Autism and Child Psychopathology Series
Series Editor: Johnny L. Matson

Johnny L. Matson *Editor*

Handbook of Intellectual Disabilities

Integrating Theory, Research, and Practice

Springer

Autism and Child Psychopathology Series

Series Editor

Johnny L. Matson
Department of Psychology
Louisiana State University
Baton Rouge, LA, USA

More information about this series at http://www.springer.com/series/8665

Johnny L. Matson
Editor

Handbook of Intellectual Disabilities

Integrating Theory, Research, and Practice

Springer

Editor
Johnny L. Matson
Department of Psychology
Louisiana State University
Baton Rouge, LA, USA

ISSN 2192-922X ISSN 2192-9238 (electronic)
Autism and Child Psychopathology Series
ISBN 978-3-030-20842-4 ISBN 978-3-030-20843-1 (eBook)
https://doi.org/10.1007/978-3-030-20843-1

© Springer Nature Switzerland AG 2019, Corrected Publication 2020
This work is subject to copyright. All rights are reserved by the Publisher, whether the whole or part of the material is concerned, specifically the rights of translation, reprinting, reuse of illustrations, recitation, broadcasting, reproduction on microfilms or in any other physical way, and transmission or information storage and retrieval, electronic adaptation, computer software, or by similar or dissimilar methodology now known or hereafter developed.
The use of general descriptive names, registered names, trademarks, service marks, etc. in this publication does not imply, even in the absence of a specific statement, that such names are exempt from the relevant protective laws and regulations and therefore free for general use.
The publisher, the authors, and the editors are safe to assume that the advice and information in this book are believed to be true and accurate at the date of publication. Neither the publisher nor the authors or the editors give a warranty, express or implied, with respect to the material contained herein or for any errors or omissions that may have been made. The publisher remains neutral with regard to jurisdictional claims in published maps and institutional affiliations.

This Springer imprint is published by the registered company Springer Nature Switzerland AG.
The registered company address is: Gewerbestrasse 11, 6330 Cham, Switzerland

Contents

Part I Foundations

Part II Philosophy of Care

Part III Parent/Professional Issues

Part IV Client Protections and Innovations

Part V Causes and Risk Factors of Intellectual Disabilities

Part VI Assessment

Part VII General Issues in Treatment

Part VIII Treatment of Problem Behaviors

Part IX Skill Building and Intellectual Disabilities

Part X Treatment for Dual Diagnosis

Part XI Health and Social Behaviors

Contributors

Wendy Aertssen Avans+, University for Professionals, Breda, The Netherlands

Jessica Akers Department of Educational Psychology, Baylor University, Waco, TX, USA

Angeleque Akin-Little Akin-Little & Little Behavioral Psychology Consultants, Malone, NY, USA

Renae Beaumont Weill Cornell Medicine, Child and Adolescent Psychiatry, New York, NY, USA

David C. Bellinger Boston Children's Hospital, Harvard Medical School, Harvard T.H. Chan School of Public Health, Boston, MA, USA

Megan O. Bird Department of Psychology, Idaho State University, Pocatello, ID, USA

Anne M. Bowers National Council of State Boards of Nursing, Chicago, IL, USA

Megan Braconnier Department of Psychology, St. John's University, Queens, NY, USA

Mary Beth Bruder University of Connecticut, Storrs, CT, USA

Larry Burd North Dakota Fetal Alcohol Syndrome Center, University of North Dakota School of Medicine and Health Sciences, Grand Forks, ND, USA

Michael Burd Department of Pediatrics, University of North Dakota School of Medicine and Health Sciences, Grand Forks, ND, USA

Claire O. Burns Department of Psychology, Louisiana State University, Baton Rouge, LA, USA

Nathan A. Call Department of Pediatrics, Division of Autism and Related Disorders, Emory University School of Medicine, Atlanta, GA, USA
Marcus Autism Center, Atlanta, GA, USA

Eileen Carey Department of Nursing and Midwifery, University of Limerick, Castletroy, Limerick, Ireland

Sarah Carlon Macquarie University, Sydney, NSW, Australia

Rachael Carroll IDS TILDA and School of Nursing and Midwifery, Trinity College Dublin, The University of Dublin, Dublin, Ireland

Mark Carter Macquarie University, Sydney, NSW, Australia

Peter Castagna Department of Psychology, Louisiana State University, Baton Rouge, LA, USA

Paige Cervantes Autism Spectrum Disorder Research and Clinical Program, Child Study Center at Hassenfeld Children's Hospital at NYU Langone, Department of Child and Adolescent Psychiatry, New York, NY, USA

Lauren Charlot Center for S.T.A.R.T. Services, University of New Hampshire, Institute on Disabilities, Concord, NH, USA

David Charnock Faculty of Medicine and Health Sciences, Queens Medical Centre, The University of Nottingham, Nottingham, UK

Hsu-Min Chiang Faculty of Education, University of Macau, Taipa, Macau, China

Emma A. Climie School & Applied Child Psychology, Werklund School of Education, University of Calgary, Calgary, AB, Canada

Tai A. Collins School of Human Sciences, University of Cincinnati, Cincinnati, OH, USA

Kaytlin Constantin Department of Psychology, University of Guelph, Guelph, ON, Canada

Brittany Cook Department of Psychology, University of Manitoba, Winnipeg, MB, Canada

Tonya Davis Department of Educational Psychology, Baylor University, Waco, TX, USA

Thompson E. Davis III Department of Psychology, Louisiana State University, Baton Rouge, LA, USA

Shoumitro Deb Division of Brain Sciences, Department of Medicine, Imperial College London, Centre for Psychiatry, London, UK

Gerda M. de Kuijper Centre for Intellectual Disabilities & Mental Health/GGZ Drenthe Research, Assen, The Netherlands

Nicole M. DeRosa Department of Pediatrics, SUNY Upstate Medical University, Syracuse, NY, USA

Meghan A. Deshais University of Florida, Gainesville, FL, USA

Robert Didden Radboud University Nijmegen, Behavioural Science Institute, Nijmegen, The Netherlands

Trajectum, Zwolle, The Netherlands

Mirjana Djordjevic Faculty of Special Education and Rehabilitation, University of Belgrade, Belgrade, Serbia

Joseph E. Donnelly Center for Physical Activity and Weight Management, University of Kansas, Lawrence, KS, USA

Lauren Donnelly Autism Spectrum Disorder Research and Clinical Program, Child Study Center at Hassenfeld Children's Hospital at NYU Langone, Department of Child and Adolescent Psychiatry, New York, NY, USA

Pamela Dunne Daughters of Charity Disability Support Service, Dublin, Ireland

Jessica Duris Department of Psychology, University of Manitoba, Winnipeg, MB, Canada

Graham M. L. Eglit Department of Psychiatry, University of California San Diego, La Jolla, CA, USA

Sam and Rose Stein Institute for Research on Aging, University of California San Diego, La Jolla, CA, USA

Patricio Erhard Department of Special Education, Clinic for Autism Research Evaluation and Support, Texas State University, San Marcos, TX, USA

Matthew J. Euler Department of Psychology, University of Utah, Salt Lake City, UT, USA

Vanessa G. Felix Department of Computer Sciences, Universidad Politecnica de Sinaloa, Mazatlan, Mexico

Cristina Fernández Conde School & Applied Child Psychology, Werklund School of Education, University of Calgary, Calgary, AB, Canada

Ralph P. Ferretti School of Education & Department of Psychological and Brain Sciences, University of Delaware, Newark, DE, USA

Laura Flanigan School & Applied Child Psychology, Werklund School of Education, University of Calgary, Calgary, AB, Canada

Jill C. Fodstad Department of Psychiatry, Indiana University School of Medicine, Indianapolis, IN, USA

Patsie Frawley Deakin University, Geelong Waterfront Campus, Locked Bag 20001, Geelong, VIC, Australia

Natalia Freitas Rossi Speech, Language and Hearing Sciences, Sao Paulo State University, (UNESP), Marilia, Sao Paulo, Brazil

Mitch Fryling California State University, Los Angeles, CA, USA

Abigail Gallivan Forward Thinking Birmingham, Finch Road Primary Care Centre, Lozells, Birmingham, UK

Amy S. Gaumer Erickson Research Collaboration, University of Kansas, Lawrence, KS, USA

Susana Gavidia-Payne RMIT University, Melbourne, VIC, Australia

Lara M. Genik Department of Psychology, University of Guelph, Guelph, ON, Canada

Kristina R. Gerencser Department of Pediatrics, Division of Autism and Related Disorders, Emory University School of Medicine, Atlanta, GA, USA
Marcus Autism Center, Atlanta, GA, USA

Stephanie Gerow Department of Educational Psychology, Baylor University, Waco, TX, USA

Reint Geuze Developmental and Clinical Neuropsychology, University of Groningen, Groningen, The Netherlands

Célia Maria Giacheti Speech, Language and Hearing Sciences, Sao Paulo State University, (UNESP), Marilia, Sao Paulo, Brazil

Shawn P. Gilroy Irish Centre for Autism and Neurodevelopmental Research, School of Psychology, National University of Ireland, Galway, Ireland

Margaret Gopaul Liberty University, Lynchburg, VA, USA

Christina Gray School & Applied Child Psychology, Werklund School of Education, University of Calgary, Calgary, AB, Canada

Joanna Greer Department of Psychology, Northumbria University, Newcastle upon Tyne, UK

Jerrica Guidry Department of Psychology, Louisiana State University, Baton Rouge, LA, USA

Michael J. Guralnick University of Washington, Seattle, WA, USA

Elizabeth Halstead Department of Psychology and Human Development, Institute of Education, University Central London, London, UK

Angela Hassiotis UCL Division of Psychiatry, Maple House, London, UK
Camden & Islington Foundation Trust, London, UK

Renee O. Hawkins School Psychology Program, sCECH | School of Human Services, University of Cincinnati, Cincinnati, OH, USA

Jenna M. Hennessey Department of Educational and School Psychology, Indiana University of Pennsylvania, Indiana, PA, USA

Miriam Heyman Ruderman Family Foundation, Boston, MA, USA

Abby Hodges Department of Educational Psychology, Baylor University, Waco, TX, USA

Esther Hong Department of Psychology, Louisiana State University, Baton Rouge, LA, USA

Abigail Issarraras Department of Psychology, Louisiana State University, Baton Rouge, LA, USA

Mervyn Jackson RMIT University, Melbourne, VIC, Australia

Holly E. Jacobs Center for Social Development and Education, University of Massachusetts Boston, Boston, MA, USA

Melissa Jeffay Department of Psychology, St. John's University, Queens, NY, USA

Callista K. Kahonde Stellenbosch University, Cape Town, South Africa

Kalli Kemp Educational Consultant, New York, New York, USA

Jacob Kerbeshian Psychiatrist, Prairie Care Medical Group, Brooklyn Park, MN, USA

Bernadette Kirkpatrick Irish Centre for Autism and Neurodevelopmental Research, School of Psychology, National University of Ireland, Galway, Ireland

Lori Klein West Chester University, West Chester, PA, USA

Marilyn G. Klug Department of Population Health, University of North Dakota School of Medicine and Health Sciences, Grand Forks, ND, USA

Russell Lang Department of Special Education, Clinic for Autism Research Evaluation and Support, Texas State University, San Marcos, TX, USA

Geraldine Leader Irish Centre for Autism and Neurodevelopmental Research, School of Psychology, National University of Ireland, Galway, Ireland

Katherine Ledbetter-Cho Department of Special Education, Clinic for Autism Research Evaluation and Support, Texas State University, San Marcos, TX, USA

The University of Texas at Austin, Austin, TX, USA

Michelle S. Lemay Department of Psychology, Idaho State University, Pocatello, ID, USA

Amy Lemelman Weill Cornell Medicine, Center for Autism and the Developing Brain, New York, NY, USA

Steven G. Little Walden University, Minneapolis, MN, USA

Julia Louw Irish Centre for Autism and Neurodevelopmental Research, School of Psychology, National University of Ireland, Galway, Ireland

Gladys E. Maestre Department of Neurosciences and Human Genetics, University of Texas Rio Grande Valley School of Medicine, Brownsville, TX, USA

Department of Human Genetics, University of Texas Rio Grande Valley School of Medicine, Brownsville, TX, USA

Arlene Mannion Irish Centre for Autism and Neurodevelopmental Research, School of Psychology, National University of Ireland, Galway, Ireland

Toby L. Martin Department of Psychology, University of Manitoba, Winnipeg, MB, Canada

St. Amant Research Centre, Winnipeg, MB, Canada

Maya Matheis MIND Institute, University of California Davis, Sacramento, CA, USA

Johnny L. Matson Department of Psychology, Louisiana State University, Baton Rouge, LA, USA

Trevor G. Mazzucchelli Child and Family Research Group, and Brain, Behaviour and Mental Health Research Group, Curtin University, Perth, WA, Australia

Parenting and Family Support Centre, School of Psychology, The University of Queensland, Brisbane, QLD, Australia

Philip McCallion School of Social Work, Temple University, Philadelphia, PA, USA

Mary McCarron Trinity Centre for Ageing and Intellectual Disability, Trinity College Dublin, The University of Dublin, Dublin, Ireland

Lindsay McCombe St. Amant Research Centre, Winnipeg, MB, Canada

Eimear McGlinchey Trinity Centre for Ageing and Intellectual Disability, Trinity College Dublin, The University of Dublin, Dublin, Ireland

Mark R. McGowan Department of Educational and School Psychology, Indiana University of Pennsylvania, Indiana, PA, USA

Catherine L. McHugh Brock University, St. Catharines, ON, Canada

Judith McKenzie Disability Studies Division, Department of Health and Rehabilitation Sciences, University of Cape Town, Cape Town, South Africa

Ty L. McKinney Department of Psychology, University of Utah, Salt Lake City, UT, USA

Laurie McLay The University of Canterbury, Christchurch, New Zealand

C. Meghan McMurtry Department of Psychology, University of Guelph, Guelph, ON, Canada

Pediatric Chronic Pain Program, McMaster Children's Hospital, Hamilton, ON, Canada

Department of Paediatrics, Western University, London, ON, Canada

Haris Memisevic Faculty of Educational Sciences, University of Sarajevo, Sarajevo, Bosnia and Herzegovina

Luis J. Mena Department of Computer Sciences, Universidad Politecnica de Sinaloa, Mazatlan, Mexico

Dunia Montalvo-Pérez Hermanas Hospitalarias-Benito Menni CASM, Service for People with Intellectual Disability, Barcelona, Spain

Lauren J. Moskowitz Department of Psychology, St. John's University, Queens, NY, USA

Colin Muething Department of Pediatrics, Division of Autism and Related Disorders, Emory University School of Medicine, Atlanta, GA, USA

Marcus Autism Center, Atlanta, GA, USA

Niamh Mulryan Daughters of Charity Disability Support Service, Dublin, Ireland

J. Meredith Murphy School of Human Sciences, University of Cincinnati, Cincinnati, OH, USA

Niri Naidoo Faculty of Health Sciences, Department of Health and Rehabilitation Sciences, University of Cape Town, Cape Town, South Africa

Karen L. Nankervis University of Queensland, St Lucia, QLD, Australia

Tom Nicholson Massey University, Auckland, New Zealand

Patricia M. Noonan Research Collaboration, University of Kansas, Lawrence, KS, USA

Matthew O'Brien University of Iowa Center for Disabilities and Development, University of Iowa Stead Family Children's Hospital, Iowa City, IA, USA

Amie O'Shea Deakin University, Geelong Waterfront Campus, Locked Bag 20001, Geelong, VIC, Australia

Rodolfo Ostos Department of Computer Sciences, Universidad Politecnica de Sinaloa, Mazatlan, Mexico

Eliana M. Pizarro University of Florida, Gainesville, FL, USA

Svetlana Popova Institute for Mental Health Policy Research, University of Toronto, Toronto, ON, Canada

Emily Porter Irish Centre for Autism and Neurodevelopmental Research, School of Psychology, National University of Ireland, Galway, Ireland

Lauren T. Ptomey Center for Physical Activity and Weight Management, University of Kansas, Lawrence, KS, USA

Evelyn Reilly Daughters of Charity Disability Support Service, Dublin, Ireland

Robert D. Rieske Department of Psychology, Idaho State University, Pocatello, ID, USA

Arne Risselada Department of Clinical Pharmacy, Wilhelmina Hospital, Assen, The Netherlands

Henry S. Roane Department of Pediatrics, SUNY Upstate Medical University, Syracuse, NY, USA

John Rose Centre for Applied Psychology, School of Psychology, The University of Birmingham, Edgbaston, Birmingham, UK

Emily A. Roth Indiana University School of Medicine, Indianapolis, IN, USA

Ruth Ryan Department of Nursing and Midwifery, University of Limerick, Castletroy, Limerick, Ireland

Matthew R. Sanders Parenting and Family Support Centre, School of Psychology, The University of Queensland, Brisbane, QLD, Australia

Shivali N. Sarawgi Indiana University School of Medicine, Indianapolis, IN, USA

Muriel D. Saunders Schiefelbush Institute for Life Span Studies, University of Kansas, Lawrence, KS, USA

Richard R. Saunders Schiefelbush Institute for Life Span Studies, University of Kansas, Lawrence, KS, USA

Dilana Schaafsma Fontys University of Applied Sciences, Eindhoven, AH, The Netherlands

Mindy Scheithauer Department of Pediatrics, Division of Autism and Related Disorders, Emory University School of Medicine, Atlanta, GA, USA

Marcus Autism Center, Atlanta, GA, USA

Jennifer Schild Weill Cornell Medicine, Child and Adolescent Psychiatry, New York, NY, USA

Georgia Shaheen Department of Psychology, Louisiana State University, Baton Rouge, LA, USA

Rebecca Shalev Autism Spectrum Disorder Research and Clinical Program, Child Study Center at Hassenfeld Children's Hospital at NYU Langone, Department of Child and Adolescent Psychiatry, New York, NY, USA

Rory Sheehan UCL Division of Psychiatry, Maple House, London, UK

Gary N. Siperstein Center for Social Development and Education, University of Massachusetts Boston, Boston, MA, USA

Bouwien Smits-Engelsman Faculty of Health Sciences, Department of Health and Rehabilitation Sciences, University of Cape Town, Cape Town, South Africa

Justin Stanley Department of Psychology, Rutgers University, New Jersey, USA

Jennifer Stephenson Macquarie University, Sydney, NSW, Australia

Karolina Štětinová Department of Psychology, Idaho State University, Pocatello, ID, USA

Regina M. Subach West Chester University, West Chester, PA, USA

William E. Sullivan Department of Pediatrics, SUNY Upstate Medical University, Syracuse, NY, USA

Frank J. Symons Department of Educational Psychology, University of Minnesota, Minneapolis, MN, USA

Karen Tang Weill Cornell Medicine, Center for Autism and the Developing Brain, New York, NY, USA

Vanessa Thompson NSW Sexology Services, Newcastle, NSW, Australia

Bruce A. Thyer College of Social Work, Florida State University, Tallahassee, FL, USA

Henrietta T. Trip Centre for Postgraduate Nursing Studies, University of Otago Christchurch, Christchurch, Canterbury, New Zealand

Gemma L. Unwin Centre for Applied Psychology, School of Psychology, The University of Birmingham, Edgbaston, Birmingham, UK

Shelley R. Upton Department of Psychology, Louisiana State University, Baton Rouge, LA, USA

Joanne E. L. VanDerNagel Tactus Addition Institute, Deventer, The Netherlands

Radboud University Nijmegen, Nijmegen Institute for Scientist-Practitioners in Addiction, Nijmegen, The Netherlands

Aveleijn, Borne, The Netherlands

Rianne van Dijken Mooi mijn lijf, Intellectual Disability Health Care & Sexology Practice, Assen, The Netherlands

Neomi van Duijvenbode Tactus Addition Institute, Deventer, The Netherlands

Radboud University Nijmegen, Nijmegen Institute for Scientist-Practitioners in Addiction, Nijmegen, The Netherlands

Radboud University Nijmegen, Behavioural Science Institute, Nijmegen, The Netherlands

Anna Costa-Vargas Department of Psychology, Universitat Autònoma de Barcelona, Barcelona, Spain

Carmen Vargas-Vargas Hermanas Hospitalarias-Benito Menni CASM, Service for People with Intellectual Disability, Barcelona, Spain

Maria V. Vassos University of Queensland, St Lucia, QLD, Australia

Endeavour Foundation, Cannon Hill, QLD, Australia

Timothy R. Vollmer University of Florida, Gainesville, FL, USA

Regan Weston Department of Educational Psychology, Baylor University, Waco, TX, USA

Keeley White Irish Centre for Autism and Neurodevelopmental Research, School of Psychology, National University of Ireland, Galway, Ireland

Lisa C. Whitehead School of Nursing and Midwifery, Edith Cowan University, Joondalup, WA, Australia

Mac Kenzie Wicker Department of Special Education, Clinic for Autism Research Evaluation and Support, Texas State University, San Marcos, TX, USA

Lienke Wilker Child and Family Research Group, and Brain, Behaviour and Mental Health Research Group, Curtin University, Perth, WA, Australia

Nathan J. Wilson School of Nursing and Midwifery, Western Sydney University, Penrith, NSW, Australia

Aline Zaragoza California State University, Los Angeles, CA, USA

Kimberley L. M. Zonneveld Brock University, St. Catharines, ON, Canada

About the Editor

Johnny L. Matson, Ph.D. is Professor and Distinguished Research Master in the Department of Psychology at Louisiana State University, Baton Rouge, LA, USA. He has also previously held a professorship in psychiatry and clinical psychology at the University of Pittsburgh. He is the author of more than 800 publications including 41 books. He served as the Founding Editor-in-Chief for the journals *Research in Developmental Disabilities* (Elsevier) and *Research in Autism Spectrum Disorders* (Elsevier) and currently serves as the Editor-in-Chief for the *Review Journal of Autism and Developmental Disorders* (Springer).

Part I

Foundations

History of Intellectual Disabilities

1

Emily A. Roth, Shivali N. Sarawgi,
and Jill C. Fodstad

Introduction

The field of intellectual disability (ID) traces its history back to early Egyptian, Greek, and Roman societies. Civilizations around the world interpret mental illness, specifically ID, in a variety of ways. Beliefs regarding ID are influenced by the current stigmas, society's understanding of mental health, and the conceptualization of the cause of ID. Furthermore, the terminology used to identify individuals with ID has paralleled the societal climate of the era. To understand the current literature and perceptions of ID, it is important to first review the history of conceptualization, terminology, research, treatment, legislation, and beliefs in this area.

On the following pages, we provide a timeline to help outline the following information presented in this chapter; however, this highlights the most important aspects of history and does not exhaust ID in its entirety. As societies across history attempted to conceptualize ID, progress toward modern thinking has been nonlinear and often includes periods of backward regression due to the religious, political, or social beliefs of the time. Advocacy groups helped propel this movement forward as they disseminated information on ID and advocated for the rights of individuals with disabilities (Fig. 1.1).

Ancient Perceptions

For much of history, ID was undifferentiated from mental illness—both are poorly understood constructs and often the target of stereotypes and stigma. As such, individuals with ID often suffered from societal injustices due to beliefs that symptoms were a by-product of a number of causes, including the individual or their families being cursed or punished for wrongful acts. The earliest definition and descriptions of ID can be found in writings as far back as ancient Egypt (Scheerenberger, 1983). During this era, ancient Egyptians were focused on developing religious and medical procedures to treat a variety of disabilities and problems (Richards, Brady, & Taylor, 2014). However, in ancient Greek and Roman civilizations, the community negatively viewed all disabilities; specifically, Sparta is infamous for killing those with disabilities (Richards et al., 2014). Ancient Greek philosophers, such as Plato and Aristotle, wrote negatively about individuals with disabilities. More specifically, Plato suggested selective mating, a foreshadowing of

---

E. A. Roth · S. N. Sarawgi
Indiana University School of Medicine,
Indianapolis, IN, USA

J. C. Fodstad (✉)
Department of Psychiatry, Indiana University School
of Medicine, Indianapolis, IN, USA
e-mail: jfodstad@iupui.ed

J. L. Matson (ed.), *Handbook of Intellectual Disabilities*, Autism and Child Psychopathology
Series, https://doi.org/10.1007/978-3-030-20843-1_1

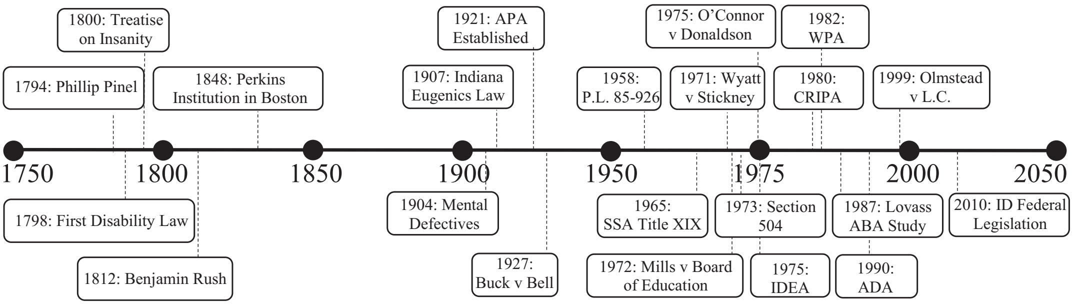


Fig. 1.1 Intellectual disability timeline. This figure illustrates key dates in the history of intellectual disabilities

the eugenics movement (Galton, 1874, 1904; Gillham, 2001a; Richards et al., 2014; Scheerenberger, 1983). Although early Romans pardoned the lives of infants born with disabilities; later Romans returned toward the tendency of infanticide for individuals with disabilities (Richards et al., 2014).

ID during the Middle Ages revolved around religion and superstitions, but Covey (1998) argued that societal response to disabilities varied by the specific disability, its context, and beliefs on etiology. Despite influences of the early Church in eliminating infanticide by providing institutions for individuals unwanted by their families due to disabilities, some argue that Luther's descriptions of the "feebleminded" were excessively negative (Harris, 2006; Richards et al., 2014). However, others argue that Luther's definition of "feebleminded" encompasses spiritual content rather than ID (Andrews, 1998). Nonetheless, during the Restoration period, "idiocy" and general mental illness were associated with immorality resulting in moral punishment (Andrews, 1998).

Philosophy's Influence

History is fraught with many philosophical descriptions and explanations for ID; however, it is important to understand the sociocultural, intellectual, and philosophical foundational contexts did not conceptualize ID as it is now defined (Groce & Scheer, 1990). Historical documentation includes words and phrases asserted to be historical terms for ID (e.g., "fool," "stupidity," and "idiot"), which refer to individuals with a variety of presentations, including mental illness and physical impairments, as opposed to specifically ID (Goodey, 2004). In the late seventeenth century, John Locke was one of the first individuals to differentiate intellectual and mental disabilities from physical disabilities. However, Locke's idea of madness and mental disability truly integrates concepts of intellectual and affective deficits and disorders (Charland, 2010). Furthermore, earlier Renaissance physicians, Paracelsus and Platter, suggested a differentiation between "foolish[ness]," "feebleminded[ness]," and mental illness (Andrews, 1998; Goodey, 2004). In 1664, Willis, another Oxford philosopher and pioneer of brain anatomy, further contributed to the understanding of "stupiditas" and idiocy by describing these states as disease-like and pinpointing various etiologies including heredity, trauma, other diseases, and "animal spirits" (Andrews, 1998; Goodey, 2004).

Asylums

During the seventeenth and eighteenth centuries, despite advances in medical and philosophical understanding (precursors to psychological understanding) of ID, affected individuals still experienced stigma and poor quality of care. They were institutionalized in asylums, hospitals, and prisons during this Age of Enlightenment and Reason. Poor living conditions, neglect, and various forms of abuse were the norm for those who lived in institutions. Furthermore, many believed that basic medical care was not a necessary privilege for individuals with ID. For example, Bethlem Hospital in England rejected individuals deemed "idiots," as they were viewed as outside of the hospital's treatment competence (Andrews, 1998). Toward the end of the seventeenth century, the beliefs of psychiatrist, Philippe Pinel, initiated a paradigm shift in the level and quality of care for individuals with ID and mental illness. Specifically, Pinel advocated for moral treatment of individuals with ID and their release from hospitals and prisons. He was and continues to be renowned for freeing patients from insane asylums in La Bicetre and later La Salpetriere.

By 1794, Pinel published an essay, "Memoir on Madness" that encouraged the study of psychopathology, and by 1798, he published the first edition of his classification of mental disorders (*Nosographie philosophique ou methode de l'analyse appliquee a la medecine*), which served as the first classification of its kind. The *Nosographie* notably differentiated between five disorders, separating ID ("ideotism or obliteration of the intellectual faculties and affections") from dementia, mania, mania without delirium, and melancholia (Pinel, 1806). Pinel's definition of idiotism is not equivalent to modern conceptions of ID. Pinel's student, Jean Marc Gaspard Itard, altered society's perception of how ID was viewed and furthered the potential for treatment and education through an emphasis on humane treatment. The combination of Pinel's work and William Tuke's establishment of humane treatment for patients with mental illness through the founding of the York Retreat for the mentally ill in 1792 served as the foundation of asylums emphasizing moral and humane treatment for patients. Additionally, these institutions began appearing in the early nineteenth century as places of refuge (Dunkel, 1983).

Pioneering the United States

As Pinel and Itard were working in France, a similar movement began in the United States started by Benjamin Rush, a founding father of the country and often considered the father of American psychiatry. Rush also advocated for more humane treatment of mental illness leading to the creation of a separate ward for such patients. In 1812, Rush published *Medical Inquiries and Observations, Upon the Diseases of the Mind*, which served as the first attempt to explain and treat disorders of the mind in the United States. Early nineteenth-century Americans witness the paradigm shift in increased attention to the ideas of mental disabilities and ID. However, at the time, the differentiation of mental disability and ID was not complete; although philosophers attempted to describe it in the past. In the 1930s, James Prichard, another notable psychiatrist, theorized a distinction between ID and mental illness or "intellectual [versus] moral insanity," serving as the foundational definition of this distinction (Carlson & Dain, 1962). Furthermore, advances in awareness of mental illness and general psychiatry significantly relate to advances in understanding and treating ID.

In 1818, the Asylum for the Insane (later the McLean Asylum for the Insane in 1826 and then McLean Hospital in 1892) opened in Massachusetts (Little, 1972) further transforming the quality of care for individuals with ID. Like other asylums of its time, the ideas of "moral treatment" influenced by Pinel and Tuke heavily influenced the model of care for the Asylum (Dunkel, 1983; Pinel, 1806). The McLean Asylum's groundbreaking research and its transition to Belmont, Massachusetts, established the transformation from institutional settings to a more residential environment (Little, 1972). By 1848, Samuel Gridley Howe opened the Experimental School for Teaching and Training Idiotic Children, within the Perkins Institution (a residential institution for the blind), in Massachusetts, serving as the first residential facility for those with developmental disabilities. Now called the Walter E. Fernald Developmental Center, this school became a leader in providing education to individuals with ID. The Experimental School for Teaching and Training Idiotic Children helped change current beliefs to demonstrate that individuals with ID are capable of learning and need to be appropriately challenged. During the mid-nineteenth century, the

Association of Medical Superintendents of American Institutions for the Insane was established, which eventually became the American Psychiatric Association in 1921 (Hirshbein, 2004). This organization was influential in the advancement of science and understanding mental illness and ID.

Eugenics Movement

While the early nineteenth century was marked by the emphasis of humane treatment of those with mental disabilities and ID, the latter part of the century was marked by the eugenics movement, leading to a de-emphasis on ethical treatment and increased stigma against individuals with various disabilities, including ID. The movement was started by Sir Francis Galton who originated the word "eugenics" in 1883. Originally developed from Platonian thought of artificial selection, eugenics occurred as a treatment of individuals with ID and other mental disabilities in early civilizations and medieval times (Richards et al., 2014; Scheerenberger, 1983). However, Galton (1904) conceptualized eugenics as a science focusing on the "improvement" of the species, with emphasis on the influence of nature versus nurture (Galton, 1874). Galton and eugenics were heavily influenced by Charles Darwin's publication, *On the Origin of Species*, in the mid-nineteenth century (Gillham, 2001a). In Galton's distinction between nature and nurture, he indicated that human ability was most strongly influenced by nature. Through this belief, Galton advocated for the heritability of intelligence and cognition (Galton, 1865, 1874; Gillham, 2001a).

As noted above, an era's sociocultural makeup heavily influences conceptualizations of intelligence and ability/disability. Galton's applications of natural selection through the eugenics movement were in opposition to other societal movements of his time during Victorian England (Galton & Galton, 1998). The eugenics movement developed at the same time as societal reforms in England, such as the Anti-slavery Bill, educational acts, protections for the poor, and other social reforms. Galton's work led to the creation of a eugenics register, an arbitrary scale of "worth," and classification of families as gifted, capable, average, or degenerate. However, this measure of intelligence was not available during his time nor proposed by Galton (Galton & Galton, 1998; Gillham, 2001b). Regardless of any scientific merit of Galton's work, it instigated a societal movement that holds significant impact on the treatment of individuals with disability (as well as other socially constructed groups) including sterilization, the restriction of movement, and segregation of individuals with ID.

Twentieth-Century Eugenics and Involuntary Sterilization Laws

Eugenics has existed for centuries, with many influential leaders including Charles Darwin, Cesare Lombroso, Thomas Malthus, Arthur de Gobineau, and Herbert Spencer (Gejman & Weilbaecher, 2002). The eugenics movement gained traction when elected officials focused on ensuring the quality of the population by minimizing and decreasing the number of feebleminded, insane, imbeciles, and idiots (Kendregan, 1966; Stern, 2007). Supporters of eugenics posited that affected individuals were a social burden to the country and should not procreate (Sofair & Kaldjian, 2000). Within the United States, Indiana was the first state to pass a compulsory eugenic sterilization bill in 1907. Thurman Rice piloted sterilization outside of institutions within the state of Indiana. Following Indiana's passing of this legislation, 29 states created their own sterilization laws, including 18 that involved involuntary sterilization (Kendregan, 1966; Sofair & Kaldjian, 2000). Within the first 40 years of these laws, almost 40,000 people underwent sterilization in the United States (Proctor, 1988). Over time, challenges to the constitutionality of these laws occurred. Indiana's sterilization laws were deemed unconstitutional in 1921; however, new legislation (reinstating sterilization) was approved in 1927 (Stern, 2007). The change in beliefs regarding sterilization was not limited to the state of Indiana: consecutive cases on the grounds of constitutionality of sterilization occurred across the United States (Kendregan, 1966; Sofair & Kaldjian, 2000; Stern, 2007).

The Supreme Court case, *Buck* v. *Bell* (1927), served as a monumental shift in the perception of the legality of involuntary sterilization (Berry, 1998; Kendregan, 1966; Siegel, 2005; Sofair & Kaldjian, 2000). The individual who led to *Buck* v. *Bell* (1927) was Carrie Buck, a young woman in Virginia whose mother could no longer care for her (Berry, 1998). Carrie, a teenager, became pregnant as a result of rape. Despite her admission, the courts considered Carrie, her mother, and her infant daughter to be "feebleminded," resulting in Carrie's institutionalization in 1924 when she was 18. The superintendent petitioned to the Board of Virginia Colony that Carrie be sterilized to prevent her from having more children that required further services. The board approved this motion; however, Carrie's lawyer appealed her case to the Virginia trial court, then the Virginia Supreme Court, and finally the US Supreme Court (Berry, 1998). The US Supreme Court ruled in favor of involuntary sterilization. Justice Holmes gave the US Supreme Court's majority's opinion in *Buck* v. *Bell* (1927), claiming that "It is better for all the world, if instead of waiting to execute degenerate offspring for crime, or to let them starve for their imbecility, society can prevent those who are manifestly unfit from continuing their kind" (p. 207). Prior to the court's determination, 23 states already enacted legislation related to involuntary sterilization (Laughlin, 2009); therefore *Buck* v. *Bell* (1927) confirmed these states' stance on sterilization. One of the largest counterarguments and a descending opinion to this case is that forcing an individual to be sterilized is "cruel and unusual punishment" (Siegel, 2005). In the aftermath of this case, upward of 50,000 Americans received involuntarily sterilization over the next few decades (Berry, 1998). In 1978, the Department of Health, Education, and Welfare altered the requirements for sterilization to include informed consent, therefore, abolishing involuntary sterilization (Title 42, 1978).

Earliest Textbook

In 1904, Martin Barr, MD, published the first textbook on ID titled *Mental Defectives: Their History, Treatment, and Training*. Barr (1904) provided professionals unprecedented information on understanding ID based upon the labels of the time: imbeciles, feebleminded, idiots, and mental deficient. Barr (1904) differentiates between an idiot and imbecile by explaining an idiot "see nothing, feels nothing, hears nothing, and knows nothing," whereas imbeciles have the capacity to understand and reason (p. 18). Furthermore, he stated that differentiating between these two is influential in selecting treatment protocols and assigning realistic prognosis. Through his definitions and classifications, Barr paved the way for others to appropriately differentiate causes and prognoses of those presenting as deficient in some way. Tredgold (1908) expands on Barr's concepts by creating a resource for professionals to educate on appropriate diagnosis, treatment, eligibility for services, and long-term placements. Barr (1904) and Tredgold (1908) provide descriptions of biological, physical, and cognitive differences of those with mental defects. Barr (1904) provides hope for those labeled as mentally defective prognosis by differentiating between disease and defect and argues that regression is preventable through appropriate treatment for those who fall within defect. However, through his classification of idiots and imbeciles, Barr postulates idiots are unable to make any gains and treatment will not result in any improvements; therefore, treatment resembled nurseries. However, treatment focused on those termed as imbeciles' emotions, body, and mind through training them in how to complete daily living activities through step-by-step instructions and avoiding any abstract concepts (Barr, 1904). The importance of differentiating and understanding the cause and prognosis of one's mental deficiency continued to inform ongoing research into the treatment of ID for centuries to come (Roberts, 1952; Schalock & Luckasson, 2013).

National Associations

After witnessing firsthand the inadequate services and advocacy for children with disabilities, Edgar Allen made it his life's effort to ensure the other children received proper care (Williams, 1994). In 1919, Mr. Allen founded the Ohio Society for Crippled Children as an organization advocating for the rights of disabled children. The organization began requesting donations to fund children's services and advocating for children's right to live. In 1967, the organization had slowly gained a national presence and officially changed its name to Easter Seals. As the Americans with Disabilities Act (ADA) developed, Easter Seals pioneered the advocacy movement for those with disabilities in Washington, DC. Easter Seals focuses on providing services to families within five categories: live, learn, work, play, and act. Easter Seals served as an example for other advocacy organizations within the United States and worldwide (Williams, 1994).

In 1930s, the National Association of Parents and Friends of Mentally Retarded Children began forming groups to advocate for improving their children's care (Hay, 1952). By 1951, the United States and Canada had over 125 groups with thousands of members. The growing membership resulted from two very important areas: (1) parents and the community's awareness that children were not receiving appropriate treatment and (2) the medical community simultaneously developing new treatments for conditions previously thought to be untreatable. This association, which later became known as the National Association for Retarded Children (ARC), focused on circulating accurate information on mental health and advocating for adequate treatment, education, vocational services, housing, and government policies (Hay, 1952).

Simultaneously, in 1946, the National Mental Health Foundation was founded, which was a civilian-run organization focused on the dissemination of factual information about mental health to legislators, educators, the public, and mental institutions. This foundation advocated for an increased standard of care for mental health treatment, education of the layperson on aiding those with mental health problems, promoting focus on psychiatry programs within already developed universities, and furthering current mental health professionals understanding of the illnesses ("National Mental Health," 1946).

In 1979, the National Alliance on Mental Illness (NAMI) originated as a small grassroots organization. NAMI focuses on educating, advocating, listening, and leading. NAMI develops educational programs to inform families and providers on mental illness and its personal implications. NAMI frequently spearheads public policy ensuring appropriate representation of the rights of individuals with mental illness in legislation. NAMI offers a helpline to assist in connecting families with local providers and supporting individuals with suicidal ideation. NAMI also hosts frequent events to increase the public's awareness of mental illness and work toward destigmatizing the field (NAMI, 2018).

Appropriate Training for Service Providers

President Eisenhower signed the Education of Mentally Retarded Children Act (1958), which allotted funds to provide additional training to universities on educating individuals with mental retardation. When the act, P.L. 85–926, was first signed, it allocated $11 million per year for educators and professionals to gain additional knowledge and understanding of mental retardation. Through this added knowledge, these leaders create programs within already established colleges and within local public school systems aimed at teaching and adapting curriculum for those with mental retardation (Martin, Martin, & Terman, 1996). An addendum was passed in 1963, P.L. 88–164, which expanded on the earlier act to include adaptation for other disabilities (e.g., emotional disability, deaf, crippled, speech impaired, blind, and other health impairments). From the initiation of P.L. 85–926 through the addendums, over 20,000 educators and professionals received training to adapt education to affected individuals. From the initial $one million allotment per year, this amount expanded to $55 million allotted in 1970 ("Programs for the Handicapped," 1969).

Terminology Changes Over Time

The terminology to refer to ID continues to change over time. In the early 1990s, labels for individuals with ID were imbecile, feeble-minded, idiots, and mental deficient in the early 1900s (Barr, 1904). In 1952, the American Psychiatric Association (APA, 1952) published the first *Diagnostic and Statistical Manual* (DSM-I) classifying this disorder as "chronic brain syndrome with mental deficiency" (p. 14) and "mental deficiency" (p. 23). In 1959, the American Association of Mental Deficiency introduced the term "mental retardation" and changed their name to the American Association of Mental Retardation (AAMR) (Fredericks & Williams, 1998). In the 1960s, APA (1968) published their second edition of the DSM-II, which incorporated the new diagnosis of "mental retardation." It was not until the publication of DSM-III in 1980 that APA (1980) addressed that mental retardation does not solely include a deficit in intellectual functioning but also impairment in adaptive skills. From 1980 through the early 2000s, this remained the definition and conceptualization for mental retardation. In 2010, the World Health Organization and the AAMR, now known as the American Association on Intellectual and Developmental Disabilities (AAIDD), formed work groups to prepare for proposed changes in the International Classification of Diseases (ICD-11). In 2010, the ICD-11 implemented the change in terminology to "intellectual disability" following President Obama signing "Rosa's Law" which changed "mental retardation" to "intellectual disability" in federal education laws (Tassé, Luckasson, & Nygren, 2013). Shortly after, DSM-5 was released in 2013 and agreed and incorporated the diagnosis "intellectual disability" (APA, 2013).

Deinstitutionalization

Many individuals with mental health diagnoses lived in state hospitals, which reached its peak census in 1955 with over 550,000 individuals hospitalized (Bachrach & Lamb, 1989). Over the next decade, the deinstitutionalization movement progressed, with the federal government's formalization of deinstitutionalization beginning in 1965 as the civil rights movement gained traction and advocates fought for the least restrictive environment (Chow & Priebe, 2013). This transition did not occur over night, and it took decades to fully transition individuals from state hospitals to outpatient community mental health centers ("Deinstitutionalization Movement," 2007). To successfully deinstitutionalize and rehabilitate, society needs proper funding to create treatment centers and transitional housing. Additionally, for this process to be effective, individuals need to be compliant with treatment recommendations and have the financial and social supports to meet these goals (Bachrach & Lamb, 1989). Unfortunately, due to limited rehabilitation services in America, many affected individuals become homeless.

Federally Funded Programs

In 1965, the federal government passed Title XIX within the Social Security Act (1965), which instated the federal- and state-level Medicaid programs (Klees, Wolfe, & Cutris, 2009). Title XIX identified individuals with low income, children, pregnant women, blind individuals, and permanently disabled individuals. An individual was considered disabled if their physical or mental diagnosis prevents them from being able to work. This was the first legislation of its type to recognize that individuals with disabilities need additional funding for appropriate services. The Medicaid plan covers a variety of treatments, including inpatient-, outpatient-, residential-, and community-based centers (Social Security Act, 1965).

Special Olympics

Through the late 1950s and early 1960s, Eunice Kennedy Shriver became abundantly aware of the injustices faced by individuals with ID; therefore, she made it her personal mission to create an atmosphere conducive to individuals with ID to thrive and enjoy life (Special Olympics, 2018). In 1962, Ms. Shriver held her first summer camp in Washington, DC, which included sporting events and outdoor activities. In 1967, Chicago proposed an idea to replicate the Olympics track meet for those with ID. The following summer of 1968, Ms. Shriver held the first International Special Olympics Summer Games in Chicago, IL, leading to the formal incorporation of Special Olympics as an organization that fall. Throughout the 1970s, the Special Olympics continued to grow within the United States, and by 1977, the winter Olympics developed. At the 1987 International Special Olympics held in Indiana, athletes from 70 countries competed (Special Olympics, 2018). The Special Olympics continues to be a remarkable organization promoting the enjoyment of life for those with disabilities and advocates for individual's rights in the progression of disability legislation.

Disability Legislation and Right to Treatment

The first legislative recognition in favor of increased funding for individuals with ID came in 1970 with the Developmental Disabilities Services and Facilities Construction Amendments (1970). This act provided states with funding for treating, giving services, and creating new treatment facilities for those with ID. The Developmentally Disabled Assistance and Bill of Rights Act (1975) served as an amendment to the earlier policy and included the advocacy of individuals with developmental disabilities, early screening, and the protection of their rights. The "rights of the developmentally disabled" were described as including individuals' right to treatment, services emphasizing the individual's potential, and appropriate funding for this treatment. An additional amendment, the Developmental Disabilities Act of 1984, addressed treatment goal of establishing services to help the individual with gaining independence, assimilation into the community, and supporting individuals to become employed. This Act and its amendments represent the shift within society in understanding the importance of quality of life of individuals with disabilities and advocating for their right to achieve this.

While legislation occurred on a federal level in the 1970s, states were also similarly grappling with legislation to improve care for individuals with ID. For example, in the state of Alabama, quality of patient care and access to safe-staffing models at Bryce State Hospital became the cornerstone of *Wyatt* v. *Stickney* (1971). The state of Alabama increased taxes on cigarettes, which affected Bryce State Hospital's ability to pay their employees. Therefore, they fired around 100 employees, leaving few professionals to care for the residents and patients. The ratios for patient to provider were outrageous: 1 psychiatrist served over 1500 patients, 1 nurse cared for 250 patients, and 1 doctor tended to over 350 patients (Carr, 2004). The focus and ruling of *Wyatt* v. *Stickney* (1971) laid the foundation for future cases recognizing patients', including those in mental health facilities, right to appropriate treatment and safe environments (Perlin, 2011).

O'Connor v. *Donaldson* (1975) was the first case to reach the US Supreme Court on the basis of right to treatment and constitutionality of confinement. After receiving a diagnosis of paranoid schizophrenia in 1957, Kenneth Donaldson's father civilly committed him. Donaldson petitioned for his release, claiming that he was not receiving treatment. Donaldson asserted that it was his constitutional right to discharge himself or receive treatment (Fields, 1976; Wold, 1975). The appellate court ruled on the presumption that Donaldson's constitutional right was to receive treatment, therefore, ruling against his discharge request. In *O'Connor* v. *Donaldson* (1975), the US Supreme Court ruled in support of Donaldson, declaring that he was involuntary committed, which deprived him of his personal liberties. The court stated that the only justification for involuntary commitment is if an individual is receiving treatment or is dangerous; given that neither of these was true for Donaldson, they ruled that a violation of his constitutional rights occurred. They further indicated that an individual's mental illness, considered independently, is not enough to qualify for involuntary commitment (Fields, 1976; Wold, 1975).

In 1980, the Civil Rights of Institutional Persons Act (CRIPA) further established the rights of individuals in institutions (e.g., prisons, hospitals, mental health facilities, nursing homes). The Attorney General claimed that any setting receiving federal aid much respects individual's rights. CRIPA (1980) specifically protects individuals against sexual abuse, inappropriate mental health treatment, inefficient rehabilitation, inadequate education, and abuse or neglect. CRIPA (1980) does not establish new rights for individuals; rather CRIPA creates a system for the Department of Justice (DOJ) to monitor and prosecute violations.

In the early twenty-first century, the Developmental Disabilities Assistance and Bill of Rights Act set the stage for increasing accessibility of treatment within the field of ID. This act supports ongoing research of developmental disabilities and ID to ensure federal policy conforms to updated research. This act also advocates for the individual's rights and protects them from unlawful abuse. The Developmental Disabilities Assistance and Bill of Rights Act of 2000 created numerous programs and legal counsel to represent those with intellectual and developmental disabilities in policy. There are state counsels which represent their rights and ensure that updated policies adhere to disability laws and remove individuals' barriers to treatment and participating within society. Protection and advocacy groups also appeared to advocate and empower individuals to represent themselves. Also created were University Centers for Excellence in Developmental Disabilities Education, Research, and Service (UCEDDS) to ensure educational organizations represent their rights and provide appropriate accommodations (Developmental Disabilities Assistance and Bill of Rights Act, 2000).

Educational Rights for the Disabled

A class action lawsuit, *Pennsylvania Association for Retarded Citizens (PARC)* v. *Commonwealth of Pennsylvania* (1972), filed petitions against the board of education and specific school districts. This was the foundational case in establishing students' right to education. *PARC* v. *Pennsylvania* (1972) claimed that students with mental retardation did not receive the same, if any, education as their peers. This case argued that the 14th Amendment of the US Constitution specifies that legislation cannot impede on citizen's rights (e.g., right to education). Therefore, school systems were violating this right by denying students with mental retardation the right to their education. The ruling on this case asserted that law requires school districts to offer public education to all children, regardless of mental abilities. Shortly after this case, a similar class action suit, *Mills* v. *Board of Education* (1972), was brought against Washington, DC. Schools excluded upwards of 18,000 children with mental and physical disabilities from receiving public education. The ruling of *Mills* v. *Board of Education* (1972) emphasized students' right to education and required schools to redistribute their financial allocations to provide adequate funding for students with mental and physical disabilities (Yell, Rogers, & Rogers, 1998).

Both cases laid the foundation for federal legislation, Section 504 of the Rehabilitation Act of 1973. This act asserts that discrimination against individuals based on their disability, including mental health diagnoses, is unlawful. This legislation particularly applies to any organization that receives federal funding, which includes educational institutions (Yell et al., 1998). The Education for all Handicapped Children Act of 1975 (EAHCA) offered states federal funding for special education services. As previously discussed, states must offer free education; therefore, EAHCA serves as an incentive and support for states to provide adequate education. EAHCA (1975) posits that students have the following rights: modified education, free education, least restrictive environment, nondiscrimination, and due process. EAHCA also incorporated

Individualized Education Program (IEP) that tailors a student's education to their particular needs (Yell et al., 1998). In 1990, EAHCA changed its name to the Individuals with Disabilities Education Act (IDEA) that altered the wording to represent person-first language (e.g., individual with a disability). IDEA (1997) amended earlier legislation to include specialized category for autism, emphasis on parental involvement, and serving individuals from infancy through the age of 21.

National and International Legislation

In 1982, the United Nations established the World Programme of Action (WPA) Concerning Disabled Persons. The WPA addresses disability advocacy from a human rights perspective, focusing on country's legislation and policies for the equality of the individual. Given the foundation within the UN, the WPA addresses disability rights and advocacy at an international level and aims to empower individuals to become fully integrated into society as active participants. This initiative tackles countries' lack of resources and this role in the perpetuation of disability among future generations. WPA strives to prevent, rehabilitate, and create equal opportunities for those with disabilities across the world (WPA, 2018).

Established within the Department of Education in 1978, the National Council on Disability (NCD) has been monumental in federal advocacy efforts for those with ID. This council briefs the president, congress, local/state governments, and other agencies on issues affecting individuals with disabilities. NCD collects data to present to their counsel, participates in debates related to monumental issues, and prepares solutions to problems related to disabilities (NCD, 2018). NCD was instrumental in the development of the Americans with Disabilities Act (ADA), which expanded upon previous legislation impacting federally funded programs to include private businesses in being responsible for respecting the rights of those with disabilities (Waterstone, 2015). ADA (1990) protects individuals against discrimination at work, at home, in public locations, on transportation, in recreational settings, institutionalizations, and accessing public services. Prior to the passing of ADA (1990), other legislations protected individuals' civil rights, but it lacked protection based on a disability. In addition to protecting individuals with physical disabilities, ADA (1990) also protects against mental impairment.

Olmstead v. *L. C.* (1999) was instrumental in establishing mental health patients' rights to appropriate treatment without confinement and institutionalization against their will. The case involved two women, Lois Curtis and Elaine Wilson, who were both diagnosed with mild ID and treated at a hospital in Georgia. Both women voluntarily committed themselves. However, after reaching treatment goals, the hospital prohibited their discharge to a community treatment facility. Lois Curtis remained hospitalized for 3 additional years after her psychiatrist considered her fit for discharge. The premise of *Olmstead* v. *L. C.* (1999) sits upon an individual's human rights, and unlawful confinement violates these rights as stated in ADA (1990). This case escalated to the US Supreme Court after numerous appeals by the state of Georgia defending their actions of not releasing these women. The US Supreme Court agreed with the previous courts that Georgia was acting in violation of ADA (1990) and the state holds the responsibility of allocating proper funding to ensure patients have access to necessary care (e.g., state hospital versus community-based setting). This case merges previous cases both on an individual's right to treatment, disability law, and adhering to federal legislation (McCants, 1999).

Applied Behavioral Analysis

The Journal of Applied Behavioral Analysis published their first periodical in Spring 1968 and continues to publish four volumes per year (JABA, 2018). O. Ivar Lovaas, often credited with developing applied behavior analysis (ABA), proved its effectiveness with children with autism spectrum disorder (ASD) and

ID. ABA is an intensive behavioral treatment that utilizes positive reinforcement and interventions to solicit positive behaviors and discourage undesirable behaviors. Lovaas pioneered the treatment for these disorders by emphasizing the importance of early intensive intervention or ABA. Lovaas' earlier research focused on understanding antecedents and consequences and functional behavior analysis (Smith & Eikeseth, 2011). Lovaas, Koegel, Simmons, and Long (1973) published a preliminary study assessing the long-term effects of ABA, which demonstrated that if parents were involved in the treatment and trained in behavioral principles, children had longer-lasting benefits and decreases in behaviors. However, when institutionalized, the initial gains from ABA dissipated, and the child returned to baseline functioning. Lovaas (1987) published a comprehensive article discussing the effectiveness of ABA for children with ASD and ID. This study assesses long-term behavior treatment and concluded that almost half the participants reached normal cognitive functioning and at grade level school performance. Additional findings by Lovaas showed that of those 50% of students placed in special education classes, around 40% of students functioned at a delayed level, with the remaining 10% being profoundly intellectually disabled (Lovaas, 1987). McEachin, Smith, and Lovaas (1993) published a longitudinal study documenting the effectiveness of long-term early intervention behavior therapy, demonstrating that by the age of 7, participants were comparable to typically developing peers. In sum, Lovaas and his colleagues' findings emphasized the importance of beginning ABA early: children who participated in extensive treatment prior to beginning school achieved more optimal outcomes. There is extensive research on ABA and its implications within the field of ID.

Disability Culture

Over the past centuries, individual's, society's, and the international community's beliefs on disability have shifted. Chicago held the first Disability Pride Parade in the United States in 2004 (Schmich, 2017). This parade symbolized the drastic shift in perception of disability within the United States and society's move toward acceptance of differences and individuality. As technology improved, national organizations, blogs, and social media outlets now host open conversations about disability and disseminate accurate information that was previously unavailable to the general public (Brown, 2015). Now, more than ever, individuals embrace their differences and disability, and there are many public figures who openly discuss their disabilities and the challenges they face. Disability culture has boomed over the past few decades as information is readily available online (Brown, 2015).

Summary

This chapter attempts to disseminate the long, complicated history of ID while highlighting key points that were influential in the forward trajectory of the perception of mental health, specifically ID. ID's early history is traced back to ancient Roman, Egyptian, and Greek philosophers. Initially, many philosophers and scientists attempted to understand mental health and individuals who did not conform to what was typically expected. Treatment facilities ranged from asylums to institutions, to inpatient hospitals and residential treatment facilities, to now, more commonly, community mental health centers. As the understanding of mental health and ID began establishing within the United States, individuals began to fear those different than themselves, leading to the eugenics movement. Stemming from eugenics and involuntary institutionalization began the legislative history in the United States, which laid the foundation for modern-day civil rights. National organizations were established, which spearheaded the civil rights movement and fought the battle against the stigma of mental illness and ID. As the understanding of ID advanced, individuals and national organizations fought for the rights of those with ID, specifically their right to quality humane treatment and appropriate education. Numerous laws were passed protecting individuals' rights and abolishing ancient treatment methods. As individuals were educated on ID, additional trainings were created for those treating individuals with ID. Newer treatment modalities were discovered that were evidence based for ID. As the cultural perception of ID shifted, the terminology used to discuss individuals with ID adapted to be more sensitive.

References

Andrews, J. (1998). Begging the question of idiocy: The definition and socio-cultural meaning of idiocy in early modern Britain: Part 2. *History of Psychiatry, 9*(34), 179–200.

American Psychiatric Association. (1952). *Diagnostic and statistical manual: Mental disorders*. Washington, DC: APA.

American Psychiatric Association. (1968). *Diagnostic and statistical manual of mental disorders* (2nd ed.). Washington, DC: APA.

American Psychiatric Association. (1980). *Diagnostic and statistical manual of mental disorders (DSM III)* (3rd ed.). Washington, DC: APA.

American Psychiatric Association. (2013). *Diagnostic and statistical manual of mental disorders (DSM 5)* (5th ed.). Washington, DC: APA.

Americans with Disabilities Act (ADA) of 1990, 42 U.S.C.A. § 12101 *et seq*. (1990).

Bachrach, L. L., & Lamb, H. R. (1989). What have we learned from deinstitutionalization? *Psychiatric Annals, 19*(1), 12–17.

Barr M. W. (1904). Sketch of the history of the treatment of mental defect. *Charities*, 879–883.

Berry, R. M. (1998). From involuntary sterilization to genetic enhancement: The unsettled legacy of Buck v. Bell. *Notre Dame Journal of Law, Ethics & Public Policy, 12*, 401.

Brown, S. E. (2015). Disability culture and the ADA. *Disability Studies Quarterly, 35*(3). Retrieved from http://dsq-sds.org/article/view/4936

Buck v. *Bell*, 274 U.S. 200 (1927).

Carlson, E. T., & Dain, N. (1962). The meaning of moral insanity. *Bulletin of the History of Medicine, 36*(2), 130–140.

Carr, L. W. (2004). Wyatt v. Stickney: A landmark decision. *Alabama Disabilities Advocacy Program Newsletter*. Retrieved from http://neurosciencecme.com/email/2006/120406 landmark.pdf

Charland, L. C. (2010). Science and morals in the affective psychopathology of Philippe Pinel. *History of Psychiatry, 21*(1), 38–53.

Chow, W. S., & Priebe, S. (2013). Understanding psychiatric institutionalization: A conceptual review. *BMC Psychiatry, 13*, 169. https://doi.org/10.1186/1471-244X-13-169

Covey, H. C. (1998). *Social perceptions of people with disabilities in history*. Charles C Thomas Pub Ltd., Springfield, IL.

CRIPA, Public Law 96-247, The U.S. Code, Title 42: The Public Health and Welfare, Chapter 21: Civil Rights, Subchapter I-A-Institutionalized Persons, Civil Rights of Institutionalized Persons Act (CRIPA) (1980).

Deinstitutionalization movement provides lessons for reforming long-term care in the United States. (2007). *Psychiatric Services; Arlington, 58*(10), 1383.

Developmental Disabilities Assistance and Bill of Rights Act of 2000, Pub. L. No. 106–402, 114 Stat. 1677–1740 (2000).

Developmental Disabilities Services and Facilities Construction Amendments of 1970, Pub. L. No. 91–517, 84 Stat. 1316–1327 (1970).

Developmentally Disabled Assistance and Bill of Rights Act, Pub. L. No. 94–103, 89 Stat. 2662–507 (1975).

Dunkel, L. M. (1983). Moral and humane: Patients' libraries in early nineteenth-century American mental hospitals. *Bulletin of the Medical Library Association, 71*(3), 274.

Education of Mentally Retarded Children Act, Pub. L. No. 85–926, 72 Stat. 1777 (1958).

Fields, J. D. (1976). O'Connor v. Donaldson. *Hofstra Law Review, 4*(2), 10.

Fredericks, D. W., & Williams, W. L. (1998). New definition of mental retardation for the American Association of Mental Retardation. *Image The Journal of Nursing Scholarship; Indianapolis, 30*(1), 53.

Galton, F. (1865). Heredity, talent, and character. *Macmillan's Magazine, 12*, 157–166, 318–327.

Galton, F. (1874). *English men of science: Their nature and nurture*. London: Macmillan & Co.

Galton, F. (1904). Eugenics: Its definition, scope, and aims. *American Journal of Sociology, 10*(1), 1–25.

Galton, D. J., & Galton, C. J. (1998). Francis Galton: And eugenics today. *Journal of Medical Ethics, 24*(2), 99–105.

Gejman, P. V., & Weilbaecher, A. (2002). History of the eugenic movement. *The Israel Journal of Psychiatry and Related Sciences; Jerusalem, 39*(4), 217–231.

Gillham, N. W. (2001a). Sir Francis Galton and the birth of eugenics. *Annual Review of Genetics, 35*, 83–101. https://doi.org/10.1146/annurev.genet.35.102401.090055

Gillham, N. W. (2001b). *A Life of Sir Francis Galton: From African Exploration to the Birth of Eugenics*. New York: Oxford University Press.

Goodey, C. F. (2004). "Foolishness" in early modern medicine and the concept of intellectual disability. *Medical History, 48*(3), 289–310.

Groce, N. E., & Scheer, J. (1990). Introduction to special issue on anthropology and disability. *Social Science and Medicine, 30*, 5–6.

Harris, J. C. (2006). *Intellectual disability: Understanding its development, causes, classification, evaluation, and treatment*. Oxford University Press.

Hay, W. (1952). Associations of parents and friends of mentally retarded children. In *Encyclopedia Americana*. New York: Americana Corp..

Hirshbein, L. D. (2004). History, memory, and profession: A view of American psychiatry through APA presidential addresses, 1883–2003. *American Journal of Psychiatry, 161*(10), 1755–1763.

Journal Applied Behavior Analysis: Society for the Experimental Analysis of Behavior (JAMA). (2018). National Center for Biotechnology Information, U.S. National Library of Medicine. Retrieved from https://www.ncbi.nlm.nih.gov/pmc/journals/309/

Kendregan, C. P. (1966). Sixty years of compulsory eugenic sterilization: Three generations of imbeciles and the Constitution of the United States. *Chicago-Kent Law Review, 43*, 123.

Klees, B. S., Wolfe, C.J ., & Cutris, C. A. (2009). Brief summaries of Medicare and Medicaid: Title XVIII and Title XIX of the Social Security Act. Department of Health and Human Services.

Laughlin, H. H. (2009). The legal status of eugenical sterilization. *Buck v Bell Documents*. Retrieved from http://readingroom.law.gsu.edu/buckvbell/79

Little, N. F. (1972). *Early years of the Mclean Hospital*. Boston, MA: Francis A. Countway of Medicine.

Lovaas, O. I. (1987). Behavioral treatment and normal educational and intellectual functioning in young autistic children. *Journal of Consulting and Clinical Psychology, 55*(1), 3–9.

Lovaas, O. I., Koegel, R., Simmons, J. Q., & Long, J. S. (1973). Some generalization and follow-up measures on autistic children in behavior therapy. *Journal of Applied Behavior Analysis, 6*(1), 131–165. https://doi.org/10.1901/jaba.1973.6-131

Martin, E. W., Martin, R., & Terman, D. L. (1996). The legislative and litigation history of special education. *The Future of Children, 6*(1), 25–39. https://doi.org/10.2307/1602492

McCants, W. D. (1999). Disability & ADA: Supreme Court rules on institutional confinement of disabled. *The Journal of Law, Medicine & Ethics; Boston, 27*(3), 281–283.

McEachin, J. J., Smith, T., & Lovaas, O. I. (1993). Long-term outcome for children with autism who received early intensive behavioral treatment. *American Journal of Mental Retardation: AJMR, 97*(4), 359–372-391.

Mills v. *Board of Education of the District of Columbia*, 348 F. Supp. 866 (1972).

National Alliance on Mental Illness (NAMI). (2018) About NAMI. Retrieved from https://www.nami.org/about-NAMI

National Counsel on Disability. (2018). About us. Retrieved from https://www.ncd.gov/about

National Mental Health Foundation Pamphlet. (1946). *The Civilian Public Service Story*. Retrieved from http://civilianpublicservice.org/sites/default/files/sites/civilianpublic service.org/files/documents/nmhf-pamphlet-nmhf.pdf

O'Connor v. *Donaldson*, 422 U.S. 563 (1975).

Olmstead v. *L. C.*, 527 U.S. 581 (1999).

Pennsylvania Association for Retarded Citizens (PARC) v. *Commonwealth of Pennsylvania*, 343 F. Supp. 279 (1972).

Perlin, M. L. (2011). *International human rights and mental disability law: When the silenced are heard*. Oxford University Press.

Pinel, P. (1806). *A treatise on insanity*. London: Messers, Cadell, & Davies, Strand.

Proctor, R. (1988). *Racial Hygiene: Medicine Under the Nazis*. Cambridge, MA: Harvard University Press.

Programs for the handicapped. (1969, November). *U.S. Department of Health, Education, and Welfare*. Retrieved from https://mn.gov/mnddc/dd_act/documents/hew_newsletters/11_3_69-PFH-HEW.pdf

Public Law 441.257, The U.S. Code, Title 42: The Public Health and Welfare, Chapter 4: Centers for Medicare and Medicaid Services, Subchapter C. Medical Assistance Programs; Subpart F. Sterilizations; Informed Consent (1978).

Richards, S. B., Brady, M. P., & Taylor, R. L. (2014). *Cognitive and intellectual disabilities: Historical perspectives, current practices, and future directions*. New York: Routledge.

Roberts, J. A. F. (1952). The genetics of mental deficiency. *The Eugenics Review, 44*(2), 71–83.

Schalock, R. L., & Luckasson, R. (2013). What's at stake in the lives of people with intellectual disability? Part I: The power of naming, defining, diagnosing, classifying, and planning supports. *Intellectual and Developmental Disabilities; Washington, 51*(2), 86–93.

Scheerenberger, R. C. (1983). *A history of mental retardation*. Baltimore, MD: Paul H. Brookes.

Schmich, M. (2017, July 20). Family marshals of city's Disability Pride Parade proud of differences. Retrieved from http://www.chicagotribune.com/news/columnists/schmich /ct-chicago-disability-pride-parade-mary-schmich-0721-20170720-column.html

Section 504, Rehabilitation Act of 1973, 29 U.S.C. § 701 (1973).

Siegel, S. A. (2005). Justice Holmes, Buck v. Bell, and the history of equal protection. *Minnesota Law Review, 90*, 106.

Smith, T., & Eikeseth, S. (2011). O. Ivar lovaas: Pioneer of applied behavior analysis and intervention for children with autism. *Journal of Autism and Developmental Disorders, 41*(3), 375–378. https://doi.org/10.1007/s10803-010-1162-0

Social Security Act, Title XIX, 42 U.S.C. §§1396–1396v (1965).

Sofair, A. N., & Kaldjian, L. C. (2000). Eugenic sterilization and a qualified Nazi analogy: The United States and Germany, 1930-1945. *Annals of Internal Medicine, 132*(4), 312–319.

Special Olympics: Our history. (2018). *The Joseph P. Kennedy Jr. Foundation for the Benefit of Persons with Intellectual disabilities*. Retrieved from https://www.specialolympics.org/Sections/What_We_Do/History/History.aspx

Stern, A. M. (2007). "We cannot make a silk purse out of a sow's ear": Eugenics in the hoosier heartland. *Indiana Magazine of History*. Retrieved from https://scholarworks.iu.edu/journals/index.php/imh/article/view/12254

Tassé, M. J., Luckasson, R., & Nygren, M. (2013). AAIDD proposed recommendations for ICD-11 and the condition previously known as mental retardation. *Intellectual and Developmental Disabilities; Washington, 51*(2), 127–131.

The Education for all Handicapped Children Act (EAHCA), Pub. L. No. 04–142, 89 Stat. 773–796 (1975).

Tredgold, A. F. (1908). *Mental Deficiency: (Amentia)*. London: Baillière, Tindall and Cox.

Waterstone, M. (2015). Backlash, Courts, and Disability Rights. *Boston University Law Review; Boston, 95*(3), 833–849.

Williams, J. E. J. (1994). 75 years of caring for the disabled. (National Easter Seal Society). Fund Raising Management, *25*(n7), p12(6).

Wold, S. M. (1975). O'Connor v. Donaldson: Due process and the involuntarily civilly committed mental patient. *Tulsa Law Journal, 11*, 604.

World Programme of Action Concerning Disabled Persons (n.d.) Retrieved from https://www.un.org/development/desa/disabilities/resources/world-programme-of-action-concerning-disabled-persons.html.

Wyatt v. *Stickney*, 325 F. Supp. 781 (1971).

Yell, M. L., Rogers, D., & Rogers, E. L. (1998). The legal history of special education: What a long, strange trip it's been! *Remedial and Special Education, 19*(4), 219–228. https://doi.org/10.1177/074193259801900404

Theories of Intelligence

2

Matthew J. Euler and Ty L. McKinney

Introduction

A prominent definition of intelligence describes the construct as follows:

> Intelligence is a very general mental capacity that, among other things, involves the ability to reason, plan, solve problems, think abstractly, comprehend complex ideas, learn quickly, and learn from experience. It is not merely book learning, a narrow academic skill, or test-taking smarts. Rather, it reflects a broader and deeper capability for comprehending our surroundings—"catching on," "making sense" of things, or "figuring out" what to do (Gottfredson, 1997a, p. 13).

This working definition has been broadly endorsed and repeated by numerous scholars in the field (Haier, 2016; Nisbett et al., 2012; Protzko, 2017) and captures many of the traits and capabilities that scientists and lay people alike would consider important parts of intelligence. Notably though, while it does a good job of describing various things that intelligent people tend to do, and distinguishes the construct from others (e.g., book knowledge), it also recapitulates a key tension at the heart of the intelligence literature. Namely, how is it that intelligence is on the one hand a very *general* capacity and yet attempts to define it very often invoke a host of apparently discrete behaviors and tendencies? For intelligence theorists, this question can be translated into the issue of whether intelligence truly represents a singular or *unitary* capacity, as much of the psychometric literature has suggested, or whether the appearance of a unitary structure is merely a statistical or measurement artifact, such that intelligence actually represents an aggregate of many capacities.

As detailed below, the issue of whether intelligence is a unitary capacity arguably represents the most important issue concerning theories of intelligence and can serve as an organizing theme for the models discussed in this chapter. Beyond that, any theory of intelligence can then be grouped according to (1) whether it is better construed as a historical or a contemporary theory; (2) whether its evidence base is primarily *psychometric* (based on statistical modeling of the correlations among various types of mental tasks), *experimental* (derived from analyses of laboratory-based measures, rather than norm-referenced standardized tests), *conceptual* (derived from literature reviews and rational considerations), or even *physiological* (arrived at through correlations of intelligence task performance with EEG or neuroimaging data); and finally (3) whether it emphasizes the importance of more *basic* (i.e., sensory perceptual) or more

M. J. Euler (✉) · T. L. McKinney
Department of Psychology, University of Utah,
Salt Lake City, UT, USA
e-mail: matt.euler@psych.utah.edu

J. L. Matson (ed.), *Handbook of Intellectual Disabilities*, Autism and Child Psychopathology
Series, https://doi.org/10.1007/978-3-030-20843-1_2

complex tasks in measuring and defining intelligence. Upon exhausting those dimensions, each theory of intelligence can ultimately be characterized in terms of its more distinguishing features and contributions to the literature. Given that psychometric research forms the bulk of the evidence base for these theories in general, the next sections outline the history of psychometric models of intelligence (also termed "structural" models), from Charles Spearman's discovery of psychometric *g* up through the development of multifactor and hierarchical models in the middle and later part of the last century.

Early Psychometric Theories of Intelligence

Spearman, the Birth of Factor Analysis, and Psychometric *g*

The most important historical figure in psychometric intelligence research is undoubtedly Charles Spearman, whose key ideas are still relevant today. As reviewed in his 1904 monograph, prior research in early psychology had produced conflicting findings about the nature of intelligence. On the one hand, Francis Galton (discussed below) and others had advocated for a view of intelligence as a single mental capacity that had its basis in relatively primitive functions such as sensory discrimination (e.g., discriminating fine differences between the weights of objects or the pitches of auditory tones). However, other scholars had either failed to find evidence for this claim or had at times contradicted it showing that more complex mental abilities better reflected the nature of intelligence (Spearman, 1904). Having reviewed the literature on both sides and found it wanting, Spearman sought out to more rigorously investigate the relation between sensory discrimination and intelligence, in an effort to improve the theoretical and empirical foundations on which the debate rested.

For that purpose, Spearman collected measures of sensory discrimination in samples of school children and related them to various tentative estimates of intellectual ability, such as class rankings in various subjects (Classics, English, French, Mathematics, and Music) and teachers' and others' general impressions of the students' abilities. Notably, Spearman also included a precursor to the contemporary IQ score in the form of a mental age measure, where he distinguished between a form of intelligence he called "present efficiency" (absolute academic performance) and "native capacity" (a student's performance in a given subject divided by their age). Overall, these studies produced several important contributions.

First, Spearman recognized the need to control for the unreliability of measurements, which, if achieved, would allow one to obtain a better estimate of the true relationship between two variables (Jensen, 1998a, p. 23). Second, Spearman also identified that by examining the variance that is shared across different types of tasks (e.g., visual and auditory discrimination, proficiency in Greek vs. piano-playing; Spearman, 1904, p. 259), one could determine the extent to which they involve one or more underlying dimensions or faculties. This marked the beginning of factor analysis (Bartholomew, 2004, p. 18), wherein Spearman laid the mathematical foundations to be able to ask whether, for example, auditory and visual discrimination reflect wholly distinct capacities, owing to their basis in different sensory modalities, or whether the variance between people on each reflects a more general sensory discrimination ability. That is, factor analysis allows one to assess how many dimensions contribute to variance in performance on a group of psychological tasks. For that reason, it has come to be the primary tool in psychometric intelligence research, including many of the following models discussed in this chapter. It also brings us to Spearman's third and most important contribution, where he showed that, indeed, there was evidence for "general discrimination" and "general intelligence" factors that were common to all of the respective measures and, further, that the two factors were related to such a large degree that they seemed to both draw on a single mental capacity (Spearman, 1904, p. 272).

While contemporary estimates of the correlation between intellectual ability and sensory discrimination suggest that the relationship is considerably smaller than Spearman's initial claim (Acton & Schroeder, 2001), the basic finding has nevertheless proved reliable (Sheppard & Vernon, 2008) and provides meaningful support for the reductive program in intelligence research, as outlined further below. More importantly, however, Spearman's broader suggestion that a single capacity might be common to all cognitive tasks has proved to be even more central.

In particular, it has now been shown that across hundreds of datasets (Carroll, 1993), whenever a sufficiently diverse set of mental tasks is administered to a sufficiently large and representative group of people, the tasks will invariably positively intercorrelate, producing the so-called *positive manifold* phenomenon. Crucially, this is an empirical phenomenon and not a logical necessity, in the sense that there is nothing inherent to the statistical procedures that require that grades in Mathematics, Classics, and French must be positively correlated. Rather, their correlation reflects an empirical truth. Not only did Spearman observe the now-ubiquitous positive manifold among the academic and sensory measures, but he further saw that the correlations between the various subjects exhibited a clear hierarchy, such that grades in English and French related more strongly to grades in Classics than either did to Math. He then showed that upon extracting what he called the common "general intelligence" factor, all of the measures—sensory and academic alike—seemed to derive their variance from only this general factor and a second, idiosyncratic test-specific factor (see Fig. 2.1). This in turn made it possible to determine how "saturated" each task was with the general factor (i.e., its "*g*-loading" in contemporary parlance) and could ultimately allow for identifying which tasks would provide the best of measures of overall intelligence (Spearman, 1904, p. 277).

Altogether, these results provided the basis for Spearman's "two-factor" theory[1] of mental ability, which argues that the variance between people on any given task appears to be a function of only two factors: that which is *specific* to each task (termed "*s*") and that which is *general* or shared, now known as psychometric *g* (Spearman, 1927).

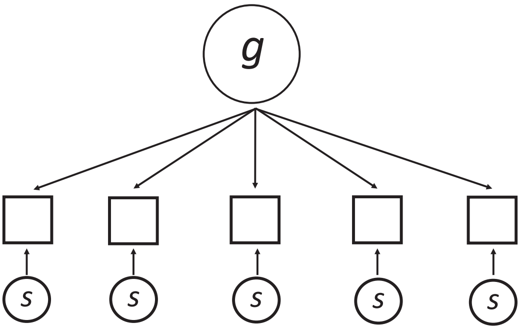


Fig. 2.1 Spearman's two-factor theory. Spearman's two-factor theory of intelligence asserts that the variation between people on all cognitive tasks is a function of two factors: general intelligence (*g*) and test-specific variance (*s*). A hypothetical battery of five tests is shown, where the variance in each test reflects only the contributions of *g* and test-specific variance. Following typical conventions for depicting structural equation models, latent factors are depicted as circles, and manifest variables (obtained test scores) are depicted as squares

In the time since Spearman's initial observations, it has become essentially universally accepted that a general factor can often account for much of the variance among cognitive tests (typically approaching 50%; Deary, 2012). Yet, as will be seen, the status and significance of this factor remain an important area of debate. On that note, it is appropriate to turn to a discussion of Spearman's primary interlocutors and the alternative models they favored.

Thurstone and Multifactor Theories

Spearman's two-factor theory of intelligence is most often and readily distinguished from Louis L. Thurstone's model of Primary Mental Abilities (Thurstone, 1938). The latter exemplifies an alternative class of models that advocated a multifactor rather than unitary structure for mental abilities (Sattler, 2008, p. 224). As the name implies, Thurstone's model emphasized the idea

---

[1] The name "two-factor" theory can be somewhat confusing, in the sense that it strongly emphasizes the importance of the single *g* factor. Nevertheless, the name refers to the assertion that any task involves contributions from two factors: *g*, which is common to all tasks, and *s*, which is unique to particular tests.

that, rather than invoking just a single general factor along with test-specific factors in explaining cognitive performance, a group of independent, lower-order processes, with a clearer psychological meaning (e.g., memory, verbal skills, spatial skills, etc.), might better explain the patterns of correlations among cognitive tests. Indeed, a form of this idea can be seen in Spearman's very own hierarchy, where he observed that grades in certain subjects (e.g., Classics, French, and English—all notably linguistic) correlated more highly with one another than they did with other measures.

Inspired by that premise, Thurstone devised a large set of cognitive tasks that were designed to be maximally pure in their reliance on what were presumed to be independent capacities. Using an alternative factor-analytic technique that forces the extraction of maximally independent factors (Jensen, 1998a, pp. 75–76), he was then able to derive a model that consisted in seven, seemingly independent, primary abilities (see Fig. 2.2). Among these were capacities like memory, perceptual speed, and spatial visualization (Sattler, 2008), which exhibit an intuitive degree of conceptual separation, as well as others like verbal comprehension and word fluency that might be expected to have more variance in common. As it happens, this turns out to be the case, as Thurstone himself later recognized. That is, although it is possible to achieve maximally independent factors by (1) carefully selecting tests on that basis and then (2) mathematically constraining the relations between resulting factors, in truth, when the latter constraints are relaxed, the primary factors will positively intercorrelate. As explained by Jensen (Jensen, 1998a, Chapter 4), this is due to the fact that although some tests may load only on one primary ability, they all will still invariably load on *g*.[2] Moreover, the latter will often account for considerably more variance in performance than do the primary abilities. In turn, the *g* factor can then be modeled at a level above the primary abilities, or even alongside them, to account for their intercorrelations, as in the contemporary hierarchical and bi-factor models described below. As will be seen, these latter models provide something of a compromise position (Deary, 2000, Chapter 1).

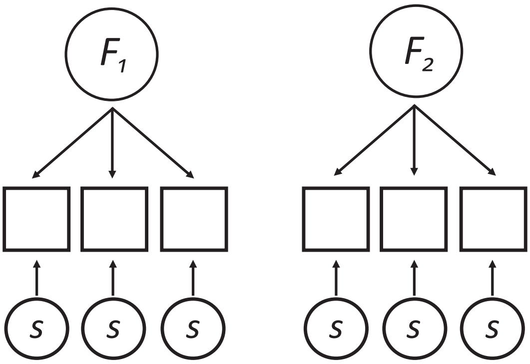


Fig. 2.2 Schematic factor model depicting two orthogonal primary abilities. A simplified version of Thurstone's primary ability model is shown here with only two factors, derived from three tests each. The key distinguishing features of Thurstone's initial model are the presence of uncorrelated (mathematically "orthogonal") primary abilities and the absence of a general factor. F_1 and F_2 designate hypothetical primary abilities (e.g., verbal and spatial skills)

Sampling Theory and Thomson's "Bonds" Model

While it is instructive to distinguish between the extremes represented by Spearman's two-factor theory and Thurstone's initial model based on Primary Mental Abilities, in recent years, a third alternative outlined by their contemporary, Godfrey Thomson, has been rediscovered. As recently described by Bartholomew and colleagues, Thomson's work is unique, in that it does not necessarily dispute the mathematical adequacy of two-factor theory, but somewhat like Thurstone's early model, it objects on conceptual grounds (Bartholomew, Deary, & Lawn, 2009). Specifically, whereas Thurstone emphasized the need for psychological and conceptual coherence in psychometric models of intelligence, Thomson was concerned with biological plausibility in terms of neural organization.

---

[2] It should be noted that Thurstone himself came to recognize these issues and acknowledged the possibility of a high-order *g* (Carroll 1993, p. 56; Major et al. 2012, p. 544).

As an intuitive, if high-level example, consider the well-known neurological findings that specific, higher-order faculties (e.g., language comprehension) can be disrupted by focal lesions, while leaving other cognitive skills intact. This of course suggests that cognitive functions are subserved by at least partly dissociable neural systems and that individual differences therein might not be the result of a single underlying factor. Yet, unlike Thurstone, Thomson did not argue that the underlying neural processes should be wholly independent, in the sense that they would never interact. Rather, just as we now know that language production and comprehension are both largely housed in the dominant hemisphere (Lezak, Howieson, Bigler, & Tranel, 2012) and even interact (Papathanassiou et al., 2000), one might expect these abilities to positively correlate based on their overlapping substrates.

In essence, Thomson extended this basic logic to develop a broader account of how diverse neural processes could give rise to the appearance of a single underlying factor in mental abilities. He reasoned that this could occur any time the items or tests in a given battery each "sampled" the same set of discrete but connected neural processes (which he termed "bonds"). In turn, as the number of bonds shared among tasks increases, so too should the correlation between them. Thus, Thomson argued, and more recent work has confirmed (Bartholomew, Allerhand, & Deary, 2013) that it is possible to develop a statistical model that replicates the *g* factor, despite actually being comprised of numerous underlying processes. Further, although not recognized at the time, Thomson's proposal makes even more sense when one considers the finding that the tasks with the highest *g*-loadings are nearly always the most complex. Given that complexity has been explicitly defined as the number of processes involved in a task (Guttman, 1954; Marshalek, Lohman, & Snow, 1983; Stankov & Raykov, 1995), it is quite suggestive that the most complex (and hence most *g*-loaded) tasks might in fact involve the most overlapping processes. Interestingly, like much of this literature, this idea, presented here in a psychometric context, would come to be echoed later on in more conceptual theories.

Cattell's Legacy: Fluid and Crystallized Intelligence and Investment Theory

Working in the middle and later part of the twentieth century, Raymond B. Cattell made a major contribution to this literature in developing the concepts of fluid and crystallized intelligence (respectively, denoted as Gf and Gc). In a seminal paper, Cattell (1963) described his effort to synthesize factor-analytic methods with developmental considerations as they pertain to the growth of intelligence. One of his key assertions was that "there is not *one* "general ability" second-order factor…but more," with each having a semi-independent basis and developmental trajectory (Cattell, 1963; emphasis in original). Specifically, he argued that it is possible to separate the *g* factor into Gf and Gc, where the former represents the capacity to respond adaptively in novel situations, while the latter is drawn upon by tasks that require learned skills and knowledge. Concretely, Gf would be expected to play more of a role in solving tasks involving unfamiliar relationships or components, such as in the well-known Raven's matrices (Raven & Court, 1998). These tasks are broadly nonlinguistic and require examines to identify the shared features among abstract figures arranged in a matrix. Gc, on the other hand, loads heavily on linguistic, academic, and other skill-based knowledge and should be more important for tasks involving things like verbal reasoning and one's general fund of information.

In line with these distinctions, Gf was hypothesized to more closely reflect one's inborn, momentary cognitive processing capacity, which is then *invested* in developing crystallized skills (thereby forming the crux of "investment theory"; Cattell, 1987). In addition, the trajectory of Gf and Gc should follow a particular developmental course, with Gf increasing as children reach adulthood but declining in middle and older age, whereas Gc would in principle continue expanding as individuals accrue further experience. Indeed, this general trajectory has been roundly confirmed (Salthouse & Davis, 2006) and represents a key consideration in developing norms for cognitive tests. Notably, although the Gc-Gf model may sound very similar to that of Primary Mental Abilities, it is distinguished from this and other "multifactor" models in that its components are allowed to intercorrelate (a so-called oblique model; Kovacs & Conway, 2016). Last, although the original Gf-Gc model contained only two factors (see Fig. 2.3), Cattell and his coworker John Horn later extended it to include ten broad abilities (McGrew, 2009).

Ultimately, while the Gf-Gc distinction has been retained in some more recent psychometric models (notably CHC theory, see below), it is not without its challenges. Most significantly, it is now well-established that although it is possible to separate Gf and Gc statistically, if one allows for a *g* factor at a third level in a higher-order model (see next section), Gf and *g* then correlate at or close to unity (Kan, Kievit, Dolan, & van der Maas, 2011; and Lichtenberger & Kaufman, 2013, p. 32–33). This of course makes them statistically and potentially substantively indistinguishable. Second, as noted by Cattell (1963), the theoretical distinction between Gf and Gc has important implications for determining which sorts of tests would be the most likely to show bias due to educational or cultural factors. Yet, although it is clearly the case that tests which presume certain linguistic and cultural information are not appropriate for some examinees, it unfortunately does not follow that the so-called "nonverbal" measures are in fact culture fair or "culture free." To the contrary, recent work examining age differences in performance on the Raven's progressive matrices suggests that such tasks are in fact highly sensitive to broader cultural knowledge, such as that which changes over successive generations (Fox & Mitchum, 2012).

Other aspects of the Gf-Gc model have received mixed support as well. On the one hand, there is meaningful support for investment theory's claim that investing in developing a certain type of skill may limit the growth of others (e.g., resulting in strong verbal and weaker math skills or vice versa; Coyle, Purcell, Snyder, & Kochunov, 2013; Coyle, Snyder, Richmond, & Little, 2015). However, at the same time, other research has undermined some of its other claims (e.g., Kievit et al., 2017). Thus, while the Gf-Gc distinction still finds support in facets of the literature, the overall challenges faced by the model have given sufficient reason to consider alternatives.

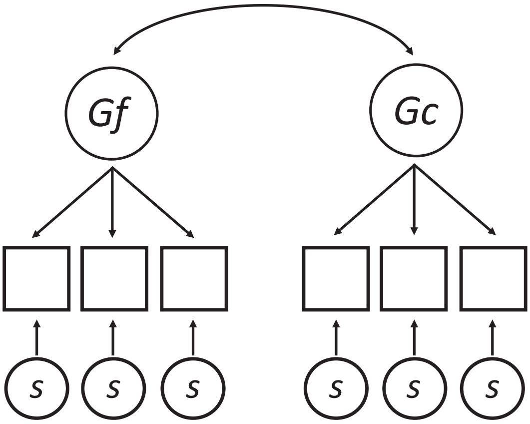


Fig. 2.3 Oblique fluid and crystallized intelligence model. Oblique factor structure depicting a schematic version of the initial Gc-Gf model. Unlike Thurstone's Primary Mental Abilities model, the two factors are allowed to intercorrelate

Carroll and Vernon's Hierarchical Models

The last group of notable historical models is the hierarchical models developed by Phillip Vernon (2014) and John Carroll (1993).[3] Like Spearman, these theorists posited a *g* factor that was common to all cognitive tasks, but like Thurstone and Cattell, they also acknowledged and incorporated other factors that had a clearer psychological meaning. They accomplished this through the process of successive factorization (developed by

---

[3] Although many authors have considered Carroll's model to be a quintessential example of a hierarchical model (Deary 2000; Lubinski 2004), it has also been argued that Carroll's view was in fact closer to the bi-factor model discussed in the next section (Beaujean 2015).

Thurstone, 1947; cited in Carroll, 1993, p. 637), where, after extracting first-order factors that corresponded to broad abilities, the remaining correlations between those factors were then factor analyzed themselves, giving rise to a single higher-order *g* factor and thereby creating a hierarchy.

Vernon's model (depicted schematically in Fig. 2.4) posited two major group factors below the level of *g*. These are v:ed, which refers to verbal or educational skills, and k:m, or kinesthetic and mechanical skills (Johnson & Bouchard, 2005b). Below those two major factors, the model then contained the six minor factors of creative abilities, numerical skills, and verbal fluency, which loaded on v:ed, and spatial, psychomotor, and mechanical ability factors which loaded on k:m (Sattler, 2008, p. 227). Although Vernon's verbal-educational and kinesthetic-mechanical factors share some similarities with Gf and Gc, in contrast to Cattell, he argued that v:ed and k:m were both susceptible to cultural influences (educational and noneducational, respectively). Further, he chose to omit Gf, due to its considerable statistical overlap with *g* (Johnson & Bouchard, 2005b).

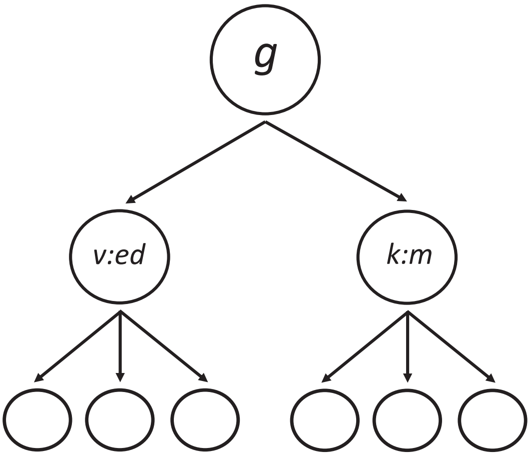


Fig. 2.4 Vernon's hierarchical model. Schematic depiction of Vernon's hierarchical model with a second-order *g* factor that explains the correlations between the verbal-educational and kinesthetic-mechanical group factors. Test-specific factors and manifest variables are omitted for simplicity

As recounted by many authors (Beaujean, 2015), Carroll's work and the resulting model are among the most celebrated achievements in psychometric intelligence research. He arrived at his framework through a meticulous reanalysis of over 450 previous datasets, from which he synthesized the then-extant literature to arrive at his "three-stratum" theory of cognitive ability. In the model, the strata, or level, at which a given ability was placed partly reflected the number of successive factorizations that were performed in deriving it but more directly corresponded to the factor's *generality* in terms of the types of lower-order factors that loaded on it (Carroll, 1993, p. 577). At the lowest level were *narrow* (Stratum I) abilities, or those that are specific to particular tasks (e.g., visualization and sequential reasoning in matrices tasks). Broad (Stratum II) factors then constituted group factors, like those employed by Thurstone and Cattell and Horn, that were common to a given class of tasks. Finally, *g* formed the single Stratum III factor representing general cognitive ability (Carroll, 1993, Chapter 16).

In summarizing his results, Carroll derived four primary conclusions: (1) that there was "abundant" evidence for a general factor of intelligence that would emerge at the highest order of factorization in any given dataset, (2) that eight broad abilities could be distinguished at Stratum II (fluid intelligence, crystallized intelligence, general memory and learning, broad auditory perception, broad retrieval ability, broad cognitive speediness, and processing speed or reaction time decision speed), (3) that additional second-order factors could further be identified in domains such as learning and memory and language (among others), and (4) that the general program of analyzing abilities into strata provided valid insights into the structure of human cognitive ability and could form the basis for such a theory.

Aside from the significance of Carroll's overall achievement, his model was also noteworthy in that, in addition to providing a relatively definitive account of the broad ability factors known at that time, he also arranged them according to their approximate *g*-loadings (Fig. 2.5).

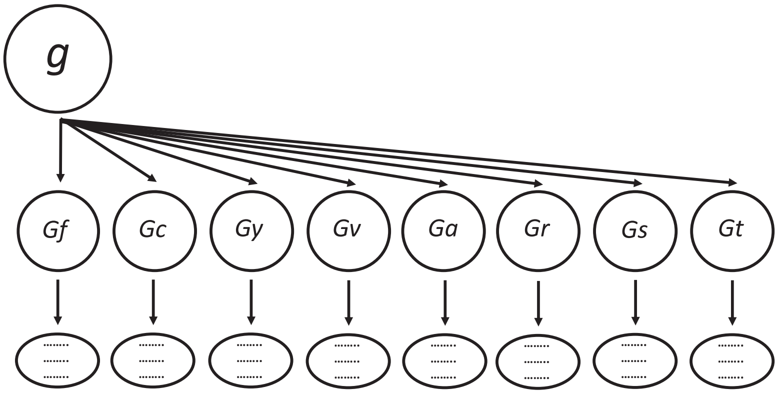


Fig. 2.5 Carroll's three-stratum model of cognitive abilities. Adaptation of Carroll's (1993) hierarchical model, with *g* at Stratum III and broad abilities ordered by their approximate *g*-loadings from left to right. Stratum II factors use contemporary abbreviations, following McGrew (2009). *Gf* fluid intelligence, *Gc* crystallized intelligence, *Gy* general memory and learning, *Gv* broad visual perception, *Ga* broad auditory perception, *Gr* broad retrieval ability, *Gs* broad cognitive speediness, *Gt* reaction time decision speed. Ovals containing ellipses depict narrow Stratum I variables, which varied in number across Stratum II factors

This is useful, because in principle, such an ordering should help to clarify which sorts of measures best predict overall intelligence and hence might inform the "true" nature of construct (e.g., does intelligence have more to do with on-the-spot reasoning or with one's depth and breadth of general knowledge?). Carroll's model is also remarkable for the fact that it truly represented a comprehensive synthesis of the available literature (Johnson & Bouchard, 2005b, p. 395). For example, although he included Gf and Gc in his model, the proximity of the former to *g* also acknowledges the debate (both then and since; Gustafsson, 1984; Colom, Rebollo, Palacios, Juan-Espinosa, & Kyllonen, 2004; Oberauer, Schulze, Wilhelm, & Süß, 2005) about whether Gf or other lower-order factors best define the limits on intelligence. Thus, it is perhaps due to the success of this synthesis that Carroll's model forms one half of the prominent, contemporary Cattell-Horn-Carroll integrated model, or simply CHC (McGrew, 2009).

Contemporary Psychometric Theories of Intelligence

Having incorporated the lessons of the prior century, the current phase of psychometric intelligence research is generally characterized by a clearer acknowledgment of the statistical validity of the *g* factor and a move toward discussing how best to represent it in model structures, what its substantive implications might be, and on determining the number and status of the broad ability factors. At present, there are arguably four major models under discussion.

Cattell-Horn-Carroll (CHC) Theory

The CHC model is likely the most prominent contemporary psychometric theory. Its elements have long been incorporated into the Woodcock-Johnson Cognitive Assessment Battery (initially as Gf-Gc theory; Woodcock, 1990; and later as CHC itself; Woodcock, McGrew, & Mather, 2001), it has received prominent treatments both in the scholarly literature and in practical assessment guides (Lichtenberger & Kaufman, 2013; Newton & McGrew, 2010), and at the time of this writing, a major review of CHC (McGrew, 2009) lists more than 700 citations in the Google Scholar database.

According to a recent chapter detailing the tenets of CHC, the theory consists in two primary components. First, it provides a "taxonomy" or classification of human cognitive abilities, and second, it provides "a set of theoretical explanations for how and why people differ" in those respects (Schneider & McGrew, 2012). As the name indicates, CHC represents a synthesis of the Cattell-Horn extended theory and John Carroll's three-stratum model. Although *g* is included, owing to the disagreement between its namesakes regarding its status (McGrew, 2009, p. 4), the substantive emphasis of CHC is decidedly on the broad and narrow factors. Not only is the casual status of *g* left undetermined, but practitioners who adopt CHC are explicitly encouraged to ignore it if they question its theoretical value and particularly for practical aspects of assessment (Schneider & McGrew, 2012, p. 111). A schematic depiction of CHC would thus be similar to Fig. 2.5 above but lacking a causal arrow from *g* to the broad factors or the emphasis on *g*-loadings.

Although the most well-known treatment of CHC lists ten broad (Stratum II) abilities along with six tentative factors, McGrew and colleagues have since reorganized aspects of the framework to clarify the conceptual groupings among the factors at that level (Schneider & McGrew, 2012). The present section draws heavily from their excellent exposition, which interested readers should consult for a deeper treatment, as well as practice guidelines. The first conceptual grouping corresponds to Domain-Free General Capacities and includes Fluid Reasoning (Gf), two memory factors (Short-Term Memory, Gsm, and Long-Term Storage and Retrieval, Glr), and three cognitive speed factors (Processing Speed, Gs; Reaction and Decision Speed, Gt; and Psychomotor Speed, Gps). These factors group together because they all emphasize more process-related aspects of cognition (e.g., how fluently can one perform a task, how readily can one recall information) as opposed to particular content.

Next, CHC emphasizes four broad, content-related factors under the heading of Acquired Knowledge. These are Comprehension-Knowledge (Gc), Domain-Specific Knowledge (Gkn), Reading and Writing (Grw), and Quantitative Knowledge (Gq). Naturally, these factors depend at least in part on educational and cultural exposure, and on that basis Schneider and McGrew (2012) emphasize their conceptual alignment with Cattell's crystallized intelligence factor. While the expression of these skills clearly depends on specific content and hence cultural exposure, it remains the case that many narrow capacities in this domain nevertheless have process aspects as well. For example, while this domain includes the narrow capacities of General Verbal Information and Lexical Knowledge (loading on Gc), it also includes skills such as Reading Comprehension and Writing Speed (including under Grw). As an interesting side note related to this domain, despite its seemingly straightforward character, Gc arguably represents one of the more controversial factors in intelligence research, both in the CHC model and in the field as a whole. For example, in contrast to early theory which predicted stronger genetic influences on fluid rather than crystallized skills (Cattell, 1963), there is now evidence that more culture-loaded factors may actually be the *most* heritable abilities (Kan, Wicherts, Dolan, & van der Maas, 2013). Indeed, even the status of Gc as a true cognitive capacity is currently under debate (for further reading, see Kan et al., 2011).

In the last conceptual grouping, CHC emphasizes a set of more modality-specific capacities, in the form of Sensory-/Motor-Linked Abilities. Each of these factors is tied to its respective sensory modality, where the list includes Visual Processing (Gv), Auditory Processing (Ga), Olfactory Abilities (Go), Tactile Abilities (Gh), Kinesthetic Abilities (Gk), and Psychomotor Abilities (Gp). Note that these factors are not thought to be mere perceptual capacities but more complex mental operations that depend upon a given modality. For example, Gv entails visualization skills such as those required for mental rotation tasks or for imagining obscured portions of objects, while Ga includes phonemic decoding (implicated in some forms of dyslexia; Coslett, 2003) and memory for sound patterns. However, these examples notwithstanding, it remains the case that assessing some of these skills often entails mere acuity tests (e.g., olfaction, tactile skills; Schneider & McGrew, 2012). Overall, while this grouping undoubtedly includes valid dimensions of individual differences, it nevertheless also seems clear that additional research in this area is needed (Stankov, 2017).

Finally, with regard to CHC's theoretical implications, the current framework is perhaps closest to Cattell and Horn's views which emphasized the diversity of abilities, as opposed to the relative primacy of any given factor in defining intelligence. This is perhaps also consistent with CHC's very practical emphasis, in that it aims to bridge the "theory-to-practice gap" in cognitive and educational assessment (McGrew, 2009, p. 4). That is, because CHC emphasizes broad and narrow abilities, it provides a very practical framework from which to guide assessment of individual people, with their unique and diverse needs. Thus, whereas the ambiguous status of *g*, or some of the less-well established Sensory-/Motor-Linked Abilities, is somewhat unsatisfying from a theoretical perspective, CHC nevertheless has the unique virtue of eschewing those sometime esoteric debates in favor of very explicitly seeking to inform practical considerations.

Verbal-Perceptual-Image Rotation (VPR)

An equally valid, but somewhat underappreciated, model is the VPR theory, which was first articulated by Johnson and Bouchard (Johnson & Bouchard, 2005b). Using a dataset comprised of 42 cognitive ability tests that had been administered to 436 adults, those authors set out to examine whether the Cattell-Horn two-factor Gf-Gc model, Vernon's v:ed-k:m model, or Carroll's three-stratum model best fit the data. Although they concluded that each model fit reasonably well, they also noted a number of issues. For example, the Gf-Gc model demonstrated a high correlation between those two factors, suggesting the presence of a *g* at a third stratum. In addition, the same model contained two lower-order abilities below the primary level that reflected fluid and crystallized skills, but these proved to be statistically indistinguishable from their respective higher-order factors. The Vernon model performed well, but did not meet the authors' a priori criteria for model fit, failed to represent memory, and also showed a very high correlation between its lower-order verbal factor and v:ed. In the three-stratum model, the fluid intelligence factor was indistinguishable from *g*.

On that basis, the authors sought to evaluate a new model, using Vernon's model as a guide but adding another factor to better account for the data. Ultimately, they arrived at what they termed the VPR model (Fig. 2.6), which, as the name indicates, identifies three broad factors below the level of *g*: verbal ability, which includes things like linguistic and scholastic skills (e.g., memorization); perceptual ability, which includes skills like the ability and speed with which one detects patterns; and image rotation, which more uniquely captures perceptual tasks that emphasize rotation per se (note that some tests load on multiple factors; Johnson & Bouchard, 2005b).

In discussing the advantages of the VPR model, the authors noted that whereas Gf-Gc includes both verbal and perceptual content as part of crystallized intelligence (e.g., mechanical ability is both learned and spatial), both Vernon's model and the VPR model distinguish these as loading on factors that reflect their respective content. Indeed, such a division of verbal and spatial skills has been repeatedly borne out in replications of VPR, where it has been shown that various tests which should theoretically reflect crystallized intelligence (e.g., naming pictures, identifying tools) have actually had *negative* loadings on the crystallized factor (Johnson & Bouchard, 2005a; Johnson, te Nijenhuis, & Bouchard, 2007). Over the course

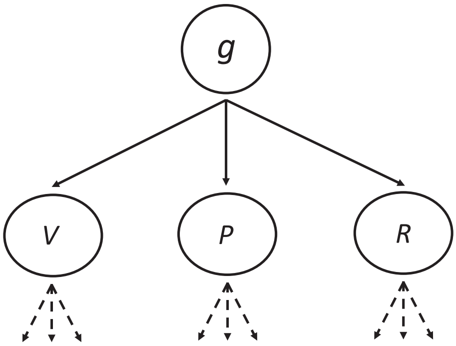


Fig. 2.6 Verbal-perceptual-image rotation model. Schematic depiction of the VPR model. Dashed arrows reflect the fact that first-order factors are unspecified (see text). *V* verbal, *P* perceptual, *R* image rotation

of those and subsequent studies, Johnson and colleagues have now consistently demonstrated the relative superiority Vernon's model over that of Gf-Gc, for both statistical and substantive reasons, as well as the superiority of VPR in turn (Johnson et al., 2007; Johnson & Bouchard, 2005a). VPR has since been shown to fit better than either the extended Gf-Gc model or a three-stratum version of CHC (Major, Johnson, & Deary, 2012).

From a theoretical standpoint, VPR is unique in emphasizing a *dimensional* view of intelligence, such that verbal and rotational skills are thought to represent two poles on a continuum (Major, Johnson, & Deary, 2012). Notably, this may conceptually align it with the well-known (relative) specializations of each cerebral hemisphere (i.e., the left hemisphere is typically dominant for language and sequential processing; the right hemisphere is typically dominant for spatial and configural processing; Lezak et al., 2012), giving it a strong degree of neurological plausibility (Johnson & Bouchard, 2005b). Relatedly, the VPR model also explicitly avoids pre-specifying the number and content of lower-order factors (Major et al., 2012, p. 544), arguing that these instead will depend on the test batteries at hand. Thus, when constructing a model based on a given sample and battery, the obtained perceptual factor may correlate more highly with verbal ability, depending on the exact tests from which it is derived (e.g., if the stimuli tend to induce verbalization), while in another battery with a greater proportion of highly spatial tasks, the perceptual factor might relate more strongly to image rotation.

Overall then, VPR represents a departure from the tradition of precisely specifying models down to all of their constituents. Rather, it aims for a more parsimonious view of intelligence, wherein a *g* factor influences task performance by virtue of its relation to just a few higher-order factors, which are aligned on a single verbal-spatial dimension. While it may be possible to enumerate all of the lower-order capacities in the manner of CHC, VPR instead prioritizes parsimony in its account of intelligence. Although its practical applications are less-developed than that of CHC, its strong base of empirical and substantive support nonetheless makes it a viable alternative to its more prominent counterpart.

Bi-factor Models of Intelligence

Another important class of models to briefly consider is the bi-factor models, which are unique in the way that they represent the relationship between *g* and the group factors. In all of the hierarchical models discussed up to this point, *g* influences individual test scores solely by virtue of its relation to the group factors (Cucina & Byle, 2017). This means that the latter account for all the shared variance among the tests that load on them (e.g., the shared variance among a group of working memory tasks would be fully explained by a working memory factor), with *g* then being modeled at a higher level to account for the correlations between the group factors. Under this scenario, any change in an individual's level of *g* can only impact their test performance through the resulting impact on the relevant group factor(s) (Beaujean, 2015).

In contrast, bi-factor models also allow one to represent *g*, but they instead place it in a nonhierarchical relation. Here, rather than first extracting primary factors and then modeling *g* based on their relations, *g* is extracted first, and primary factors are extracted separately from the remaining shared variance among the tests (Jensen, 1998a, p. 78). This means that the resulting factors are independent of variance due to *g*, with all the latent factors existing at the same level of the model (Gignac, 2016b; Morgan, Hodge, Wells, & Watkins, 2015; and see Fig. 2.7). The result is that variation in test performance across individuals can reflect *either* variance due to *g* or that due to the primary factors, with the two being completely independent (Beaujean, 2015; Morgan et al., 2015).

Following from the previous example, in a bi-factor model, one's score on a given working memory test would be a function of *both* one's level of "general intelligence" and their independent, more specific working memory ability. Setting aside the various technical considerations

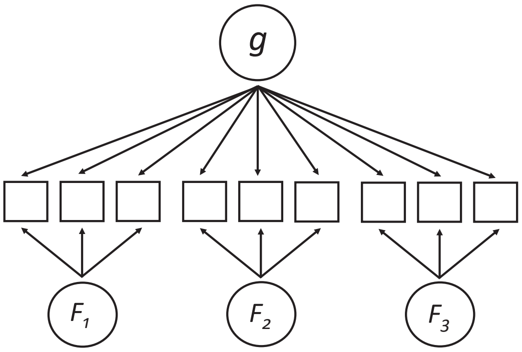


Fig. 2.7 Example bi-factor model. Schematic bi-factor model with three group factors. Boxes represent measured variables (specific tests), and circles represent latent factors as above. Each arrow going from a latent factor to a measured variable reflects the former's unique contribution to variance in the latter

related to the challenges and virtues of the bi-factor model (see Gignac, 2016b; Morgan et al., 2015; Murray & Johnson, 2013), these models are of substantive interest because they provide an alternative way to think about *g*. Namely, because both the general and group factors independently account for test performance in these models, they seem to allow a greater reconciliation between the empirical evidence for *g*, on the one hand, and the notion that more specialized skills might have additional explanatory utility on the other (e.g., Benson, Hulac, & Kranzler, 2010).

The Mutualism Model

The final psychometric model is somewhat akin to Thomson's sampling theory in also offering a very different perspective on psychometric *g*. Specifically, whereas sampling theory argues that the positive manifold arises from a situation where cognitive tests recruit overlapping sets of neural connections, the mutualism model holds that initially uncorrelated processes, such as group factors like memory and perceptual ability, come to be correlated through reciprocal positive interactions that occur during development (van der Maas et al., 2006). In essence, what are initially independent processes come to support the growth of one another, thereby increasing their interrelations. Some related examples from the empirical literature come from the work of Demetriou and colleagues, who have shown that various lower-order factors appear to play different roles in intelligence at different developmental phases (Demetriou et al., 2013; Makris, Tachmatzidis, Demetriou, & Spanoudis, 2017).

Using simulation studies,[4] van der Maas et al. (2006) demonstrated that given a model containing no relationships among limiting resources but with positive correlations between otherwise independent processes, factor analyses of the simulated data are nonetheless consistent with a single major latent factor such as psychometric *g*. As a result, because the model is comprised of multiple, separate but *correlated* processes, the resulting statistical pattern cannot be said to result from a single underlying cause. Thus, although fully hypothetical, the mutualism model suggests an alternative route by which *g* could statistically arise, but without a unitary cognitive or neurological basis. In turn, van der Maas and colleagues further demonstrated that the mutualism model could also explain several other important phenomena in the intelligence literature. These include the more complex cognitive hierarchies like the models described above, as well as patterns of intellectual development, intra-subject variability, and cohort effects on IQ test performance (see discussion and details in van der Maas et al., 2006).

In the time since mutualism was first proposed, the theory has received mixed support. For example, although mutualism suggests that *g* should account for more variance in test performance as children grow older, a study that examined this in a cross-sectional sample ranging from 2 to 90 years old failed to support this (Gignac, 2014). A second study also argued against mutualism when it failed to find expected patterns in factor-analytic results (Gignac, 2016a). However, proponents of mutualism debated the prior

---

[4] Primary simulations were based on 16 hypothetical neurocognitive processes, which were each independently sampled from pre-specified distributions to define 1000 simulated subjects. Different models specified various constraints and relationships among model parameters.

study's premise (van der Maas & Kan, 2016). These issues notwithstanding, in the most recent and direct test of mutualism to date, a longitudinal study did find evidence for reciprocal positive interactions between two broad abilities over the span of almost 2 years (Kievit et al., 2017).

Finally, the mutualism model provides a good venue to raise an additional point related to modern conceptions of psychometric ability models. Specifically, while most of the models considered here are said to be *reflective* in nature, mutualism (along with sampling theory and possibly VPR; Deary, Cox, & Ritchie, 2016; Major et al., 2012, p. 544) instead represents what is known as a *formative* model (van der Maas, Kan, & Borsboom, 2014). In a reflective model, the latent factors are understood to be the cause of the observed variation in the individuals' test scores, in the sense that factors are taken to represent real entities (Borsboom, Mellenbergh, & Van Heerden, 2003; Kievit et al., 2011), without which the observed results would not be obtained. As an example, whenever one attributes a global quality to an individual, e.g., he or she broke the law because they are simply a "bad person," one is implicitly making a reflective explanation—an underlying attribute of the person was the cause of their behavior. Under this scenario, because of the specified causal direction, changing the subtests that are used to measure the variable (intelligence in this case) would change the accuracy of the measurement, but could not logically affect the variable itself (Kievit et al., 2011).

In contrast, although formative models also relate latent factors to observed data, here, the factors are merely understood to be weighted composite scores, such that the data are responsible for the composition of the factor, as opposed to vice versa (van der Maas et al,. 2014). Common examples of formative variables include constructs like socioeconomic status and "overall health," where it can be readily seen that many diverse factors come together to define them. Researchers might disagree about whether cardiovascular efficiency or strength and flexibility are more important aspects of health, but they likely would not argue that an underlying "health factor" was the cause of variation in both. The two are of course often related in healthy people, but it is possible to have one without the other. In turn, to the extent that one chose to measure one dimension at the exclusion of the other, these choices could produce very different pictures of a person's overall health. In this instance, the choice of indicators determines the attributes of the factor, rather than the reverse (see Fig. 2.8). Finally then, returning back to intelligence, it can be seen how the formative/reflective distinction provides a useful way for thinking about *g*. Although reflective models *imply* a unitary cause for variation in intelligence, the mutualism model (along with sampling theory) shows that this is not necessarily the case. Rather, *g* could in fact be legitimately unitary in a statistical sense, while still ultimately resulting from many underlying processes (Deary et al., 2016).

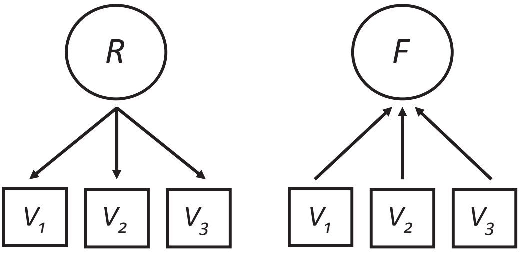


Fig. 2.8 Reflective vs. formative models. In a reflective model (left), including most models of intelligence, the latent factors are to understood as causing the variation in the obtained scores (V_1–V_3). In a formative model (right), the casual direction is reversed such that the composition, and thus the nature, of the latent factor entirely depends on the variables used to measure it. Error and residual terms and other parameters are omitted for simplicity.

Summary of Psychometric Theories

To summarize this section, psychometric theories of intelligence have addressed the "structure" of the construct, considering the role of various specific and broad factors (e.g., Gf and Gc) and of course the role of *g*. While early historical accounts debated whether intelligence is better reflected by a single general factor (*g*) or a series of independent skills, more recent theories have generally endorsed a hybrid of the two. That is, intelligence likely has a hierarchical structure, with *g* at the top, and broad factors operating between *g* and performance on particular types of tests. Practically, this suggests that a single general capacity influences all tasks, regardless of their content, although more specific abilities govern the relation between *g* and specific tasks. More recent theories have also debated the nature of these broad factors, with CHC perhaps being the most comprehensive, though VPR offers a conceptually simpler account centered around a verbal-spatial continuum. These two theories emphasize continuity with different theoretical traditions, but they differ in their emphasis on practical utility vs. theoretical consistency. Still other psychometric theories acknowledge the empirical phenomenon of *g* but propose alternative ways that it might come about, in terms of the interrelations between lower-level cognitive and neural processes. Overall, while psychometric theories are useful for thinking about the structure of intelligence (i.e., is it a single capacity or many?), they say less about its "nature." These latter issues, such as whether intelligence relates to more basic or complex processes, and its more-detailed information processing basis, are better addressed by conceptual theories of intelligence, as reviewed in the next section.

Conceptual Theories of Intelligence

Because psychometric theories involve a high degree of mathematical rigor, they have perhaps justifiably dominated discussions about intelligence. Nevertheless, a number of more conceptually oriented theories still have their place in filling out the scope of these debates. These models run the gamut from being primarily theoretical and rationally derived, to those that are backed by a substantial body of empirical results. In the latter category, some models (especially those that emphasize working memory and processing speed) could also claim substantial psychometric support. Yet, insofar as they advocate for *particular* processes in defining intelligence and also bring experimental methods to bear on their views, they are somewhat more similar to conceptual models than the primarily statistical frameworks outlined above. In every case, though, the models and theorists highlighted below have been selected for review because of the ways in which they exemplify other key or controversial ideas in theories of intelligence.

Francis Galton: The Reductive Tradition in Intelligence Research

Sir Francis Galton is generally regarded as the most important early figure in the modern era of intelligence research.[5] His work took place in the context of Victorian England with evolutionary theory in the intellectual milieu (Jensen, 1998a), including its emphasis on the role of genes in determining the traits of organisms. Whereas many at the time held that intelligence could "act independently of natural laws," Galton held the contrary, evolutionary view that genes and the biological processes they govern should be important determinants of intellectual ability (Galton, 1883). In turn, by examining the implications of just that single assertion, one can derive what are perhaps Galton's three most important intellectual contributions.

First, if mental capacities are akin to physical traits in being subject to biological principles, this means that, like other phenomena in the natural sciences, intellectual capacity should be amenable to objective quantification. Thus, it should be possible to develop reliable measures of intellectual ability that can quantify differences among individuals. Second, if mental ability is largely determined by genetic variation, then more fundamental attributes of individuals (those which are more closely tied to their genes) might place constraints on higher-order mental skills. For Galton, this meant that the basic efficiency of lower-order, sensory-motor capacities (those that should be more directly tied to basic nervous system functions, like nerve conduction

---

[5] Though see Deary's (2000, Chap. 2, and p. 68) account of several authors who espoused similar views but predated Galton

speed) should limit "the field upon which our judgment and intelligence can act" (Galton, 1907; cited in Wasserman, 2012). Thus, while Galton did not necessarily hold that intelligence could be *fully* reduced to lower-order capacities, the primacy he placed on them nevertheless exemplifies the reductive tradition. Third, Galton also made a number of statistical contributions, being the first to develop a measure of the relation between two variables (the forerunner to Pearson's correlation coefficient) and to use the normal distribution to rank individuals on an interval scale (Jensen, 1998a, pp. 10–11). Thus, he provided a foundation not only for differential psychology but also for normative principles that are still used today.

In pursuit of these various ideas, Galton established his "anthropometric laboratory" in London, where, for a small fee, members of the public could be measured on mostly low-level traits and capacities, such as height, grip strength, breathing capacity, and visual and auditory acuity (Sattler, 2008; Wasserman, 2012). Although he amassed data on more than 9000 individuals, likely owing in part to methodological limitations, his measures proved only weakly related to indices of intelligence (e.g., occupational attainment; Jensen, 1998a, Chapter 11). As recounted by Deary (2000, Chapter 2), subsequent prominent but likely overemphasized failures to apply Galton's measures to *practical* assessment may in part account for the waning of interest in Galton's approach and the following stagnation in reductive research. In the time since, the approach has rebounded, and a more balanced view has emerged. On the one hand, many recent studies, including meta-analyses and population-based research, clearly support Galton's contention that lower-order processes should relate to intelligence (e.g., Acton & Schroeder, 2001; Deary, Der, & Ford, 2001; Euler, McKinney, Schryver, & Okabe, 2017; Melnick, Harrison, Park, Bennetto, & Tadin, 2013). Nevertheless, to the extent that effect sizes are modest (typically correlating near $r = -0.30$; Sheppard & Vernon, 2008), their conceptual significance remains open to debate.

Alfred Binet and David Wechsler: Theoretical Insights Gleaned from Practical Assessors

In contrast to Galton's decidedly theoretical aims, the work of Alfred Binet and David Wechsler, who respectively developed the Binet-Simon and Wechsler Intelligence Scales, illustrate a number of insights that were gleaned through more practical endeavors. Specifically, while it would be difficult to discount the predominant role of theory and basic research in advancing this field, the systematic research through which these measures were developed nevertheless yielded important principles that have stood the test of time. Indeed, even if there were no principles guiding their development (untrue in either case; Binet & Simon, 1916; Wechsler & Edwards, 1974), the immense practical success of these instruments (and the tests they inspired) would seem to speak to their construct validity. To wit, if an instrument is near-universally accepted as a valid measure of intelligence, then it stands to reason that the principles that govern it may help elucidate the construct.

In the case of Binet, he and his colleague Theodore Simon developed what is now generally recognized as the first useful test of intelligence in the Binet-Simon scale, with the goal of better identifying children with intellectual disabilities (Sattler, 2008). In the course of their studies, Binet articulated what has proved to be a fundamental principle not only of intelligence testing but of intelligence itself. Specifically, he identified that more complex measures involving multiple dimensions are far superior to simple tasks as measures of intelligence—for the reason that complex tasks elicit more variability across individuals (Wasserman, 2012, p. 14). Here then, is a clear contrast between the lessons suggested by Galton and Binet's work. As Binet acknowledged, simpler measures can be more precisely controlled with respect to stimulus characteristics and the like. Yet, because simple measures elicit little variability across individuals, this mitigates their advantage over less precise but more *sensitive* tasks that involve more complex mental operations (Binet & Henri, 1894; quoted in Wasserman, 2012). Thus, although narrow psychophysical capacities may meaningfully relate to, and even constrain, overall intelligence, the construct itself seems to inherently represent a *higher-order* capacity. In addition, Binet also emphasized the necessity of using multiple tasks to arrive at a composite intelligence measure (Binet & Simon, 1916), and he considered individual tasks to be of questionable value (Boake, 2002). Not only has this become a foundational principle of most intelligence tests, but it also reflects the core theoretical notion that intelligence represents a fundamentally general capacity (Gottfredson, 1997a).

Although David Wechsler was more theoretically inclined than Binet (and produced a number of theoretical papers; Wechsler & Edwards, 1974), he was also largely concerned with practical assessment. Here again though, it can be seen how his practical considerations served to clarify a key feature of intelligence. In particular, while serving in the army, Wechsler was tasked with individually examining unschooled recruits or those with limited English proficiency, who had been deemed intellectually disabled based on their performance on the very verbally dependent Stanford-Binet (Boake, 2002). Yet, in many cases it was evident to Wechsler that these recruits had previously held significant social and professional responsibilities, attesting to their average if not higher intelligence. Based on these experiences, Wechsler came to believe that intelligence could be expressed equally well through verbal and "performance-based" measures (i.e., perceptual and visuospatial tasks) and strove to include diverse measures spanning both dimensions in developing his own scales (Matarazzo, 1972, Chapter 8).

On the one hand, this represents a practical and clinical necessity. For certain examinees, such as those with various neurological syndromes or with limited English proficiency, one might indeed expect their intelligence to be better expressed through a particular route. Yet, like Binet, Wechsler's clinical insight around this reflected a deeper fact about intelligence itself. Namely, that although most healthy individuals will not demonstrate such discrepancies, intelligence itself is broader than any single dimension and thus can be expressed through multiple routes. Not only is this evident in the ubiquity of *g* but also in the somewhat lesser-known phenomenon of the "indifference of the indicator." As discovered by Spearman (1927), this refers to the finding that highly diverse measures with no content in common (e.g., vocabulary vs. visuospatial reasoning) can in some instances predict intelligence equally well (see: Wechsler, 2008; Table 5.1). Thus, as in Binet's case, although practical considerations drove Wechsler's approach, the insights he derived reflected important aspects of intelligence.

Arthur Jensen and Lazar Stankov: Contemporary Debates Around Reductive Approaches

On the spectrum of unitary-non-unitary perspectives of intelligence, Arthur Jensen is a strong supporter of an overall *g* factor. Where Jensen differs from psychometric approaches, however, is in his extensive use of mental chronometry (reaction time-based tasks; RT) as a means to assess information processing accounts of intelligence. As a *g*-theorist, Jensen believed that intelligence could be reduced down to one fundamental process, which he tried to capture through patterns of RT performance (Jensen, 1981). He proposed that speed of information processing (SOP; Jensen, 1993) was this fundamental process and tried to link IQ scores to mathematical models of RT task performance (Hick's Law; Hick, 1952). These studies argue that intelligence is about how long it takes one to process a "bit" of information (Jensen, 1981, 1982, 1998a, 1998b), with variation in other processes (e.g., working memory—one's ability to hold and manipulate information held in mind) being the result of variability in SOP (Jensen, 1993). While a correlation between RT and IQ scores is a well-replicated finding (Sheppard & Vernon, 2008), there is much debate about the nature of this relationship (Stankov & Roberts, 1997) and what it means for understanding intelligence.

In an attempt to resolve these questions about RT-IQ correlations, mental chronometry has explored how discrete processes typically examined in cognitive psychology (working memory, etc.) could be related to intelligence. For example, while overall RT correlates with IQ scores, so does the variability of one's RT within a given task (RT standard deviation; RTSD; Doebler & Scheffler, 2016; Jensen, 1992). Likewise, one's slowest individual RT trial often correlates better with intelligence than their fastest RT (Rammsayer & Troche, 2016; Ratcliff, Schmiedek, & McKoon, 2008). This led to theories that higher-ability people may experience less "neural noise" (Jensen, 1993) or have better "evidence accumulation" during decision-making (Schmiedek, Oberauer, Wilhelm, Süß, & Wittmann, 2007). Finally, since chronometric tasks lend themselves well to neuroscience methods, the two approaches have begun to merge. Indeed, Jensen argued that explanations for chronometric effects would only be valid insofar as they were biologically plausible (Jensen, 1993). In a way, many contemporary neuroscience-based theories of intelligence (such as Process Overlap Theory, discussed below) could be considered intellectual descendants of Jensen's initial linkage of experimental and differential psychology.

An interesting contrast with Arthur Jensen's reductive approach can be found in the work of Lazar Stankov and colleagues. While Jensen tried to link chronometric performance to a unitary *g*, Stankov instead used experimental approaches to consider how the coordination of several processes, as opposed to a particular one, might define intelligence. Specifically, he argues that intelligence reflects the capacity to engage multiple processes simultaneously to accomplish a task. Indeed, Stankov has provided experimental evidence demonstrating that tasks with increasing numbers of components (i.e., more complex tasks) correlate better with intelligence (Stankov & Crawford, 1993; Stankov & ,Raykov 1995). While Stankov acknowledges *g*, he argues that it is generally overemphasized and that second-stratum factors are more important to understanding intelligence (Stankov, 2017). In line with his emphasis on higher-order coordination of discrete processes, Stankov has also advocated for the integration of non-cognitive process into models of cognitive ability (e.g., emotional intelligence and meta-cognition; Stankov, 2017). Thus, in contrast to Jensen who emphasized the primacy of *g*, and speed of processing as its ultimate basis, Stankov's work illustrates an alternative experimental approach that eschews strong reductionism and emphasizes a more holistic perspective.

The Role of Executive Abilities: Working Memory Capacity and Process Overlap Theory

The negative correlation between intraindividual variability in RT and IQ scores (meta-analysis: Doebler & Scheffler, 2016) suggests that attention may be an important component of intelligence, for the reason that higher variability may reflect attentional lapses. The execution of the top-down attention (commonly called executive functioning; EF) is thought to reduce RT variability and promote higher Working Memory Capacity (WMC; Bellgrove, Hester, & Garavan, 2004; Miyake, Friedman, Rettinger, Shah, & Hegarty, 2001). Given these findings, it is perhaps not surprising that structural models often find strong relationships between intelligence and WMC. Research examining the known neurophysiological correlates of these constructs has highlighted the importance of the prefrontal cortex (Kane & Engle, 2002, 2003) as a common neural substrate of both EF and Gf (a robust correlate of *g*). These authors argue that fluid reasoning is achieved through the manipulation of information held in working memory and that WMC (and thus EF as well) acts like a "bottleneck" on reasoning and the expression of intelligence (Kane & Engle, 2002). In the time since, process overlap theory (POT; Kovacs & Conway, 2016) has been developed from this original WMC model. POT integrates experimental (e.g., RT variability, complexity effects) and psychometric (e.g., IQ-WMC correlation) findings to propose frontal-parietal networks as the ultimate neural basis of the aforementioned bottleneck, consistent with the known neuroimaging correlates of intelligence (Basten, Hilger, & Fiebach, 2015; Jung & Haier, 2007). While POT is a relatively new theory, it integrates a number of findings in the intelligence literature and, unlike many theories, proposes fairly concrete neurological correlates. While POT strongly emphasizes WMC, the latter's relation to intelligence is perhaps best thought of as a necessary, but not sufficient explanation (Redick et al., 2013), with intelligence being a broader construct and WMC acting as a limiting factor.

The Planning, Attention, Simultaneous, and Successive Model

The Planning, Attention, Simultaneous, and Successive (PASS) model of intelligence was developed by Naglieri and Das (1990) and was heavily influenced by the work of the esteemed neuropsychologist Alexander Luria. Like POT, PASS also emphasizes the role of EF but also considers lower-level cognitive operations in its conceptualization of intelligence. PASS theory outlines how any complex task involves three different functional units that are differentially involved depending on the task demands. The first unit corresponds to basic attention processes, the second unit corresponds to perceptual processing and multimodal sensory integration, and the third unit corresponds to the planning, maintenance, and correction of behavior. Das and Naglieri argue that these three units are involved in all tasks, whose effective coordination and integration with prior knowledge constitute intelligence. That is, intelligence is the capacity to (1) effectively engage with the environment (2), process information from the environment (3), and plan/execute effective behavior in the environment based on that information. The authors argue that the structured nature of current IQ tests do not adequately capture the first and third functional units (specifically basic attention and planning processes), thus limiting their capacity to make more sophisticated claims about an individual's real-world achievement.

In support of the PASS model, studies have found that neuropsychological tests of executive functioning (corresponding to the third unit) are more predictive of daily functioning (Barkley & Fischer, 2011) and achievement (Clark, Prior, & Kinsella, 2002) than IQ tests alone. A handful of studies suggest that PASS-based measures are at least as sensitive to dysfunction in ADHD and academic achievement as traditional IQ tests (Naglieri & Bornstein, 2003; Naglieri, Goldstein, Delauder, & Schwebach, 2005; Naglieri, Goldstein, Iseman, & Schwebach, 2003), though some argue that PASS is merely a recapitulation of certain CHC factors (Keith, Kranzler, & Flanagan, 2001). While the evidence for PASS is somewhat limited, it highlights the synergistic importance of processes at all levels of the cognitive hierarchy, from basic attention to complex planning and reasoning, when considering real-world success.

Detterman's System Theory of Intelligence

Finally, Detterman's (1987) system theory of intelligence is noteworthy here because in addition to trying to explain the nature of intelligence, it explicitly addresses intellectual disability. As outlined above, a major debate in intelligence research concerns whether intelligence is fundamentally unitary, as at least superficially implied by psychometric *g*, or whether various correlated but ultimately independent factors better represent the construct's true structure. For individuals with cognitive disabilities, this could (hypothetically) translate into either (1) a deficit in overall ability (*g*) or, possibly, (2) a deficit in some but not all cognitive abilities. Detterman's model provides an innovative, partial synthesis of these two ideas, in that it recognizes and explains *g* but in a way that also acknowledges the multiple factors that make up intelligence.

Specifically, Detterman takes a *systems* view, in arguing that intelligence arises through the interactions of "independent but interrelated parts" (Detterman, 1987). In explaining this concept, he likens IQ scores (which aggregate performance over multiple tests) to the ways in which one might rank universities. For example, one might evaluate various different aspects of a university, like the quality of the faculty and the facilities, the size of the endowment, its physical location and layout, etc., and sum them together to derive an overall score. As just described, each aspect could in principle be fully independent, with the resulting score reflecting an aggregate index of the university's overall quality. Note however, that in practice, such factors will likely not only be correlated, but any *central* factors, like the size of the endowment, will set a limit on the level of the others (Detterman, 1987). Thus, in the case of intelligence, various group factors (or still lower-level processes) might seem independent in principle, yet if some are more central than others, they will place limits on the rest. Indeed, the working memory and process overlap theories of intelligence make exactly this case.

Detterman's theory also has specific implications for intellectual disability. He argues that if intelligence reflects the operation of an interconnected neural system, where some parts are more central than others, this implies that inefficiencies in central resources should cause scores among cognitive tests to be more interrelated at lower levels of ability (Detterman, 2002). Conversely, individuals with highly efficient central processing resources will have fewer constraints on the development of their more specific capacities, and hence cognitive test scores should be less highly related at higher ability levels. Indeed, Detterman and Daniel (1989) found evidence for this exact phenomenon (also known as cognitive differentiation), which has been replicated in numerous other studies (Blum & Holling, 2017; Tucker-Drob, 2009). In the case of intellectual disability, Detterman predicted that it "would be shown to be a deficit in one or more basic abilities…with a moderate to high degree of centrality (Detterman, 1987, p. 8)." Notably though, as discussed further on, he was also careful to note that this does not mean that all intellectual disabilities result from a single cause.

Other Prominent Theories

Finally, there are two other prominent theories of intelligence that also deserve brief mention. Both Howard Gardner's theory of multiple intelligences and Robert Sternberg's Triarchic Theory of Successful Intelligence are widely cited accounts, which remain prominent in some fields. Although these theories do not have strong support among most contemporary intelligence researchers (Warne, Astle, & Hill, 2018), their popularity in applied settings warrants a brief review.

Gardner's Theory of Multiple Intelligences

On the spectrum of intelligence theories which include few to many factors, multiple intelligences (MI) is perhaps only less expansive than that of CHC. Published in the book *Frames of Mind* (Gardner, 2006), MI outlines the view that there are eight conceptually distinct intelligences, which are all of equal importance, with no hierarchical structure. Gardner arrived at these ideas through extensive readings in a variety of different fields, but did not generate any data to empirically test his theory. To date, experimental studies trying to validate MI theory have yielded limited empirical support (Almeida et al., 2010; Visser, Ashton, & Vernon, 2006).

Overall, while the notion that there can be multiple distinct forms of intelligence, which are not all cognitive in nature, is appealing in some respects, this idea is better supported by other theories, with a larger empirical base. For example, Stankov (2017) has argued that emotional intelligence should be considered as a second-order factor in CHC, thereby presenting a more empirically grounded alternative to Gardner's interpersonal and intrapersonal intelligences. Likewise, Carroll convincingly argued that capacities such as musical and mathematical ability, which are included in MI in various forms, actually express themselves via different contents at different levels of ability and also involve numerous lower-order skills (e.g., visualization, induction, sequential reasoning, in the case of mathematical ability). Thus, Carroll argued that such constructs should be regarded as "inexact… popular concepts[s]" rather than distinct, scientifically supported cognitive capacities (Carroll, 1993, pp. 626–627). Finally, insofar as there is essentially overwhelming evidence for the *psychometric* reality of *g* (as distinct from its potential neural basis, which remains undetermined), the literature provides little support for the view that all aspects of intelligences are equally influential, with no hierarchical features.

Sternberg's Triarchic Theory of Successful Intelligence

Finally, Robert J. Sternberg's prominent theory of successful intelligence also deserves a mention here. Overall, Sternberg emphasizes the ecological aspects of intelligence, contrasting between intelligence as typically discussed, and *successful intelligence*, which is argued to be a more comprehensive construct that considers one's sociocultural context and idiographic patterns of strengths, weaknesses, and experience (Sternberg, 1999). His conceptualization of intelligence strongly emphasizes the interactive, transactional nature of an individual within their environment (i.e., the adaption, selection, and shaping of one's environment), focusing on how factors like specific goals, prior experience, and contextual information can change the expression of intelligence (Sternberg, 2012). As such, he argues that the measurement of intelligence should depend on the goals and the environmental context of the individual, as opposed to traditional IQ tests. Sternberg's triarchic theory emphasizes the role of not only analytical skills (his term for what most IQ tests measure) but also creative (i.e., idea generation) and practical skills (i.e., implementation of ideas in real-world settings), so that an individual can be successful as they define it within their environment (Sternberg, 2012). Collectively, these skills are referred to as process skills, which together with meta-components (somewhat like executive functions as described by Suchy, 2015) and knowledge acquisition skills (i.e., learning) are argued to better capture the notion of success in a culturally neutral way. Overall, Sternberg's notion of successful intelligence integrates many often-disparate approaches to studying intelligence (i.e., factor-analytic, experimental, culturally relative, application-based approaches, etc.), and has both some strengths and weaknesses.

Perhaps the most controversial aspect of the triarchic theory relates to the distinctiveness of analytical and practical intelligence and to the extent to which the latter is more predictive of real-world success. Psychometric evaluation of an intelligence test designed specifically to assess the triarchic theory found that a modified *g* model was the best fit for the data (Chooi, Long, & Thompson, 2014). A separate study highlighted that many of Sternberg's findings could potentially be explained by measurement error (Brody, 2003). Additionally, Gottfredson (2003) critically reviewed the studies supporting the triarchic theory and concluded that practical intelligence and traditional IQ scores are statistically related (as opposed to distinct entities), especially when methodological aspects of the studies are considered (e.g., size and diversity of the samples). She further asserted that there is no evidence supporting the idea that a "practical *g*" exists as a separate factor apart from the conventional *g* (an argument supplemented by a separate paper; McDaniel & Whetzel, 2005). In summary, although both the triarchic and MI theories have conceptual merits, relative to the other theories discussed in this chapter, they each have more limited empirical support.

Overall Summary

The preceding sections aimed to give an overview of the historical development of theories of intelligence, with an eye toward articulating the key principles and debates and clarifying current thinking in the area. The overall consensus from psychometric approaches suggests that intelligence consists in an individual's general cognitive capacity, which influences every mental task they might undertake, irrespective of its nature. Although lower-order abilities have a clear and legitimate place in the cognitive hierarchy—consisting in more domain-general, content-related, and modality-specific skills (to borrow from CHC)—their unique significance in determining one's overall intelligence remains a matter of debate. Unlike most contemporary models, which are largely hierarchical, sampling theory

(Bartholomew et al., 2009) and mutualism (van der Maas et al., 2006) both highlight how *g* can arise from a non-unitary basis, giving psychometric credence to the idea that intelligence may result from a collection of interacting processes rather than a single fundamental one.

While psychometric approaches clarify the relationships between cognitive abilities in a detailed way, they are somewhat agnostic as to what *g* and various broad factors reflect in a more functional sense. This is where conceptual theories can help fill in the gap. This latter work has clarified that while lower-order processes play a role in intelligence, it is clearly best characterized as a higher-order capacity and one that is particularly implicated in managing cognitive complexity (Gottfredson, 1997b). Thus, intelligence appears to relate not only to the level of one or more discrete abilities but also to their efficient coordination in support of adaptive behavior. Finally, insofar as the most recent era of experimental and theoretical studies has supported the centrality of particular processes, it appears likely that capacities like WMC and other executive skills (Kovacs & Conway, 2016) or possibly speed of processing (Jensen, 1993; Schubert, Hagemann, & Frischkorn, 2017) may constrain overall intelligence. These constraints are apt to be strongest at lower levels of ability, whereas at higher ability levels, profiles should be more differentiated (Detterman, Petersen, & Frey, 2016). Tables 2.1 and 2.2 summarize the major features of the models discussed in this chapter.

Table 2.1 Overview of historical theories of intelligence

Theory	Major theme	Status of *g*	Emphasizes basic vs. complex processes	Evidence base
Galton	Reductionism	Unitary	Basic	Psychometric, experimental
Spearman	Discoverer of *g*	Unitary	Complex	Psychometric, experimental
Binet	Complex tasks, practical assessment	Ambiguous	Complex	Psychometric
Thurstone	Primary abilities	Non-unitary	Complex	Psychometric
Thomson	Sampling mental bonds	Non-unitary	Basic	Simulation, psychometric
Cattell	Fluid vs. crystallized intelligence	Non-unitary	Complex	Psychometric
Carroll	Hierarchical factor structure	Hierarchical	Complex	Psychometric
Wechsler	Performance-based, practical assessment	Non-unitary	Complex	Psychometric

Table 2.2 Overview of contemporary theories of intelligence

Theory	Major theme	Status of *g*	Emphasizes basic vs. complex processes	Evidence base
Jensen	Speed of processing	Hierarchical	Basic	Psychometric, experimental
Gardner	Multiple intelligences	Non-unitary	N/A	Conceptual
Sternberg	Triarchic/successful intelligence	De-emphasized	Complex	Conceptual, experimental
PASS	Planning, attention, simultaneously, successive	Non-unitary	Complex	Conceptual, psychometric
Detterman	Systems	Non-unitary	Basic	Psychometric, experimental, simulation
Stankov	Role of complexity and group factors	De-emphasized	Complex	Psychometric, experimental

(continued)

Table 2.2 (continued)

Theory	Major theme	Status of *g*	Emphasizes basic vs. complex processes	Evidence base
VPR	Verbal, perceptual, spatial rotation	Hierarchical	Complex	Psychometric
CHC	≥10 broad factors	De-emphasized	Complex	Psychometric
Bi-factor	Simultaneously models *g* and group factors	Unitary	Complex	Psychometric
Mutualism	Developmental, *g* as epiphenomenal	Non-unitary	Basic	Simulation, psychometric
Working memory/process overlap theory	Executive attention	Hierarchical/hybrid	Complex	Psychometric, experimental, physiological

Implications for Intellectual Disability

In applying these concepts to intellectual disability (ID), perhaps the biggest implication is that intelligence, as it manifests empirically through *g* and in turn through IQ scores, is indeed a very general capacity (and note that IQ scores share >80% of their variance with *g*; Kranzler, Benson, & Floyd, 2015). This is all the more so in ID, given that narrow abilities are more strongly interrelated at lower levels of IQ. In turn, since intellectual functioning is of course one of the two central features in the diagnosis of ID (the other being adaptive functioning; American Psychiatric Association, 2013), the effect and hence the goal of the diagnosis should be to identify individuals whose cognitive challenges span diverse content and situations. Thus, unlike other cognitive disorders which are characterized by some degree of specificity (e.g., specific-learning disorders, ADHD), a diagnosis of ID indicates more pervasive cognitive difficulties.

Notably, the ID diagnosis also helps to clarify what intelligence is not. Because ID also requires adaptive impairments, which further establish diagnostic severity, it highlights the fact that cognitive functioning does not wholly determine one's functional capabilities. This same distinction holds within much of intelligence research, where although earlier theories did emphasize the role of non-cognitive factors (e.g., motivation; Wechsler, 1943), current theories (and particularly psychometric models) tend to restrict themselves to purer cognitive measures. Importantly, this is not to deny the role of such variables in adaptive behavior (e.g., Duckworth, Quinn, Lynam, Loeber, & Stouthamer-Loeber, 2011) but merely to distinguish between cognition and intelligence strictly construed and the other traits and capacities that people bring to bear on life's challenges.

A second implication relevant to the concept of ID is the fact that while psychometric theories seem to make implicit claims about *neural* organization (i.e., they address the relatedness of one cognitive function to another), ID is of course a *behavioral* diagnosis the reflects a diverse group of underlying causes (Lee & Harris, 2006, Chapter 5). Thus, while the ID diagnosis clearly depends upon cognitive capacities that are mediated by the brain, it encompasses a wide group of ultimate neural etiologies. This means that although the diagnosis should correctly identify persons who face broad cognitive challenges, because it does emphasize intelligence, it will tend to obscure more subtle, individual needs. Given that many of the syndromes responsible for ID are now known to be associated with more specific cognitive profiles (Edgin, 2013; Grigsby, 2016), this calls for bringing the neuropsychological approach into individual assessment of ID (see Chap. 27, this volume).

On a final more conceptual note, as Detterman observed (Detterman, 1987), different potential causes for psychometric *g* have different implications for the ways in which ID might arise. In the case of hierarchical theories or those that emphasize one or more central factors in intelligence (e.g., POT, system theory), one might expect the functioning of various core networks to strongly determine one's level of intelligence. Physiologically, this is perhaps most akin to parieto-frontal integration theory (P-FIT; Jung & Haier, 2007) and similar accounts (Duncan, 2010) that emphasize the centrality of those specific regions to overall intelligence. In contrast, mutualism makes a developmental case that initially independent factors become increasingly integrated through time. Finally, as recently pointed out by Deary, Cox, and Ritchie (Deary et al., 2016; citing Burt, 1940), *g* might also arise though a shared adaptive *property* of the brain, as opposed to the features of one or more major structures. That is, while a single neuroanatomical network might account for the appearance of a unitary *g*, individual, lower-level modules (or even single neurons) could also give rise to *g* if they are all homogenous in terms of their lower-level attributes (e.g., dendritic arborization, neural membrane properties; Jensen, 1998a, p. 121). This too could give rise to a *g* factor but for a very different reason. Thus, although there is strong evidence for the *effective* unity of intelligence as it manifests through testing, the ultimate causes of that cohesion could be numerous and multifaceted.

References

Acton, G. S., & Schroeder, D. H. (2001). Sensory discrimination as related to general intelligence. *Intelligence, 29*(3), 263–271. https://doi.org/10.1016/S0160-2896(01)00066-6

Almeida, L. S., Prieto, M. D., Ferreira, A. I., Bermejo, M. R., Ferrando, M., & Ferrándiz, C. (2010). Intelligence assessment: Gardner multiple intelligence theory as an alternative. *Learning and Individual Differences, 20*(3), 225–230. https://doi.org/10.1016/j.lindif.2009.12.010

American Psychiatric Association. (2013). *Diagnostic and statistical manual of mental disorders (DSM-5)*. Arlington, VA: American Psychiatric Pub.

Barkley, R. A., & Fischer, M. (2011). Predicting impairment in major life activities and occupational functioning in hyperactive children as adults: Self-reported executive function (EF) deficits versus EF tests. *Developmental Neuropsychology, 36*(2), 137–161. https://doi.org/10.1080/87565641.2010.549877

Bartholomew, D. J. (2004). *Measuring intelligence: Facts and fallacies*. Retrieved from https://books.google.com/books?hl=en&lr=&id=sPaS7R5-Wk4C&oi=fnd&pg=PR9&dq=Bartholomew+measuring+intelligencce&ots=33RGo6Zgnb&sig=X3H3VntF0Fa1Re0Vx6PFgel10Zs

Bartholomew, D. J., Allerhand, M., & Deary, I. J. (2013). Measuring mental capacity: Thomson's bonds model and Spearman's g-model compared. *Intelligence, 41*(4), 222–233. https://doi.org/10.1016/j.intell.2013.03.007

Bartholomew, D. J., Deary, I. J., & Lawn, M. (2009). A new lease of life for Thomson's bonds model of intelligence. *Psychological Review, 116*(3), 567–579.

Basten, U., Hilger, K., & Fiebach, C. J. (2015). Where smart brains are different: A quantitative meta-analysis of functional and structural brain imaging studies on intelligence. *Intelligence, 51*, 10–27. https://doi.org/10.1016/j.intell.2015.04.009

Beaujean, A. (2015). John Carroll's views on intelligence: Bi-factor vs. higher-order models. *Journal of Intelligence, 3*(4), 121–136. https://doi.org/10.3390/jintelligence3040121

Bellgrove, M. A., Hester, R., & Garavan, H. (2004). The functional neuroanatomical correlates of response variability: Evidence from a response inhibition task. *Neuropsychologia, 42*(14), 1910–1916. https://doi.org/10.1016/j.neuropsychologia.2004.05.007

Benedek, M., Jauk, E., Sommer, M., Arendasy, M., & Neubauer, A. C. (2014). Intelligence, creativity, and cognitive control the common and differential involvement of executive functions in intelligence and c. *Intelligence, 46*, 73–83.

Benson, N., Hulac, D. M., & Kranzler, J. H. (2010). Independent examination of the Wechsler adult intelligence scale—Fourth edition (WAIS-IV): What does the WAIS-IV measure? *Psychological Assessment, 22*(1), 121–130.

Binet, A., & Henri, V. (1894). Le développement de la mémoire visuelle chez les enfants. *Revue Générale Des Sciences Pures et Appliquées, 5*, 162–169. Retrieved from https://scholar.google.com/scholar?hl=en&as_sdt=0%2C45&q=binet+henri+1894+visuelle&btnG=#d=gs_cit&p=&u=%2Fscholar%3Fq%3Dinfo%3AQJ9OesbTwCkJ%3Ascholar.google.com%2F%26output%3Dcite%26scirp%3D0%26hl%3Den

Binet, A., & Simon, T. (1916). The development of intelligence in children: The Binet-Simon scale. In J. J. Jenkins & D. G. Paterson (Eds.), *Studies in individual differences: The search for intelligence* (pp. 81–111). East Norwalk, CT: Appleton-Century-Crofts. https://doi.org/10.1037/11491-008

Blum, D., & Holling, H. (2017). Spearman's law of diminishing returns. A meta-analysis. *Intelligence, 65*, 60–66. https://doi.org/10.1016/J.INTELL.2017.07.004

Boake, C. (2002). From the Binet-Simon to the Wechsler-Bellevue: Tracing the history of intelligence testing. *Journal of Clinical and Experimental Neuropsychology, 24*(3), 383–405. https://doi.org/10.1076/jcen.24.3.383.981

Borsboom, D., Mellenbergh, G. J., & Van Heerden, J. (2003). The theoretical status of latent variables. *Psychological Review, 110*(2), 203–219. https://doi.org/10.1037/0033-295X.110.2.203

Brody, N. (2003). Construct validation of the Sternberg Triarchic abilities test comment and reanalysis. *Intelligence, 31*(4), 319–329. https://doi.org/10.1016/S0160-2896(01)00087-3

Burt, C. (1940). *The factors of the mind*. London: University of London Press.

Carroll, J. (1993). *Human cognitive abilities: A survey of factor-analytic studies*. Retrieved from https://books.google.com/books?hl=en&lr=&id=jp9dt4_0_cIC&oi=fnd&pg=PA3&dq=Carroll+higher+order+cognitive+ability&ots=dBCUQdPkR3&sig=11w_WN2WUrtcYQbni9H0F8ZI78U

Cattell, R. B. (1963). Theory of fluid and crystallized intelligence: A critical experiment. *Journal of Educational Psychology, 54*(1), 1.

Cattell, R. B. (1987). *Intelligence: Its structure, growth, and action*. North-Holland. Retrieved from https://books.google.com/books?hl=en&lr=&id=flX770mG2HcC&oi=fnd&pg=PP1&dq=Catell+intelligence+growth,+structure,+action&ots=8WbUnyTDvG&sig=7E5O5M7XplQWUHXrinFn2HaBBbM#v=onepage&q=Catell intelligence growth%2C structure%2C action&f=false.

Chooi, W.-T., Long, H., & Thompson, L. (2014). The Sternberg Triarchic abilities test (level-H) is a measure of g. *Journal of Intelligence, 2*(3), 56–67. https://doi.org/10.3390/jintelligence2030056

Clark, C., Prior, M., & Kinsella, G. (2002). The relationship between executive function abilities, adaptive, behaviour, and academic achievement in children with externalising behaviour problems. *Journal of Child Psychology and Psychiatry and Allied Disciplines, 43*(6), 785–796. https://doi.org/10.1111/1469-7610.00084

Colom, R., Rebollo, I., Palacios, A., Juan-Espinosa, M., & Kyllonen, P. C. (2004). Working memory is (almost) perfectly predicted by g. *Intelligence, 32*(3), 277–296. https://doi.org/10.1016/j.intell.2003.12.002

Coslett, H. B. (2003). Acquired dyslexia. In K. M. Heilman & E. Valenstein (Eds.), *Clinical Neuropsychology* (4th ed., pp. 108–128). New York: Oxford.

Coyle, T. R., Purcell, J. M., Snyder, A. C., & Kochunov, P. (2013). Non-g residuals of the SAT and ACT predict specific abilities. *Intelligence, 41*(2), 114–120. https://doi.org/10.1016/j.intell.2012.12.001

Coyle, T. R., Snyder, A. C., Richmond, M. C., & Little, M. (2015). SAT non-g residuals predict course specific GPAs: Support for investment theory. *Intelligence, 51*, 57–66. https://doi.org/10.1016/j.intell.2015.05.003

Cucina, J., & Byle, K. (2017). The Bifactor model fits better than the higher-order model in more than 90% of comparisons for mental abilities test batteries. *Journal of Intelligence, 5*(3), 27. https://doi.org/10.3390/jintelligence5030027

Deary, I. J. (2000). *Looking down on human intelligence: From psychometrics to the brain* (Oxford Psychology Series). Oxford: Oxford University Press. https://doi.org/10.1093/acprof:oso/9780198524175.001.0001

Deary, I. J. (2012). Intelligence. *Annual Review of Psychology, 63*(1), 453–482. https://doi.org/10.1146/annurev-psych-120710-100353

Deary, I. J., Cox, S. R., & Ritchie, S. J. (2016). Getting Spearman off the skyhook: One more in a century (since Thomson, 1916) of attempts to vanquish g. *Psychological Inquiry, 27*(3), 192–199. https://doi.org/10.1080/1047840X.2016.1186525

Deary, I. J., Der, G., & Ford, G. (2001). Reaction times and intelligence differences: A population-based cohort study. *Intelligence, 29*(5), 389–399. https://doi.org/10.1016/S0160-2896(01)00062-9

Demetriou, A., Spanoudis, G., Shayer, M., Mouyi, A., Kazi, S., & Platsidou, M. (2013). Intelligence cycles in speed-working memory-G relations: Towards a developmental – Differential theory of the mind. *Intelligence, 41*(1), 34–50. https://doi.org/10.1016/j.intell.2012.10.010

Detterman, D. K. (1987). Theoretical notions of intelligence and mental retardation. *American Journal of Mental Deficiency, 92*(1), 2–11. Retrieved from http://psycnet.apa.org/record/1987–31598-001.

Detterman, D. K. (2002). General intelligence: Cognitive and biological explanations. In R. J. Sternberg & E. L. Grigorenko (Eds.), *The general factor of intelligence: How general is it* (pp. 223–243). Mahwah, NJ: Lawrence Erlbaum Associates Publishers.

Detterman, D. K., & Daniel, M. H. (1989). Correlations of mental tests with each other and with cognitive variables are highest for low IQ groups. *Intelligence, 13*(4), 349–359. https://doi.org/10.1016/S0160-2896(89)80007-8

Detterman, D. K., Petersen, E., & Frey, M. C. (2016). Process overlap and system theory: A simulation of, comment on, and integration of Kovacs and Conway. *Psychological Inquiry, 27*(3), 200–204. https://doi.org/10.1080/1047840X.2016.1181514

Doebler, P., & Scheffler, B. (2016). The relationship of choice reaction time variability and intelligence: A meta-analysis. *Learning and Individual Differences, 52*, 157–166. https://doi.org/10.1016/j.lindif.2015.02.009

Duckworth, A. L., Quinn, P. D., Lynam, D. R., Loeber, R., & Stouthamer-Loeber, M. (2011). Role of test motivation in intelligence testing. *Proceedings of the National Academy of Sciences of the United States of America, 108*(19), 7716–7720.

Duncan, J. (2010). The multiple-demand (MD) system of the primate brain: Mental programs for intelligent behaviour. *Trends in Cognitive Sciences, 14*(4), 172–179. https://doi.org/10.1016/j.tics.2010.01.004

Edgin, J. O. (2013). Cognition in down syndrome: A developmental cognitive neuroscience perspective. *Wiley Interdisciplinary Reviews: Cognitive Science, 4*(3), 307–317. https://doi.org/10.1002/wcs.1221

Euler, M. J., McKinney, T. L., Schryver, H. M., & Okabe, H. (2017). ERP correlates of the decision time-IQ relationship: The role of complexity in task- and brain-IQ effects. *Intelligence, 65*, 1–10. https://doi.org/10.1016/j.intell.2017.08.003

Fox, M. C., & Mitchum, A. L. (2012). A knowledge-based theory of rising scores on "culture-free" tests. *Journal of Experimental Psychology: General, 142*(3), 979–1000.

Galton, F. (1883). Inquiries into human faculty and its development. *Inquiries into Human Faculty and Its Development*. Galton: https://doi.org/10.1037/10913-000

Galton, F. (1907). *Inquiries into human faculty and its development* (2nd ed.). London: Dent.

Gardner, H. (2006). *Frames of mind: The theory of multiple intelligences*. New York: Basic Books.

Gignac, G. E. (2014). Dynamic mutualism versus g factor theory: An empirical test. *Intelligence, 42*, 89–97. https://doi.org/10.1016/J.INTELL.2013.11.004

Gignac, G. E. (2016a). Residual group-level factor associations: Possibly negative implications for the mutualism theory of general intelligence. *Intelligence, 55*, 69–78. https://doi.org/10.1016/J.INTELL.2016.01.007

Gignac, G. E. (2016b). The higher-order model imposes a proportionality constraint: That is why the bifactor model tends to fit better. *Intelligence, 55*, 57–68. https://doi.org/10.1016/J.INTELL.2016.01.006

Gottfredson, L. S. (1997a). Mainstream science on intelligence: An editorial with 52 signatories, history, and bibliography. *Intelligence, 24*(1), 13–23. https://doi.org/10.1016/S0160-2896(97)90011-8

Gottfredson, L. S. (1997b). Why g matters: The complexity of everyday life. *Intelligence, 24*(1), 79–132. https://doi.org/10.1016/S0160-2896(97)90014-3

Gottfredson, L. S. (2003). Dissecting practical intelligence theory: Its claims and evidence. *Intelligence, 31*(4), 343–397. https://doi.org/10.1016/S0160-2896(02)00085-5

Grigsby, J. (2016). The fragile X mental retardation 1 gene (FMR1): Historical perspective, phenotypes, mechanism, pathology, and epidemiology. *The Clinical Neuropsychologist, 30*(6), 815–833. https://doi.org/10.1080/13854046.2016.1184652

Gustafsson, J.-E. (1984). A unifying model for the structure of intellectual abilities. *Intelligence, 8*(3), 179–203. https://doi.org/10.1016/0160-2896(84)90008-4

Guttman, L. (1954). A new approach to factor analysis: The Radex. In P. Lazarsfeld (Ed.), *Mathematical thinking in the social science* (pp. 258–348). New York: The Free Press. Retrieved from http://doi.apa.org/psycinfo/1955–02329-001

Haier, R. J. (2016). *The neuroscience of intelligence*. Cambridge: Cambridge University Press.

Hick, W. E. (1952). On the rate of gain of information. *Quarterly Journal of Experimental Psychology, 4*(1), 11–26.

Jauk, E., Benedek, M., Dunst, B., & Neubauer, A. C. (2013). The relationship between intelligence and creativity: New support for the threshold hypothesis by means of empirical breakpoint detection. *Intelligence, 41*(4), 212–221. https://doi.org/10.1016/j.intell.2013.03.003

Jensen, A. R. (1981). Reaction time and intelligence. *Intelligence and Learning*, 39–50.

Jensen, A. R. (1982). Reaction time and psychometric g. *A Model for Intelligence*, 93–132. https://doi.org/10.1007/978-3-642-68664-1_4

Jensen, A. R. (1992). The importance of intraindividual variation in reaction time. *Personality and Individual Differences, 13*(8), 869–881. https://doi.org/10.1016/0191-8869(92)90004-9

Jensen, A. R. (1993). Why is reaction time corelated with psychometric g? *Current Directions in Psychological Science, 2*(2), 53–56.

Jensen, A. R. (1998a). *The g factor: The science of mental ability*. Westport, CT: Praeger.

Jensen, A. R. (1998b). The suppressed relationship between IQ and the reaction time slope parameter of the Hick function. *Intelligence, 26*(l), 43–52.

Johnson, W., & Bouchard, T. J. (2005a). Constructive replication of the visual - perceptual-image rotation model in Thurstone's (1941) battery of 60 tests of mental ability. *Intelligence, 33*(4), 417–430. https://doi.org/10.1016/j.intell.2004.12.001

Johnson, W., & Bouchard, T. J. (2005b). The structure of human intelligence: It is verbal, perceptual, and image rotation (VPR), not fluid and crystallized. *Intelligence, 33*(4), 393–416.

Johnson, W., te Nijenhuis, J., & Bouchard, T. J. (2007). Replication of the hierarchical visual-perceptual-image rotation model in de Wolff and Buiten's (1963) battery of 46 tests of mental ability. *Intelligence, 35*(1), 69–81. https://doi.org/10.1016/j.intell.2006.05.002

Jung, R. E., & Haier, R. J. (2007). The Parieto-frontal integration theory (P-FIT) of intelligence: Converging neuroimaging evidence. *Behav Brain Sci, 30*(2), 135–187. https://doi.org/10.1017/S0140525X07001185

Kan, K. J., Kievit, R. A., Dolan, C., & van der Maas, H. (2011). On the interpretation of the CHC factor Gc. *Intelligence, 39*(5), 292–302.

Kan, K. J., Wicherts, J. M., Dolan, C. V., & van der Maas, H. L. J. (2013). On the nature and nurture of intelligence and specific cognitive abilities: The more heritable, the more culture dependent. *Psychological Science, 24*(12), 2420–2428. Retrieved from http://www.ncbi.nlm.nih.gov/pubmed/24104504

Kane, M. J., & Engle, R. W. (2002). The role of prefrontal cortex in working memory capacity, executive attention, and general fluid intelligence: An individual-differences perspective. *Psychonomic Bulletin & Review, 9*(4), 637–671.

Kane, M. J., & Engle, R. W. (2003). Working-memory capacity and the control of attention: The contributions of goal neglect, response competition, and task set to Stroop interference. *Journal of Experimental Psychology: General, 132*(1), 47–70. https://doi.org/10.1037/0096-3445.132.1.47

Keith, T., Kranzler, J., & Flanagan, D. (2001). What does the cognitive assessment system (CAS) measure? Joint confirmatory factor analysis of the CAS and the Woodcock-Johnson tests of cognitive ability (3rd edition). *School Psychology Review, 30*(1), 89–119.

Kievit, R. A., Lindenberger, U., Goodyer, I. M., Jones, P. B., Fonagy, P., Bullmore, E. T., & Dolan, R. J. (2017). Mutualistic coupling between vocabulary and reasoning supports cognitive development during late adolescence and early adulthood. *Psychological Science, 28*(10), 95679761771078. https://doi.org/10.1177/0956797617710785

Kievit, R. A., Romeijn, J.-W., Waldorp, L. J., Wicherts, J. M., Scholte, H. S., & Borsboom, D. (2011). Mind the gap: A psychometric approach to the reduction problem. *Psychological Inquiry, 22*(2), 67–87. https://doi.org/10.1080/1047840X.2011.550181

Kovacs, K., & Conway, A. R. A. (2016). Process overlap theory: A unified account of the general factor of intelligence. *Psychological Inquiry, 27*(3), 151–177. https://doi.org/10.1080/1047840X.2016.1153946

Kranzler, J. H., Benson, N., & Floyd, R. G. (2015). Using estimated factor scores from a bifactor analysis to examine the unique effects of the latent variables measured by the WAIS-IV on academic achievement. *Psychological Assessment, 27*(4), 1402–1416. https://doi.org/10.1037/pas0000119

Lee, D. G., & Harris, J. C. (2006). *Intellectual disability: Understanding its development, causes, classification, evaluation, and treatment*. New York, NY: Oxford University Press.

Lezak, M. D., Howieson, D. B., Bigler, E. D., & Tranel, D. (2012). *Neuropsychological Assessment* (5th ed.). New York: Oxford University Press.

Lichtenberger, E., & Kaufman, A. (2013). *Essentials of WAIS-IV assessment* (2nd ed.). Hoboken, NJ: John Wiley & Sons Inc. Retrieved from https://books.google.com/books?hl=en&lr=&id=4iHTzkqdQYQC&oi=fnd&pg=PR11&dq=lichtenberger+kaufman+essentials+WAIS-IV&ots=DoAKgKQZo0&sig=ngwhgFRGScK0HrpVyw7OI8-Gp4Y.

Lubinski, D. (2004). Introduction to the special section on cognitive abilities: 100 years after Spearman's (1904) 'general intelligence,' objectively determined and measured. *Journal of Personality and Social Psychology, 86*(1), 96–111. https://doi.org/10.1037/0022-3514.86.1.96

Major, J. T., Johnson, W., & Deary, I. J. (2012). Comparing models of intelligence in project TALENT: The VPR model fits better than the CHC and extended Gf–Gc models. *Intelligence, 40*(6), 543–559. https://doi.org/10.1016/j.intell.2012.07.006

Makris, N., Tachmatzidis, D., Demetriou, A., & Spanoudis, G. (2017). Mapping the evolving core of intelligence: Changing relations between executive control, reasoning, language, and awareness. *Intelligence, 62*, 12–30. https://doi.org/10.1016/J.INTELL.2017.01.006

Marshalek, B., Lohman, D. F., & Snow, R. E. (1983). The complexity continuum in radex and hierarchical models of intelligence. *Intelligence, 4*, 107–127. Retrieved from http://www.sciencedirect.com/science/article/pii/0160289683900235

Matarazzo, J. D. (1972). *Wechsler's measurement and appraisal of adult intelligence* (5th ed.). Oxford: Williams & Wilkins.

McDaniel, M. A., & Whetzel, D. L. (2005). Situational judgment test research: Informing the debate on practical intelligence theory. *Intelligence, 33*(5), 515–525. https://doi.org/10.1016/j.intell.2005.02.001

McGrew, K. S. (2009). CHC theory and the human cognitive abilities project: Standing on the shoulders of the giants of psychometric intelligence research. *Intelligence, 37*(1), 1–10. https://doi.org/10.1016/j.intell.2008.08.004

Melnick, M. D., Harrison, B. R., Park, S., Bennetto, L., & Tadin, D. (2013). A strong interactive link between sensory discriminations and intelligence. *Current Biology, 23*(11), 1013–1017.

Miyake, A., Friedman, N. P., Rettinger, D. A., Shah, P., & Hegarty, M. (2001). How are visuospatial working memory, executive functioning, and spatial abilities related? A latent-variable analysis. *Journal of Experimental Psychology: General, 130*(4), 621–640. https://doi.org/10.1037//0096-3445.130.4.621

Morgan, G., Hodge, K., Wells, K., & Watkins, M. (2015). Are fit indices biased in favor of bi-factor models in cognitive ability research?: A comparison of fit in correlated factors, higher-order, and bi-factor models via Monte Carlo simulations. *Journal of Intelligence, 3*(1), 2–20. https://doi.org/10.3390/jintelligence3010002

Murray, A. L., & Johnson, W. (2013). The limitations of model fit in comparing the bi-factor versus higher-order models of human cognitive ability structure. *Intelligence, 41*(5), 407–422. https://doi.org/10.1016/j.intell.2013.06.004

Naglieri, J. A., & Bornstein, B. T. (2003). Intelligence and achievement: Just how correlated are they? *Journal of Psychoeducational Assessment, 21*, 244–260.

Naglieri, J. A., & Das, J. P. (1990). Planning, attention, simultaneous, and successive (PASS) cognitive processes as a model for intelligence. *Journal of Psychoeducational Assessment, 8*(3), 303–337. https://doi.org/10.1177/073428299000800308

Naglieri, J. A., Goldstein, S., Delauder, B. Y., & Schwebach, A. (2005). Relationships between the WISC-III and the cognitive assessment system with Conners' rating scales and continuous performance tests. *Archives of Clinical Neuropsychology, 20*(3), 385–401. https://doi.org/10.1016/j.acn.2004.09.008

Naglieri, J. A., Goldstein, S., Iseman, J. S., & Schwebach, A. (2003). Performance of children with attention deficit hyperactivity disorder and anxiety/depression on the WISC-III and cognitive assessment system (CAS). *Journal of Psychoeducational Assessment, 21*(1), 32–42. https://doi.org/10.1177/073428290302100103

Newton, J. H., & McGrew, K. S. (2010). Introduction to the special issue: Current research in Cattell-Horn-Carroll-based assessment. *Psychology in the Schools, 47*(7), 621–634. https://doi.org/10.1002/pits.20495

Nisbett, R. E., Aronson, J., Blair, C., Dickens, W., Flynn, J., Halpern, D. F., & Turkheimer, E. (2012). Intelligence: New findings and theoretical developments. *American Psychologist, 67*(2), 130–159.

Oberauer, K., Schulze, R., Wilhelm, O., & Süß, H. (2005). Working memory and intelligence--their correlation and their relation: Comment on Ackerman, Beier, and Boyle. Retrieved from http://psycnet.apa.org/record/2004-22408-003

Papathanassiou, D., Etard, O., Mellet, E., Zago, L., Mazoyer, B., & Tzourio-Mazoyer, N. (2000). A common language network for comprehension and production: A contribution to the definition of language epicenters with PET. *NeuroImage, 11*(4), 347–357. https://doi.org/10.1006/nimg.2000.0546

Protzko, J. (2017). Effects of cognitive training on the structure of intelligence. *Psychonomic Bulletin & Review, 24*(4), 1022–1031. https://doi.org/10.3758/s13423-016-1196-1

Rammsayer, T. H., & Troche, S. (2016). Validity of the worst performance rule as a function of task complexity and psychometric g: On the crucial role of g saturation. *Journal of Intelligence, 4*(1), 5. Retrieved from http://www.mdpi.com/2079–3200/4/1/5/htm

Ratcliff, R., Schmiedek, F., & McKoon, G. (2008). A diffusion model explanation of the worst performance rule for reaction time and IQ. *Intelligence, 36*(1), 10–17. https://doi.org/10.1016/j.intell.2006.12.002

Raven, J. C., & Court, J. H. (1998). *Raven's progressive matrices and vocabulary scales*. Oxford: Oxford Psychologists Press.

Redick, T. S., Shipstead, Z., Harrison, T. L., Hicks, K. L., Fried, D. E., Hambrick, D. Z., ... & Engle, R. W. (2013). No evidence of intelligence improvement after working memory training: a randomized, placebo-controlled study. *Journal of Experimental Psychology: General, 142*(2), 359. https://doi.org/10.1037/a0029082

Salthouse, T. A., & Davis, H. P. (2006). Organization of cognitive abilities and neuropsychological variables across the lifespan. *Developmental Review, 26*(1), 31–54. https://doi.org/10.1016/j.dr.2005.09.001

Sattler, J. M. (2008). *Assessment of children: Cognitive foundations* (5th ed.). San Diego, CA: Jerome M Sattler Publisher, Inc..

Schmiedek, F., Oberauer, K., Wilhelm, O., Süß, H.-M., & Wittmann, W. W. (2007). Individual differences in components of reaction time distributions and their relations to working memory and intelligence. *Journal of Experimental Psychology: General, 136*(3), 414–429. https://doi.org/10.1037/0096-3445.136.3.414

Schneider, J., & McGrew, K. S. (2012). The Cattell-Horn-Carroll (CHC) Model of Intelligence. In *Contemporary intellectual assessment: Theories, tests, and issues* (3rd ed.). https://doi.org/10.3233/978-1-60750-588-4-1344

Schubert, A.-L., Hagemann, D., & Frischkorn, G. T. (2017). Is general intelligence little more than the speed of higher-order processing? *Journal of Experimental Psychology: General, 146*(10), 1498–1512. https://doi.org/10.1037/xge0000325

Sheppard, L. D., & Vernon, P. A. (2008). Intelligence and speed of information-processing: A review of 50 years of research. *Personality and Individual Differences, 44*(3), 535–551. https://doi.org/10.1016/j.paid.2007.09.015

Spearman, C. (1904). "General intelligence," objectively determined and measured. *The American Journal of Psychology, 15*(2), 201–292. https://doi.org/10.2307/1412107

Spearman, C. (1927). The abilities of man. Retrieved from http://doi.apa.org/psycinfo/1927-01860-000

Stankov, L. (2017). Overemphasized "g". Journal of Intelligence, 5(33), 1–10. Retrieved from http://www.mdpi.com/2079-3200/5/4/33

Stankov, L., & Crawford, J. D. (1993). Ingredients of complexity in fluid intelligence. *Learning and Individual Differences, 5*(2), 73–111.

Stankov, L., & Raykov, T. (1995). Modeling complexity and difficulty in measures of fluid intelligence. *Structural Equation Modeling, 2*(4), 335–366.

Stankov, L., & Roberts, R. D. (1997). Mental speed is not the basic process of intelligence. *Personality and Individual Differences, 22*(1), 69–84.

Sternberg, R. J. (1999). The theory of successful intelligence. *Review of General Psychology, 3*(4), 292–316. https://doi.org/10.1037/1089-2680.3.4.292

Sternberg, R. J. (2012). The Triarchic theory of successful intelligence. In D. Flanagan & P. Harrison (Eds.), *Contemporary intellectual assessment* (3rd ed., pp. 156–177). New York, NY: Guidlford Press.

Suchy, Y. (2015). *Executive Functioning*.

Thurstone, L. (1938). *Primary mental abilities*. Chicago, IL: University of Chicago Press. Retrieved from http://psycnet.apa.org/record/1938-15070-000.

Thurstone, L. (1947). *Multiple factor analysis*. Chicago, IL: University of Chicago Press. Retrieved from http://doi.apa.org/psycinfo/1947-15068-000

Tucker-Drob, E. M. (2009). Differentiation of cognitive abilities across the life span. *Developmental Psychology, 45*(4), 1097–1118. Retrieved from http://www.pubmedcentral.nih.gov/articlerender.fcgi?artid=2855504&tool=pmcentrez&rendertype=abstract

van de Vijver, F., & Tanzer, N. K. (2004). Bias and equivalence in cross-cultural assessment: An overview. *Revue Europeenne de Psychologie Appliquee, 54*(2), 119–135. https://doi.org/10.1016/j.erap.2003.12.004

van der Maas, H. L. J., Dolan, C. V., Grasman, R. P. P. P., Wicherts, J. M., Huizenga, H. M., & Raijmakers, M. E. J. (2006). A dynamical model of general intelligence: The positive manifold of intelligence by mutualism. *Psychological Review, 113*(4), 842.

van der Maas, H. L. J., & Kan, K. J. (2016). Comment on "residual group-level factor associations: Possibly negative implications for the mutualism theory of general intelligence" by Gilles E. Gignac (2016). *Intelligence, 57*, 81–83. https://doi.org/10.1016/j.intell.2016.03.008

van der Maas, H. L. J., Kan, K.-J., & Borsboom, D. (2014). Intelligence is what the intelligence test measures. Seriously. *Journal of Intelligence, 2*(1), 12–15. https://doi.org/10.3390/jintelligence2010012

Vernon, P. (2014). *The structure of human abilities (psychology revivals)*. Retrieved from https://books.google.com/books?hl=en&lr=&id=i8y2AgAAQBAJ&oi=fnd&pg=PP1&dq=Phillip+vernon+abilities&ots=NdTRZ5TxVT&sig=4As0OIfnlDicZ_7v-HliNK1dw1A

Visser, B. A., Ashton, M. C., & Vernon, P. A. (2006). Beyond g: Putting multiple intelligences theory to the test. *Intelligence, 34*(5), 487–502. https://doi.org/10.1016/j.intell.2006.02.004

Warne, R. T., Astle, M. C., & Hill, J. C. (2018). What do undergraduates learn about human intelligence? An analysis of introductory psychology textbooks. *Archives of Scientific Psychology, 6*, 32–50. https://doi.org/10.1037/arc0000038

Wasserman, J. (2012). A history of intelligence assessment: The unfinished tapestry. In D. Flanagan & P. L. Harrison (Eds.), *Contemporary intellectual assessment: Theories, tests, and issues* (3rd ed., pp. 3–55). New York: Guilford Press.

Wechsler, D. (1943). Non-intellective factors in general intelligence. *The Journal of Abnormal and Social Psychology, 38*, 101. Retrieved from.

Wechsler, D. (2008). *Wechsler adult intelligence scale-4th edition: Technical and interpretative manual*. San Antonio, TX: Psychological Corporation.

Wechsler, D., & Edwards, A. J. (1974). *Selected papers of David Wechsler*. (A. Edwards, Ed.).

Woodcock, R. W. (1990). Theoretical foundations of the WJ-R measures of cognitive ability. *Journal of Psychoeducational Assessment, 8*(3), 231–258. https://doi.org/10.1177/073428299000800303

Woodcock, R. W., McGrew, K. S., & Mather, N. (2001). *Woodcock-Johnson III tests of cognitive abilities. Test*. Itasca, IL: Riverside.

Definition and Diagnosis

3

Paige Cervantes, Rebecca Shalev,
and Lauren Donnelly

Definition and Diagnosis

Intellectual disability (ID) has been an evolving construct beginning early in history and has been generally defined by significant, lifelong impairments in cognition and daily functioning. While intellectual and adaptive behavior deficits are essential in the diagnosis of ID, the presentation of ID can vary widely between individuals in relation to the degree of impairment and in distinct strengths and weaknesses. Further, the ID population, across severity levels, has an increased prevalence of a range of comorbid medical and psychiatric disorders. Therefore, ID is a highly heterogeneous condition that requires thoughtful, comprehensive, and individualized assessment across the life course. Providing appropriate diagnostic and evaluation services is essential for informing treatment planning and ensuring individuals with ID can optimize their prognosis and reach their potential.

This chapter will briefly review the evolution of the definition and conceptualization of ID. In addition, a detailed discussion will be presented regarding the current definition and how to interpret this definition in the context of the diagnostic process. A brief review of specific measurement techniques and how they are used in concert will also be provided. Further, a developmental perspective will be used to explain the differences in the diagnostic process across age groups based on severity level of the disability.

The Evolving Definition of Intellectual Disability

Since antiquity, people have conceptualized and labeled ID in many different ways. The field has made many important advances in policy and research over the course of decades that have led to our current understanding of the definition of ID and to support needs related to ID. However, this attitude of inclusion and intervention has not always been the norm. Individuals with ID have historically been subjected to significant discrimination and injustices (Witwer, Lawton, & Aman, 2014). For example, during ancient times, ID was thought to result from possession by a supernatural being, and those with ID were mistreated and subjected to exorcism and even death. Other early theorists believed that ID was the product of sin and evil or divine visitation (Cardona, 1994), and again treatment of ID fell in line with this conceptualization. Fortunately, in the eighteenth century, Jean Marie Gaspar

P. Cervantes (✉) · R. Shalev · L. Donnelly
Autism Spectrum Disorder Research and Clinical Program, Child Study Center at Hassenfeld Children's Hospital at NYU Langone,
Department of Child and Adolescent Psychiatry,
New York, NY, USA
e-mail: paige.cervantes@nyumc.org

J. L. Matson (ed.), *Handbook of Intellectual Disabilities*, Autism and Child Psychopathology Series, https://doi.org/10.1007/978-3-030-20843-1_3

Itard showed that people with ID were able to learn and many improvements in the field followed. For instance, in the nineteenth century, Samuel Howe followed up on this critical discovery by founding the first school for children with ID in 1848 (Cardona, 1994).

Improvements in our knowledge regarding etiology of ID have also contributed to advances in the provision of services. Historically, the definition of ID focused on inadequate social behavior (Doll, 1941). However, medical advancements in the twentieth century most notably resulted in the scientific discovery that ID is hereditary. Following this discovery, ID came to be seen as a medical construct (Devlieger, 2003). The conceptualization of ID changed once more as the intelligence testing movement took hold: with the advent of valid and reliable intelligence measures, the emphasis shifted to an individual's IQ score, which was compared to a normative sample (Devlieger, 2003; Schalock, Luckasson, & Shogren, 2007).

The nomenclature has also evolved. In the late 1800s and early 1900s, people with ID were labeled feebleminded, imbeciles, and idiots (Cardona, 1994). These terms had technical definitions; however, they quickly took on derogatory meanings. In the mid- and late 1900s, the terms mental deficiency and then mental retardation were used to classify individuals with ID (Cardona, 1994; Greenspan & Switzky, 2003). It was not until the introduction of the *DSM-5* that the term mental retardation was formally changed to intellectual disability (Schalock et al., 2007, 2010). ID was chosen to replace mental retardation for several reasons. The term ID better aligned with the new construct of disability from the World Health Organization (WHO) and the American Association on Intellectual and Developmental Disabilities (AAIDD). This new conceptualization emphasized a move away from a person-centered definition of innate deficit to defining functional limitations based on a person-environment interaction. Relatedly, the term ID matched up with current clinical practice that focuses on functional behaviors and individualized, environmental supports necessary to improve a person's functioning. Finally, in addition to addressing the negative connotation associated with the term mental retardation, the new terminology improved consistency with terminology used internationally (Schalock et al., 2007).

Despite the evolving vocabulary used to classify ID and the associated advancements in research, policy, and clinical practice in recent decades, the construct of ID has remained largely the same. Three essential elements have been retained and include (1) deficits in cognitive functioning, (2) limitations in adaptive behavior, and (3) an early onset of symptoms (Schalock et al., 2007).

Diagnostic and Statistical Manual (DSM)

ID was termed mental deficiency in the first edition of the *Diagnostic and Statistical Manual* (*DSM-I;* American Psychiatric Association [APA], 1952) and was used to classify individuals who presented with "a defect of intelligence existing since birth, without demonstrated organic brain disease or known prenatal cause." Chronic brain syndrome with mental deficiency was diagnosed when the etiology of intellectual deficits was known. In the *DSM-I*, IQ estimates were provided to correspond with severity levels ranging from mild to severe; however, an emphasis was placed on functional behaviors as well, noting that severity level should be informed by both cultural, physical, and emotional factors as well as occupational, educational, and social success (APA, 1952).

In the *DSM-II*, ID was called mental retardation and was defined as "subnormal general intellectual functioning, which originates during the developmental period" (APA, 1968). Severity levels within the *DSM-II* included borderline, mild, moderate, severe, and profound mental retardation. As in the *DSM-I*, discrete IQ cutoffs were provided for determining severity level, but the need to evaluate adaptive behavior in conjunction with IQ was stated as well. Clinical subcategories were also offered to detail any known etiological information related to an individual's intellectual deficits. Unspecified mental retardation became available in the *DSM-II* to diagnose individuals who could not participate in a formal assessment of IQ but were clinically "subnormal" (APA, 1968).

The *DSM-III* (APA, 1980) emphasized the dual-criterion approach to a greater degree than in previous editions; the *DSM-III* definition hinged upon significantly "subaverage" intellectual functioning in addition to deficits or impairments in adaptive behavior. IQ estimates corresponding to severity levels continued to be present; however, the authors of the *DSM-III* highlighted the error of measurement and encouraged clinicians to treat IQ scores with "some flexibility." Adaptive behavior in the *DSM-III* was defined as the "effectiveness with which an individual meets the standards of personal independence and social responsibility expected for his or her age and cultural group." As tools for standardized measurement were limited at this time, the authors encouraged the use of clinical judgment in evaluating adaptive functioning. Further, symptom onset had to occur prior to 18 years old. Of note, borderline mental retardation was removed, and "borderline intellectual functioning" was added as a "V" code within the *DSM-III*. The definition of ID remained largely the same in the 1987 *DSM-III-Revision* (APA, 1987).

The criteria for diagnosis in the *DSM-IV* (APA, 1994) and *DSM-IV-Text Revision* (APA, 2000) were also similar (APA, 1994). Again, discrete IQ cutoffs were provided to determine an individual's severity level (i.e., mild, moderate, severe, and profound). Though, criteria for adaptive functioning was clarified to specify limitations in at least two of the following areas: communication, self-care, home living, social/interpersonal skills, use of community resources, self-direction, functional academic skills, work, leisure, health, and safety (APA, 1994, 2000). Notably, while mentioned in previous editions, a greater emphasis was placed on psychiatric comorbidity within the *DSM-IV* and *DSM-IV-TR* conceptualization. The increased vulnerability for mental health disorders and the complexity in assessing for these disorders were clearly stated (APA, 2000).

The DSM-5 Definition of ID

The *DSM-5* (APA, 2013) presented several changes to the previous conceptualization of ID, including but not limited to the change in terminology from mental retardation, and provided comprehensive guidance for diagnosing ID. Following revisions to the *DSM-IV-TR* definition, the *DSM-5* classification is also more equivalent to the *International Classification of Disease*, *11th Revision*'s (*ICD-11*) diagnosis of intellectual developmental disorder (APA, 2013). Within the *DSM-5*, ID is presented alongside other neurodevelopmental disorders, emphasizing its enduring nature and association with early brain dysfunction. The core diagnostic features of ID remained the same and include deficits in two domains—intellectual functions and adaptive skills—relative to age, gender, and sociocultural expectations, which are based on clinical judgment and standardized assessment results.

In the *DSM-5*, the term intellectual functions refers to reasoning abilities, problem-solving skills, planning, abstract thinking, judgment, practical understanding, and learning. Deficits in adaptive functioning are characterized by difficulties performing skills of daily living in a variety of conceptual (academic), social, and practical areas, across multiple contexts (i.e., home, school, vocational, and community settings). Impairments in intellectual and adaptive functioning must manifest during the developmental period; this is a change from previous editions of the *DSM* where an age of onset prior to 18 years old was explicitly required (APA, 2013).

One of the most notable changes in the *DSM-5* was the removal of IQ scores from the diagnostic criteria. Individuals are instead diagnosed with mild, moderate, severe, or profound ID based upon adaptive functioning (APA, 2013). Since severity classifications are used to communicate the level of support an individual requires, and adaptive functioning refers to skills in practical contexts, the *DSM-5* approach better identifies treatment needs and therefore has greater clinical utility. This paradigm shift also reduces an over-reliance on IQ scores, which is critical in light of research that suggests IQ measures are less valid in the lower end of the IQ range (Hessl et al., 2009). The APA also notes that an IQ score alone may not reflect skills for reasoning in real-life situations nor is a single IQ score useful in understanding intellectual abilities compared to comprehensive neuropsychological profiles (APA, 2013). However, the APA is clear that IQ tests should continue to be administered in the assessment of intellectual functioning. Further, the upper limit of an IQ of approximately 70 remains (APA, 2013).

According to current estimates, ID occurs in 1% of the population (Witwer et al., 2014). However, prevalence differs as a function of severity level. Mild ID is the most prevalent, encompassing approximately 85% of the ID population (van Bokhoven, 2011). Further, ID occurs in both males and females across sociodemographic backgrounds. However, the numbers are higher in males and in lower-income populations (Maulik, Mascarenhas, Mathers, Dua, & Saxena, 2011; Witwer et al., 2014). Gender differences are likely a result of sex-linked genetic factors (Harris, 2006), and the increased prevalence in low-income populations may be due to several factors, including heightened risk for poor prenatal care, malnutrition, and exposure to illness, injury, toxins, or environments with reduced stimulation (Matson & Cervantes, 2019; Maulik et al., 2011).

While etiology of mild ID is often unclear, individuals with more severe impairments (e.g., moderate to profound ID) are more likely to have an identifiable, organic cause, like a genetic disorder or significant pre-, peri-, or postnatal complications (Witwer et al., 2014). Prenatal screening can be done to identify if a genetic disorder associated with ID is present (de Jong, Dondorp, Frints, de Die-Smulders, & de Wert, 2011; Genetic Alliance, 2009). Examples of genetic disorders associated with ID include Down syndrome, Prader-Willi syndrome, Angelman syndrome, Williams syndrome, and Fragile X syndrome. When a genetic disorder is responsible for intellectual impairment, the individual may also present with dysmorphic features at birth. For example, newborns with Down syndrome present with brachycephaly, which is characterized by shortened, anteroposterior dimension of the head (Wechsler & McDonald, 2018). Other genetic disorders may become evident during infancy and early childhood. For instance, Lesch-Nyhan syndrome is associated with a distinct behavioral phenotype, including delayed motor development evident at 3–6 months of age and persistent self-injurious behavior during toddlerhood (Harris, 2010; Nyhan, O'Neill, Jinnah, & Harris, 2014). ID can be acquired prenatally through factors such as inadequate prenatal care, maternal infections during pregnancy, and maternal alcohol or substance use during pregnancy. Perinatal causes can include infections, intracranial hemorrhage, and hypoglycemia, and ID may be acquired postnatally through events like meningitis and encephalitis from bacterial or viral infections, significant psychosocial trauma (e.g., abuse, neglect, and malnutrition), exposure to environmental toxins (e.g., lead), and head injury (Harris, 2006; Shevell, 2008).

In terms of age of onset, diagnosis often occurs at different times based upon severity level. Children with mild ID may not be recognized until school years when increased academic and social demands make symptoms more evident, whereas children with moderate ID are typically diagnosed in the preschool years, and children with severe and profound ID are typically identified before or shortly after birth (Witwer et al., 2014). Often though, individuals with ID experience some degree of apparent developmental delays in infancy and early childhood. Generally, one or more of the following early symptoms are present: motor delays (e.g., sitting, crawling, or walking later than same-age peers), language delays (e.g., talking later than expected or having trouble speaking), memory deficiencies, problem-solving weaknesses, and difficulties understanding the results of their actions (Centers for Disease Control and Prevention [CDC], 2017).

For these young children who present with clear developmental difficulties but are too young to participate in structured testing, a diagnosis of global developmental delay (GDD) may be warranted. GDD is diagnosed in children under

5 years old when significant deficits are present across two or more developmental domains (e.g., motor skills, speech/language, social skills, activities of daily living, cognition; APA, 2013; Moeschler, Shevell,, & Committee on Genetics, 2014). As IQ is not considered stable in early childhood (Ozonoff, Goodlin-Jones, & Solomon, 2005), GDD is an appropriate label for young children until valid and reliable IQ testing can be conducted and a diagnosis of ID, or a different neurodevelopmental disorder, can be confirmed (Moeschler et al., 2014). A diagnosis of GDD highlights the need for reassessment after a period of time (APA, 2013) and predicts later diagnosis of ID (Shevell, 2008). When an individual is over the age of 5 but cannot participate in structured testing due to sensory or physical impairments, severe problem behaviors, or comorbid psychopathology, a diagnosis of unspecified ID should be provided until data from a reassessment can confirm a more specific diagnosis (APA, 2013).

The Diagnosis of DSM-5 ID

Assessment for ID focuses on measurement in the two primary domains discussed above: intellectual functioning and adaptive behavior. It is imperative to measure both intellectual and adaptive domains in a systematic and objective manner in order to generate a complete picture of an individual's strengths and weaknesses. Further, obtaining information regarding how impairments manifested across the life span is necessary for the diagnosis of ID.

As with all diagnostic evaluations, a multimodal assessment process incorporating clinical interviews, behavioral observations, and standardized measures should be used. It may also be helpful to obtain educational and medical records, especially if a diagnosis of ID is being made in adolescence or adulthood. The evaluation should begin with a comprehensive interview of developmental history and current functioning. Such an interview should be conducted with an individual's primary caregiver(s) and inquire about medical, psychiatric, developmental, behavioral, and educational realms. Within the medical history, given the known etiological bases for ID, history of prenatal, perinatal, and postnatal complications should be obtained. Specifically, it is necessary to assess for possible maternal substance use and exposure to alcohol, smoke, and illicit drugs in utero, as well as exposure to toxins. Information should be collected regarding the pregnancy and birth, including gestational age, birth weight, and APGAR scores if available, any history of head trauma, injuries, and illnesses, and timing of early developmental milestones. It is also necessary to obtain a family history of developmental disorders (DDs), ID, and other medical and psychiatric conditions (McDonald, 2006; Shevell, 2008; Tirosh & Jaffe, 2011). After attaining a comprehensive history, current behaviors, including cognitive, communication, social, adaptive, and academic functioning, should also be evaluated through interview, behavioral observations, and standardized measures.

As previously stated, the diagnosis of GDD is typically considered for children under 5 years old instead of a diagnosis of ID. The diagnostic process for GDD should not be dissimilar to that described above for ID. However, the tools used for the assessment of GDD differ from those used in the evaluation for ID and will be addressed below. Notably, in more severe cases of intellectual and adaptive impairment, a diagnosis of ID may be warranted prior to age of 5 years old. This may be particularly true for young children with genetic disorders associated with ID.

Evaluation of Intellectual Abilities

Intellectual ability is measured using standardized cognitive assessments, resulting in an IQ score. IQ was originally defined as a ratio of one's mental age to their chronological age, but the ratio IQ has since been replaced by a more complicated deviation IQ (Brue & Wilmshurst, 2016). Previous recommendations encouraged the use of a composite or total test score to be compared to a cutoff score (two standard deviations below the mean, i.e., an IQ of 70) in examining symptom threshold (National Research Council [NRC], 2002); however, the *DSM-5* conceptualization warns against overreliance on an IQ score. Instead, an individual's complete cognitive profile, as measured by a reliable and valid IQ test, should be used as one piece of evidence in the diagnosis of ID.

Cognitive measures typically assess a variety of areas including verbal knowledge, spatial skills, processing/psychomotor speed, memory, and verbal and nonverbal reasoning abilities (Sattler, 2008). Several cognitive assessments are available, and important considerations should be made in choosing the optimal tool for the individual being assessed. These include patient age, developmental level, culture, language, communication abilities, and environment (Schalock & Luckasson, 2004). In addition, clinicians assessing for ID should be experienced in working with children with DDs, as the application of strategies to improve patient motivation and on-task behavior may be necessary to achieve an accurate estimation of ability (Ozonoff et al., 2005).

Assessment tools

Measurement techniques vary as a function of one's age and severity of symptoms. Therefore, examples of assessment tools will be presented based upon their utility across different developmental periods.

Early childhood assessment

In young children, clinicians may choose to obtain a developmental quotient (DQ) instead of an IQ score. A DQ score is a ratio of one's developmental age to chronological age. As IQ is unstable in early childhood, using a DQ to quantify strengths and weaknesses related to developmental milestones may be more precise and clinically useful in young childhood (Matson, Mahan, Hess, & Fodstad, 2010). Therefore, assessment of cognitive abilities should occur in relation to developmental milestones using developmental inventories. The *Bayley Scales of Infant and Toddler Development-Third Edition* (*Bayley-3*; Bayley, 2006) is one example of a developmental measure. The *Bayley-3* is designed for use with children 1–42 months of age and examines developmental functioning across five domains. The cognitive, language, and motor skill domains are administered to the child and are directly assessed by the clinician; social-emotional and adaptive skill domains are obtained through informant-report. Other developmental measures include the *Mullen Scales of Early Learning* (*Mullen*; Mullen, 1995) and the *Battelle Developmental Inventory-Second Edition* (*BDI-2*; Newborg, 2005).

Measures of IQ designed to assess young children are also available. These are typically reserved for use with young children who are closer to 5 years old and less impaired. In these cases, the *Differential Abilities Scale, Second Edition-Early Years Battery* (*DAS-2*; Elliott, 2007) or the *Wechsler Preschool and Primary Scale of Intelligence-Fourth Edition* (*WPPSI-IV*; Wechsler, 2012) can be administered to obtain an estimate of intellectual functioning.

School-aged to adulthood

There are many cognitive tools available for children and adults with verbal language. The Wechsler scales are one of the most widely used. The *Wechsler Intelligence Scale for Children-Fifth Edition* (*WISC-V*; Wechsler, 2014) is designed for use with children aged 6–16 years old, and the *Wechsler Adult Intelligence Scale-Fourth Edition* (*WAIS-IV;* Wechsler, 2008) is used for individuals 16 years of age and older. The Wechsler scales, however, have a high verbal requirement and therefore are generally administered in the assessment of individuals with a high degree of spoken language (Matson & Cervantes, 2019; Ozonoff et al., 2005).

In addition, the *Stanford-Binet Intelligence Scales-Fifth Edition* (*SB5*; Roid, 2003) is routinely used to assess cognitive abilities within evaluations for ID. The *SB5* is designed for use with individuals 2 years of age and older and results in a Full Scale IQ (FSIQ), nonverbal and verbal IQ scores, and five index scores. Based on assessment needs, the clinician can administer solely the verbal or nonverbal IQ domain. If available assessment time is short, an Abbreviated Battery IQ (ABIQ) can also be obtained from the *SB5* in approximately 15 min with the use of two subtests (Roid, 2003). General consensus indicates that the *SB5* is able to assess a wide range of abilities, allowing one to obtain a greater differential at the lower ends of the cognitive spectrum (Silverman et al., 2010). Therefore, the *SB5* may be preferred in the assessment for ID in individuals across severity levels.

The *DAS-II School-Age Battery* (Elliott, 2007) offers another option for the assessment of children with greater impairments, including language difficulties. This measure is designed for use with children 7–17 years old and assesses verbal, spatial, and nonverbal reasoning abilities. Because there is a degree of fluidity across the early years and school-age batteries, the *DAS-II* offers clinicians the flexibility to administer subtests originally designed for younger children if a child is unable to complete a subtest that is meant for their chronological age range. While no data were found comparing the *DAS-II* with the *SB5*, researchers have shown that the *DAS-II* resulted in higher scores when compared to the *Wechsler Intelligence Scale for Children-Fourth Edition* (*WISC-IV*) in a population of children with autism spectrum disorder (ASD). This difference was hypothesized to be due to the inclusion of processing speed on the *WISC-IV* (Kuriakose, 2014).

Measures with minimal verbal requirement

Due to substantial impairments in communication, more traditional tools may not be effective in the assessment of individuals with severe to profound ID. In these cases, the *Leiter International Performance Scale-Third Edition* (*Leiter-3*; Roid, Miller, Pomplun, & Koch, 2013), the *Test of Nonverbal Intelligence-Fourth Edition* (*TONI-4;* Brown, Sherbenou, & Johnsen, 2010), or the *Comprehensive Test of Nonverbal Intelligence-Second Edition* (*CTONI-2*; Hammill, Pearson, & Weiderholt, 2009) may be utilized. These nonverbal assessments are administered without verbal instructions and do not require verbal responses, which allows clinicians to assess nonverbal cognitive abilities in individuals with minimal to no language. Further, developmental measures, such as the *Mullen*, may also be utilized to evaluate the abilities of individuals with severe impairments. When administering developmental measures out of the age range of the standardized administration, age equivalents may be more informative than the resulting IQ score (Bishop, Guthrie, Coffing, & Lord, 2011).

Evaluation of Adaptive Skills

Adaptive behavior deficits across conceptual, social, and practical domains should also be measured using standardized, norm-referenced assessments. Standardized measures provide the age-referenced results necessary for evaluating an individual's degree of impairment against what is expected (Cervantes & Jang, 2016; Schalock & Luckasson, 2004). In regard to evaluating whether an individual meets the adaptive behavior criteria for diagnosis, the *DSM-5* stipulates that an individual must demonstrate difficulties in at least one of the three domains, to the extent that ongoing support is required in at least one setting (APA, 2013).

Assessment tools

Typically, reports of adaptive skills are given by caregivers. However, it can be helpful to obtain information regarding adaptive functioning from multiple sources, including employers, job coaches, teachers, and, in some cases, the individual being assessed, in order to generate a wide-ranging assessment of strengths and weaknesses.

Vineland Adaptive Behavior Scales, Third Edition (Vineland-3)

The *Vineland Adaptive Behavior Scales-Third Edition* (*Vineland – 3;* Sparrow, Cicchetti, & Saulnier, 2016) is one of the most popular and well-known assessments of adaptive functioning. The *Vineland-3* is available in several formats, including interviews and rating scales to be completed by caregivers and teachers. The measure is intended for use with individuals across the life-span. The instrument assesses an individual's adaptive skills across five domains (i.e., communication, daily living skills, socialization, motor skills, maladaptive behavior) depending on the individual's age and evaluation needs. Subdomains exist within each of the larger domains to better define an individual's strengths and weaknesses. The *Vineland-3* also results in an adaptive behavior composite score, as well as standard scores for each domain and subdomain, allowing for comparison of scores across a population.

Adaptive Behavior Assessment System, Third Edition (ABAS-3)

The *Adaptive Behavior Assessment System-Third Edition* (*ABAS-3*; Harrison & Oakland, 2015) is another widely used assessment of adaptive functioning. The *ABAS-3* has five rating forms available to be used across the life-span and to be completed by caregivers, by teachers, or by the individual being assessed. Parent and teacher forms are available for the assessment of individuals 0–21 years old. The Adult form can be administered as self-report for individuals over 16 years old. This self-assessment provides adults with ID autonomy in influencing treatment goals and allows caregivers and service providers to support individuals in generating and implementing self-directed plans. The *ABAS-3* results in scores across conceptual, social, and practical domains, through the assessment of nine or ten adaptive skill areas based upon the individual's age.

Additional Considerations

Because symptoms of ID affect multiple domains of functioning, multidisciplinary assessment is essential in developing appropriate interventions. If an interdisciplinary approach is unavailable during the initial assessment and based upon the results of the evaluation, individuals with ID should be referred to professionals within the speech and language, medical, and occupational and physical therapy fields for further evaluation and support.

Additionally, once a diagnosis is made, medical and genetic testing is often recommended to determine if there is a known etiology of the ID, such as a genetic disorder. Further investigation into the presence of a genetic disorder is especially indicated in the case that there is a family history of genetic disorders and/or a physical examination identifies signs related to genetic disorders (e.g., dysmorphic features, aberrant motor development). This is important because identification of a known etiology offers an individual and their family more detailed prognostic information and more specific supports and services. However, the etiology of many cases of ID, particularly more mild cases, cannot be discerned, and genetic testing is both financially and labor intensive. Therefore, practitioners should be mindful of this when referring families for follow-up evaluation (de Jong et al., 2011; Genetic Alliance, 2009; Matson & Cervantes, 2019).

In addition, even when a diagnosis of ID is made early in life, re-evaluations throughout the life span are necessary for the determination of educational classification and support services, program planning, eligibility for benefits and legal protections, and reimbursement/funding. As limitations related to ID are defined by person-environment interactions, an individual's support needs will change across the life course and as the context in which they are functioning changes.

Dual Diagnosis

In addition to intellectual and adaptive skill deficits and although not necessary for diagnosis, many individuals with ID experience comorbid health and psychiatric conditions as well as a variety of challenging behaviors (e.g., tantrums, aggression, self-injury). Prognosis is typically poorer when comorbidities are present (Matson & Shoemaker, 2011); therefore, comprehensive assessment and targeted treatment of an individual's complete clinical picture is imperative. However, several challenges are present when addressing medical and psychiatric symptoms within the ID population (Tonge, Torr, & Trollor, 2017).

Comorbid Medical Disorders

Individuals with ID commonly present with a range of medical conditions (van Schrojenstein Lantman-de Valk et al., 1997; van Schrojenstein

Lantman-de Valk, Metsemakers, Haveman, & Crebolder, 2000). For instance, hearing and visual impairments, motor and seizure disorders, congenital abnormalities related to areas like cardiac and gastrointestinal functioning, and metabolic, endocrine, and immune system disorders occur more often in individuals with ID (Tonge et al., 2017; van Schrojenstein Lantman-de Valk et al., 1997). Of note, medical comorbidities frequently relate to the etiology of ID. As previously mentioned, ID is often associated with various genetic disorders, and each disorder has related medical problems. For example, individuals with Down syndrome often experience hypothyroidism, hearing loss, and early-onset dementia, and cleft palate, congenital heart disease, and immune system disorders are prevalent in individuals with DiGeorge syndrome (Tonge et al., 2017). The presence of medical conditions is also associated with severity of ID and age; individuals with more severe ID and who are older adults (i.e., 50 years or older) are at higher risk for medical difficulties (van Schrojenstein Lantman-de Valk et al., 1997).

Individuals with ID also experience general health problems more often than typically developing peers, such as obesity, sleep problems, pneumonia, seizures, and gastroesophageal reflux (Didden, Korzilius, van Aperlo, van Overloop, & de Vries, 2002; Tonge et al., 2017; van Schrojenstein Lantman-de Valk et al., 2000). Age-related conditions also occur more frequently in adults with ID; for example, dementia has a higher incidence and an earlier onset for adults with ID than for the general population (Tonge et al., 2017). Though these conditions occur commonly in the general population, their identification and treatment in individuals with ID is complex, thus contributing to the high rates of unmet service needs experienced by the ID population (Peterson-Besse et al., 2014; Tonge et al., 2017).

Assessment of physical symptoms in individuals with ID is particularly difficult due to the communication impairments inherent to ID. Medical professionals often have to rely on behavioral indices of pain rather than verbal report of symptoms. Pain may be communicated through an increase in challenging behaviors (e.g., self-injury, repetitive behavior, noncompliance) or other indirect cues. For instance, behavioral markers for abdominal pain in individuals with ASD and communication impairments include crying with no clear cause, frequent swallowing, facial grimacing, gritting teeth, wincing, unusual posturing, and sleep problems (Buie et al., 2010; Tonge et al., 2017). Symptom presentation may also be complicated by the presence of medication side effects. The ID population is a highly medicated group; many individuals with ID are on multiple psychotropic medications from the same or different classes (Tonge et al., 2017). This can lead to side effects that are difficult to discern from organic medical problems. Further complicating the evaluation process, administering standard examination procedures (e.g., blood draws, neuroimaging) is not always possible given the individual's level of cooperation and challenging behaviors. However, medical conditions that are not recognized and therefore are not treated can lead to more serious conditions later in life as well as greatly reduce an individual's quality of life (Matson & Cervantes, 2019; van Schrojenstein Lantman-de Valk et al., 1997).

To improve our care for the ID population, emphasis should be placed on continued advancements in our understanding of the health risks and preventative service needs of the ID population. It is essential that individuals with ID receive routine checkups on physical health, including dental health, to monitor symptoms, as medical conditions often present as an increase in challenging behavior (Tonge et al., 2017). Regular assessment of medication effectiveness and side effects should also be conducted; the use of tools like the *Matson Evaluation of Drug Side effects* (*MEDS*), which was developed for use with the ID population, would be helpful in this process (Matson & Cervantes, 2013). Medical professionals should be trained in frequently occurring medical problems within the ID population as well as necessary adaptations for medical service provision of people with communication and behavioral difficulties. Improving these assessment procedures should be at the forefront of ID research. Current best practice involves a multidisciplinary approach in which professionals from a variety of disciplines (e.g., speech and language therapy, psychology, medicine) work together in the evaluation and treatment of medical comorbidities within the ID population.

Comorbid Neurodevelopmental and Psychiatric Disorders

In addition to medical conditions, the ID population presents with increased vulnerability for co-occurring neurodevelopmental and psychiatric disorders (Emerson, 2003; Matson & Shoemaker, 2011; Tonge et al., 2017; Wallander, Dekker, & Koot, 2006). Historically, researchers and clinicians did not believe that individuals with ID could have comorbid psychiatric diagnoses (Matson & Shoemaker, 2011). However, it is now clear that ID and mental health conditions are distinct and can and often do present together (Tonge et al., 2017). Of note, prevalence estimates of mental health problems range widely in the ID research literature due to difficulties in assessing for symptoms (Costello & Bouras, 2006). With regard to specific comorbid conditions, attention-deficit/hyperactivity disorder (ADHD) is most common and occurs within the ID population at rates four times higher than in the general population (Neece, Baker, Crnic, & Blacher, 2013). ASD is also highly prevalent within the ID population, with up to 40% of the population meeting ASD diagnostic criteria (La Malfa, Lassi, Bertelli, Salvini, & Placidi, 2004). Further, individuals with ID are at higher risk for psychiatric diagnoses like anxiety and depression (Tonge et al., 2017).

As in medical comorbidities, mental health conditions are also associated with the etiologies of ID. For example, individuals with Prader-Willi syndrome often experience obsessive-compulsive disorder, individuals with Williams syndrome are at increased risk for specific phobias and generalized anxiety, and individuals with DiGeorge syndrome have higher rates of bipolar disorder and psychosis (Tonge et al., 2017). Further, several genetic disorders associated with ID are implicated in ASD, such as Fragile X syndrome and tuberous sclerosis (Muhle, Trentacoste, & Rapin, 2004). While mental health problems in individuals with ID tend to be more stable across the life course (Wallander et al., 2006), emotional and behavioral symptoms are most prevalent in children and adolescence (Tonge et al., 2017). As is the case in the general population, gender differences in psychopathology exist in the ID population. ASD and ADHD are more common in males, whereas females with moderate to severe ID are at heightened risk for anxiety (Tonge et al., 2017). Other risk factors for psychiatric comorbidity in ID include socioeconomic deprivation, living in settings with limited social and occupational opportunities, experiencing co-occurring physical health symptoms, and increased parental and familial distress (Tonge et al., 2017; Wallander et al., 2006).

Despite the high prevalence of mental health problems in the ID population, it is not uncommon for individuals with ID to go without receiving a comorbid diagnosis or to be misdiagnosed, as psychiatric evaluation is complex due to intellectual, language, and social difficulties. First, diagnostic overshadowing may occur where clinicians inaccurately attribute symptoms of concurrent disorders to ID. Further, while psychopathology in individuals with mild-to-moderate ID may be similar to that of the general population, symptoms of psychiatric disorders may present atypically in individuals with more severe ID (Costello & Bouras, 2006; Matson & Shoemaker, 2011).

Given these difficulties, current diagnostic systems are limited in their utility with the ID population, and a range of adaptations must be made for appropriate assessment (Matson & Shoemaker, 2011; Tonge et al., 2017). Psychiatric evaluation should include extensive interview of individuals with ID (to the extent possible) and their caregivers about symptom presentation, the contexts in which symptoms are occurring, and how current functioning differs from premorbid functioning. Accommodations to improve the interviewing of individuals with ID may include visual supports like mood scales and increased session time to allow time for anxiety reduction and rapport development. Ensuring that self- and informant-report measures are valid for use with the ID population prior to administration is also integral. Finally, behavioral observations are paramount to assess for nonverbal signs of psychopathology (Matson & Cervantes, 2019; Matson & Shoemaker, 2011; Tonge et al., 2017).

Following identification of a comorbid psychiatric condition, evidence-based interventions used within the general population (e.g., cognitive behavior therapy) can be applied to individuals with ID. Nevertheless, psychological treatments typically require thoughtful adaptations. These include simplifying concepts by the use of visuals, repetition of concepts and behavioral rehearsal of skills to encourage learning, a heavier focus on behavior than on cognitions, and reinforcement-based behavioral strategies (Matson & Shoemaker, 2011; Tonge et al., 2017). Medication management is also often provided in the treatment of comorbidity. The range of participants with ID being prescribed at least one psychotropic medication is 40–89%, with higher rates associated with comorbid psychiatric concerns and the presence of challenging behavior (Bowring, Totsika, Hastings, Toogood, & McMahon, 2017). However, research into medication efficacy and side effect profiles is limited for the ID population specifically, and adverse effects may occur more frequently in individuals with ID (Tonge et al., 2017). Therefore, close monitoring of symptoms and adverse effects by caregivers and within periodic reassessments is imperative in optimizing care for individuals with ID.

Associated Symptomology

Individuals with ID, with and without comorbidity, also present with a range of associated symptoms. Among these are challenging behaviors including but not limited to tantrums, aggression, noncompliance, property destruction, self-injury, repetitive behaviors, and feeding challenges. While these behaviors are not necessary for a diagnosis of ID, systematic assessment of commonly associated difficulties is important for informing treatment and monitoring an individual's progress across time. Evaluation and treatment of associated symptomology is particularly important given the significant impact challenging behaviors have on the inclusion of individuals with ID in educational, social, and vocational settings and on the quality of life of individuals with ID and their caregivers (Crocker et al., 2006).

Researchers have found that more than half of the ID population demonstrates some degree of challenging behavior, with a minority engaging in severe behavior causing significant bodily injury (Matson & Cervantes, 2019; Crocker et al., 2006). Challenging behavior is also the most common reason for referral for services in children with ID (Costello & Bouras, 2006). Of note, nearly 40% of individuals with ID receiving antipsychotic medication treatment did not have a psychiatric diagnosis, suggesting prescription related to the presence of challenging behavior. Psychotropic medication use is significantly associated with aggression and self-injurious behavior, older age, and male sex. These trends are concerning given the limited research evidence indicating effectiveness for treatment of challenging behavior as well as the large adverse effect profile of long-term use of many psychotropic medications (Bowring et al., 2017).

Behavioral interventions are currently the best practice for the treatment of challenging behaviors in ID; therefore, functional assessment of presenting challenging behaviors should be a priority within the evaluation process. Functional assessment is designed to identify maintaining variables to challenging behavior or behavioral functions. Functions assessed include social positive reinforcement (e.g., access to preferred items, access to attention), social negative reinforcement (e.g., escape from demands, avoidance), and automatic reinforcement (Lloyd & Kennedy, 2014). Functional assessments can be conducted indirectly through caregiver interview or informant-report measures (e.g., *Questions About Behavioral Function* [*QABF*]; Matson & Vollmer, 1995), descriptively through direct observation and measurement of challenging behavior or through an experimental functional analysis involving direct manipulation of environmental conditions to determine maintaining variables. Indirect functional assessment can be done rapidly but is subject to bias, and the challenging behavior is never observed. Descriptive functional assessment includes direct observation across many environments to describe behavior-environment patterns but can be time intensive and only yields hypotheses of functional relations. Experimental functional analyses is most accurate in determining the behavioral function but is time and cost intensive, requires expertise, and exposes individuals to conditions that likely induce challenging behavior. Because of this, experimental methods are rarely used within clinical settings (Lloyd & Kennedy, 2014).

After the completion of a functional assessment, an informed function-based intervention can be developed and implemented utilizing evidence-based strategies like extinction and differential reinforcement. Although these practices are empirically supported, an individual with ID should not be treated in isolation. Given the interaction between caregiver stress and behavior problems in the ID population, where parent behavior contributes to challenging behavior and challenging behavior then influences parenting, a family-focused approach is imperative. Treatment should not only incorporate behavioral parent training to improve parent-child interactions and parent effectiveness in managing challenging behaviors but also include strategies to reduce parent stress, like mindfulness. This addition would help improve caregiver ability to access parent training instruction and implement strategies with fidelity (Crnic, Neece, McIntyre, Blacher, & Baker, 2017).

Conclusion

Comprehensive and individualized assessment and reassessment across the life course is integral in developing adequate supports for individuals with ID and their families. Within these assessments, clinicians should use a developmental perspective when building a testing battery, interpreting results, and providing recommendations. For example, when evaluating a young child with cognitive and adaptive delays, an understanding of relative strengths and weaknesses within the context of developmental milestones would inform appropriate early intervention referral. Further, providing recommendations encouraging the development of skills required for learning (e.g., attending behavior, understanding contingencies) would be imperative at this young age. In contrast, when evaluating an older adult with ID, assessment should be directed toward informing vocational opportunities and appropriate residential settings.

As mentioned, ID increases an individual's vulnerability for medical and psychiatric comorbidity and behavioral difficulties. Therefore, an evaluation at any age that is not inclusive of screening for commonly co-occurring but distinct conditions would be inadequate. A multidisciplinary team approach is essential in ensuring the assessment is as wide-ranging as the symptom clusters associated with ID. As mentioned, when a team approach is unavailable, clinicians should acknowledge that individuals with ID are at increased vulnerability for a variety of difficulties, some of which may be outside the clinician's realm of specialty. Therefore, increasing competence in screening for these difficulties and then making appropriate referrals is necessary to improve access to care and address the significant proportion of the population experiencing unmet service needs.

Finally, caregivers of individuals with ID experience heightened rates of stress (Patton, Ware, McPherson, Emerson, & Lennox, 2016). These caregivers undertake tremendous responsibilities throughout the developmental course, from navigating federal agencies and insurance companies to access care and supports to making decisions regarding appropriate school placements and intervention services and researching employment options and residential settings preparing for the transition to adulthood. Importantly, researchers have found that increased social supports is associated with reduced caregiver stress

(Patton et al., 2016). Therefore, professionals should screen for caregiver stress within evaluations, make appropriate recommendations for support and advocacy groups, and be available to help caregivers make well-informed decisions for their child (Matson & Cervantes, 2019).

References

American Psychiatric Association. (1952). *Diagnostic and statistical manual of mental disorders* (1st ed.). Washington, DC: Author.

American Psychiatric Association. (1968). *Diagnostic and statistical manual of mental disorders* (2nd ed.). Washington, DC: Author.

American Psychiatric Association. (1980). *Diagnostic and statistical manual of mental disorders* (3rd ed.). Washington, DC: Author.

American Psychiatric Association. (1987). *Diagnostic and statistical manual of mental disorders*. (3rd ed., Rev.). Washington, DC: Author.

American Psychiatric Association. (1994). *Diagnostic and statistical manual of mental disorders* (4th ed.). Washington, DC: Author.

American Psychiatric Association. (2000). *Diagnostic and statistical manual of mental disorders*. (4th ed., Text rev.). Washington, DC: Author.

American Psychiatric Association. (2013). *Diagnostic and statistical manual of mental disorders* (5th ed.). Washington, DC: Author.

Bayley, N. (2006). *Bayley scales of infant and toddler development, Third Edition: Technical manual*. San Antonio, TX: Harcourt.

Bishop, S. L., Guthrie, W., Coffing, M., & Lord, C. (2011). Convergent validity of the mullen scales of early learning and the differential ability scales in children with autism spectrum disorders. *American Journal on Intellectual and Developmental Disabilities; Washington, 116*(5), 331–343.

Bowring, D. L., Totsika, V., Hastings, R. P., Toogood, S., & McMahon, M. (2017). Prevalence of psychotropic medication use and association with challenging behaviour in adults with an intellectual disability. A total population study. *Journal of Intellectual Disability Research, 61*(6), 604–617.

Brown, L., Sherbenou, R. J., & Johnsen, S. K. (2010). *Test of Nonverbal Intelligence, Fourth Edition (TONI-4)*. Austin, TX: PRO-ED.

Brue, A. W., & Wilmshurst, L. (2016). *Essentials of intellectual disability assessment and identification*. Hoboken, NJ: John Wiley & Sons.

Buie, T., Campbell, D. B., Fuchs, G. J., Furuta, G. T., Levy, J., VandeWater, J., … Winter, H. (2010). Evaluation, diagnosis, and treatment of gastrointestinal disorders in individuals with asds: A consensus report. *Pediatrics, 125*(Supplement), S1–S18.

Cardona, F. A. (1994). Milestones in the history of mental retardation. *Journal of the South Carolina Medical Association (1975), 90*(6), 285–288.

Centers for Disease Control and Prevention (2017). Developmental disabilities definitions. Retrieved March 1, 2018, from https://ephtracking.cdc.gov/showDevelopmentalDisabilitiesDefinitions

Cervantes, P., & Jang, J. (2016). Methods and procedures for measuring comorbid disorders: medical. In J. Matson (Ed.), *Comorbid conditions among children with autism spectrum disorders* (pp. 65–90). Cham: Springer.

Costello, H., & Bouras, N. (2006). Assessment of mental health problems in people with intellectual disabilities. *Israel Journal of Psychiatry and Related Sciences, 43*(4), 241.

Crnic, K. A., Neece, C. L., McIntyre, L. L., Blacher, J., & Baker, B. L. (2017). Intellectual disability and developmental risk: Promoting intervention to improve child and family well-being. *Child Development, 88*(2), 436–445.

Crocker, A. G., Mercier, C., Lachapelle, Y., Brunet, A., Morin, D., & Roy, M.-E. (2006). Prevalence and types of aggressive behaviour among adults with intellectual disabilities. *Journal of Intellectual Disability Research, 50*(9), 652–661.

de Jong, A., Dondorp, W. J., Frints, S. G. M., de Die-Smulders, C. E. M., & de Wert, G. M. W. R. (2011). Advances in prenatal screening: The ethical dimension. *Nature Reviews Genetics, 12*(9), 657–663.

Devlieger, J. P. (2003). From 'idiot' to 'person with mental retardation': Defining differences in an effort to dissolve it. In J. P. Devlieger, F. Rusch, & D. Pfeiffer (Eds.), *Rethinking disability: The emergence of new definitions, concepts, and communities* (pp. 169–188). Antwerp: Garant.

Didden, R., Korzilius, H., van Aperlo, B., van Overloop, C., & de Vries, M. (2002). Sleep problems and daytime problem behaviours in children with intellectual disability. *Journal of Intellectual Disability Research, 46*(7), 537–547.

Doll, E. A. (1941). Notes on the concept of mental deficiency. *The American Journal of Psychology, 54*(1), 116–124.

Elliott, C. (2007). *Differential Ability Scales* (2nd ed.). San Antonio, TX: Harcourt Assessment.

Emerson, E. (2003). Prevalence of psychiatric disorders in children and adolescents with and without intellectual disability. *Journal of Intellectual Disability Research, 47*(1), 51–58.

Genetic Alliance. (2009). *Understanding genetics: A new york, mid-atlantic guide for patients and health professionals; the new york-mid-atlantic consortium for genetic and newborn screening services*. Washington, DC: Genetic Alliance.

Greenspan, S., & Switzky, H. S. (2003). Forty years of AAMR manuals. In H. S. Switzky & S. Greenspan (Eds.), *What is mental retardation? Ideas for an evolving disability in the 21st century* (pp. 3–28). Washington, DC: American Association on Mental Retardation.

Hammill, D. D., Pearson, N. A., & Weiderholt, J. L. (2009). *Comprehensive Test of Nonverbal Intelligence, Second Edition (CTONI-2)*. Austin, TX: PRO-ED.

Harris, J. C. (2006). *Intellectual disability: Understanding its development, causes, classification, evaluation, and treatment*. USA: Oxford University Press.

Harris, J. C. (2010). Advances in understanding behavioral phenotypes in neurogenetic syndromes. *American Journal of Medical Genetics Part C: Seminars in Medical Genetics, 154C*(4), 389–399.

Harrison, P., & Oakland, T. (2015). *Adaptive behavior assessment system, Third Edition (ABAS)*. San Antonio, TX: Pearson.

Hessl, D., Nguyen, D. V., Green, C., Chavez, A., Tassone, F., Hagerman, R. J., … Hall, S. (2009). A solution to limitations of cognitive testing in children with intellectual disabilities: The case of fragile X syndrome. *Journal of Neurodevelopmental Disorders, 1*(1), 33–45.

Kuriakose, S. (2014). Concurrent validity of the WISC-IV and DAS-II in children with autism spectrum disorder. *Journal of Psychoeducational Assessment, 32*(4), 283–294.

La Malfa, G., Lassi, S., Bertelli, M., Salvini, R., & Placidi, G. F. (2004). Autism and intellectual disability: A study of prevalence on a sample of the Italian population. *Journal of Intellectual Disability Research, 48*(3), 262–267.

Lloyd, B. P., & Kennedy, C. H. (2014). Assessment and treatment of challenging behaviour for individuals with intellectual disability: A research review. *Journal of Applied Research in Intellectual Disabilities, 27*(3), 187–199.

Matson, J. L., & Cervantes, P. E. (2013). Current status of the matson evaluation of drug side effects (MEDS). *Research in Developmental Disabilities, 34*(5), 1849–1853.

Matson, J. L., & Cervantes, P. E. (2019). Intellectual disabilities. In T. H. Ollendick, S.W. White, & B. A. White (Eds.), *The oxford handbook of clinical child psychology*. (pp. 165–176). Oxford, UK: Oxford University Press.

Matson, J. L., Mahan, S., Hess, J. A., & Fodstad, J. C. (2010). Effect of developmental quotient on symptoms of inattention and impulsivity among toddlers with Autism Spectrum Disorders. *Research in Developmental Disabilities, 31*(2), 464–469.

Matson, J. L., & Shoemaker, M. E. (2011). Psychopathology and intellectual disability. *Current Opinion in Psychiatry, 24*(5), 367–371.

Matson, J. L., & Vollmer, T. (1995). *Questions about behavioral function (QABF)*. Baton Rouge, LA: Disability Consultants, LLC.

Maulik, P. K., Mascarenhas, M. N., Mathers, C. D., Dua, T., & Saxena, S. (2011). Prevalence of intellectual disability: A meta-analysis of population-based studies. *Research in Developmental Disabilities, 32*(2), 419–436.

McDonald, L. (2006). Investigation of global developmental delay. *Archives of Disease in Childhood, 91*(8), 701–705.

Moeschler, J. B., Shevell, M., & Committee on Genetics. (2014). Comprehensive evaluation of the child with intellectual disability or global developmental delays. *Pediatrics, 134*(3), e903–e918.

Muhle, R., Trentacoste, S. V., & Rapin, I. (2004). The genetics of autism. *Pediatrics, 113*(5), e472–e486.

Mullen, E. M. (1995). *Mullen scales of early learning: AGS edition*. Circle Pines, MN: American Guidance Service.

National Research Council. (2002). *Mental retardation: Determining eligibility for social security benefits*. Washington, DC: National Academies Press.

Neece, C. L., Baker, B. L., Crnic, K., & Blacher, J. (2013). Examining the validity of ADHD as a diagnosis for adolescents with intellectual disabilities: Clinical presentation. *Journal of Abnormal Child Psychology, 41*(4), 597–612.

Newborg, J. (2005). *Battelle developmental inventory* (2nd ed.). Itasca, IL: Riverside.

Nyhan, W. L., O'Neill, J. P., Jinnah, H. A., & Harris, J. C. (2014). Lesch-Nyhan syndrome. In M. P. Adam, H. H. Ardinger, R. A. Pagon, S. E. Wallace, L. J. Bean, K. Stephens, & A. Amemiya (Eds.), *GeneReviews®*. Seattle, WA: University of Washington, Seattle.

Ozonoff, S., Goodlin-Jones, B. L., & Solomon, M. (2005). Evidence-based assessment of autism spectrum disorders in children and adolescents. *Journal of Clinical Child and Adolescent Psychology, 34*(3), 523–540.

Patton, K. A., Ware, R., McPherson, L., Emerson, E., & Lennox, N. (2016). Parent-related stress of male and female carers of adolescents with intellectual disabilities and carers of children within the general population: A cross-sectional comparison. *Journal of Applied Research in Intellectual Disabilities, 31*(1), 51–61.

Peterson-Besse, J. J., O'Brien, M. S., Walsh, E. S., Monroe-Gulick, A., White, G., & Drum, C. E. (2014). Clinical preventive service use disparities among subgroups of people with disabilities: A scoping review. *Disability and Health Journal, 7*(4), 373–393.

Roid, G. H. (2003). *Stanford-Binet Intelligence Scales, Fifth Edition: Examiner's Manual*. Itasca, IL: Riverside Publishing.

Roid, G. H., Miller, L. J., Pomplun, M., & Koch, C. (2013). *Leiter International performance scale, Third Edition (Leiter-3)*. Wood Dale, IL: Stoelting.

Sattler, J. M. (2008). *Assessment of children: cognitive foundations* (5th ed.). San Diego, CA: Jerome M. Sattler, Publisher.

Schalock, R. L., Borthwick-Duffy, S. A., Bradley, V. J., Buntinx, W. H. E., Coulter, D. L., Craig, E. M., … Yeager, M. H. (2010). *Intellectual disability: Definition, classification, and systems of supports* (11th ed.). American Association on Intellectual and Developmental Disabilities.

Schalock, R. L., & Luckasson, R. (2004). American association on mental retardation's definition, classification, and system of supports and its relation to international trends and issues in the field of intellectual disabilities. *Journal of Policy and Practice in Intellectual Disabilities, 1*(3–4), 136–146.

Schalock, R. L., Luckasson, R. A., & Shogren, K. A. (2007). The renaming of mental retardation: Understanding the change to the term intellectual disability. *Intellectual and Developmental Disabilities, 45*(2), 116–124.

Shevell, M. (2008). Global developmental delay and mental retardation or intellectual disability: Conceptualization, evaluation, and etiology. *Pediatric Clinics of North America, 55*(5), 1071–1084.

Silverman, W., Miezejeski, C., Ryan, R., Zigman, W., Krinsky-McHale, S., & Urv, T. (2010). Stanford-binet & WAIS IQ differences and their implications for adults with intellectual disability (aka mental retardation). *Intelligence, 38*(2), 242–248.

Sparrow, S. S., Cicchetti, D. V., & Saulnier, C. A. (2016). *Vineland adaptive behavior scales, Third Edition (Vineland-3)*. Minneapolis, MN: Pearson Assessments.

Tirosh, E., & Jaffe, M. (2011). Global developmental Delay and mental retardation: A pediatric perspective. *Developmental Disabilities Research Reviews, 17*(2), 85–92.

Tonge, B., Torr, J., & Trollor, J. (2017). Mental health and wellbeing in people with intellectual disability. In S. Bloch, S. A. Green, A. Janca, P. B. Mitchell, & M. Robertson (Eds.), *Foundations of clinical psychiatry* (4th ed.). Melbourne: Melbourne University Publishing Limited.

van Bokhoven, H. (2011). Genetic and epigenetic networks in intellectual disabilities. *Annual Review of Genetics, 45*(1), 81–104.

van Schrojenstein Lantman-de Valk, H. M., Metsemakers, J. F., Haveman, M. J., & Crebolder, H. F. (2000). Health problems in people with intellectual disability in general practice: a comparative study. *Family Practice, 17*(5), 405–407.

van Schrojenstein Lantman-de Valk, H. M. J., van den Akker, M., Maaskant, M. A., Haveman, M. J., Urlings, H. F. J., Kessels, A., & Crebolder, H. F. J. (1997). Prevalence and incidence of health problems in people with intellectual disability. *Journal of Intellectual Disability Research, 41*(1), 42–51.

Wallander, J. L., Dekker, M. C., & Koot, H. M. (2006). Risk factors for psychopathology in children with intellectual disability: A prospective longitudinal population-based study. *Journal of Intellectual Disability Research, 50*(4), 259–268.

Wechsler, D. (2008). *Wechsler Adult Intelligence Scale–Fourth Edition (WAIS-IV)*. San Antonio, TX: Psychological Corporation.

Wechsler, D. (2012). *The Wechsler Preschool and Primary Scale of Intelligence, Fourth Edition (WPPSI-IV)*. San Antonio, TX: The Psychological Corporation.

Wechsler, D. (2014). *Wechsler Intelligence Scale for Children-Fifth Edition*. Bloomington, MN: Pearson.

Wechsler, S. B., & McDonald, M. (2018). The dysmorphic newborn. In E. T. Alboliras, Z. M. Hijazi, C. Lopez, & D. J. Hagler (Eds.), *Visual guide to neonatal cardiology*. Hoboken, NJ: John Wiley and Sons, Inc..

Witwer, A. N., Lawton, K., & Aman, M. G. (2014). Intellectual disability. In E. J. Mash & R. A. Barkley (Eds.), *Child psychopathology* (3rd ed., pp. 593–624). New York, NY: Guilford Publications.

4

Problem-Solving and Working Memory in People with Intellectual Disabilities: An Historical Perspective

Ralph P. Ferretti

The experimental study of human memory was marked by the publication of *Memory: A Contribution to Experimental Psychology* (Ebbinghaus, 1964). In this seminal work, Ebbinghaus (1964) described the rigorous application of carefully controlled experimental methods to study fundamental aspects of human memory. Using himself as a subject, he investigated the rapidity with which he learned nonsense syllables, forgot them over time, and retained them as a function of the number of repetitions and repeated relearning. Among his many discoveries include the learning curve, the forgetting curve, and the serial-position effect. While Ebbinghaus self-consciously sought to control the effects of meaningful material on learning and memory, he noted, almost as an aside and somewhat presciently, that his "occasional tests" with Byron's *Don Juan* revealed "…an extraordinary advantage which the combined ties of meaning, rhythm, rhyme, and a common language give to material to be memorized."

The work of Norman Ellis (1963, 1970, 1978) occupies a similarly important place in the history of research about intelligence-related differences in memory and problem-solving. He was among the first scholars to experimentally investigate the possibility of structural constraints on the learning and memory of people with intellectual disabilities (Ellis, 1963). His later studies (Ellis, 1970) provided evidence for intelligence-related differences in strategic and controllable aspects of memory. It is not hyperbole to say that this work inaugurated decades of research about the memory and problem-solving of people with intellectual disabilities. Professor Ellis, who was my dissertation advisor, passed away as I was writing this paper. It is to his memory that this chapter is dedicated.

This report is a selective review of the literature focusing on the problem-solving and working memory[1] of people with intellectual disabilities. Any effort to undertake a comprehensive review of the literature about the cognition of people with intellectual disabilities would require at least a book-length manuscript. My purpose here is to highlight the influence of Norman Ellis's work on the development of theory and research about problem-solving and working memory of people with intellectual disabilities. I begin by describing a theory of

R. P. Ferretti (✉)
School of Education & Department of Psychological and Brain Sciences, University of Delaware,
Newark, DE, USA
e-mail: ferretti@udel.edu

---

[1] Short-term memory (STM) and working memory (WM) have been used interchangeably to refer to a cognitive system responsible for the temporary storage of information. A theoretical distinction between STM and WM is based on whether the system is simply responsible for maintaining information (STM) or maintaining and manipulating information (WM). However, the concepts have not been consistently distinguished in behavioral data (Aben, Stapret, & Blokland 2012). In this paper, these terms will be cited as they were used by the scholars cited in this manuscript.

J. L. Matson (ed.), *Handbook of Intellectual Disabilities*, Autism and Child Psychopathology Series, https://doi.org/10.1007/978-3-030-20843-1_4

problem-solving (Ferretti & Belmont, 1983; Ferretti & Cavalier, 1991) to provide an interpretative framework for Ellis's pioneering work about intelligence-related differences in memory processes. I then discuss scholarship that was given impetus by his research, including studies focusing on strategic and self-regulatory determinants of intelligence-related differences in memory and problem-solving (e.g., Belmont & Butterfield, 1969; Belmont, Butterfield, & Ferretti, 1982; Borkowski & Cavanaugh, 1979; Borkowski, Reid, & Kurtz, 1984; Bray, 1987; Bray, Fletcher, & Turner, 1997; Brown, 1974; Butterfield & Ferretti, 1984; Butterfield, Siladi, & Belmont, 1980; Campione & Brown, 1977; Ferretti & Cavalier, 1991). I conclude the chapter with a discussion of scholarship about the self-determination of students with intellectual disabilities (Wehmeyer, 2001, 2007). As the reader will see, the historical research about self-regulation, adaptation, and problem-solving informs current efforts to ensure that people with intellectual disabilities are empowered to determine their lives.

Problem-Solving and Adaptation

I take it as axiomatic that people with intellectual disabilities experience deficits in intellectual skills and adaptive functioning (American Association on Intellectual and Developmental Disabilities, 2010). Intelligent adaptation depends upon the ability to solve problems (Ferretti & Belmont, 1983) that arise when a discrepancy exists between a person's current state and a desired goal. A major challenge for the problem-solver is to construct an internal representation of possible solution paths that may reduce this discrepancy (Ferretti & Belmont, 1983). In short, problem-solving involves the strategic deployment of cognitive resources to achieve one's goals (Newell & Simon, 1972). Research about problem-solving focuses on the processes by which people adapt to problematic conditions, as well as the conditions that require adaptation. Viewed this way, the study of problem-solving is a natural extension of the pioneering psychometric work about individual differences in intelligence and the adaptive consequences that result from those differences (Ferretti & Belmont, 1983; Ferretti & Butterfield, 1989; Ferretti & Cavalier, 1991).

Problem-solvers use subordinate and superordinate processes to achieve their goals. Subordinate processes involve the use of step-by-step computational procedures for eliminating the discrepancy between the current state and the goal, i.e., *strategies* (Ferretti & Cavalier, 1991). Of course, strategies differ in their likelihood of success. If a strategy is unsuccessful, it will need to be revised or abandoned in favor of a different strategy. It should be clear that problem-solvers must judiciously choose strategies, monitor their implementation, and attend to feedback about their efficacy. Problem-solving, reasoning, and decision-making are interdependent and affected by individual differences in the psychological processes upon which they depend (Ferretti & Cavalier, 1991).

Superordinate processes are thought to be responsible for overseeing and managing subordinate computational strategies. These *executive or self-management processes* (Atkinson & Shiffrin, 1968; Belmont, Butterfield, & Ferretti, 1982; Butterfield & Belmont, 1977) have been imbued with powerful responsibilities, including goal-setting, problem identification, strategic planning, implementation, monitoring, and revision (Ferretti & Cavalier, 1991). There has been considerable debate in the literature about the necessity of an omniscient homunculus with nearly magical powers of self-regulation (see Baddeley, 1998). However, there can be no doubt that the concept of an executive has been a powerful heuristic for a set of interrelated and potentially malleable processes that can be taught and have been shown to positively impact learning and memory (Belmont, Butterfield, & Ferretti, 1982; Hacker, Dunlosky, & Graesser, 2009).

Problem-solving operates within an information-processing system that has limited capacity (Atkinson & Shiffrin, 1968; Newell & Simon, 1972). Information-processing theories recognize that goal-directed problem-solving requires the temporary storage and manipulation of information in memory and that problem-solving may make demands that exceed the system's working memory capacity (Ferretti & Cavalier, 1991). It should be noted, however, that the functional capacity of working memory can be affected by the organization of information, the use of strategies for maintaining and recoding that information, and the automatization of processes that would otherwise place demands on the system's limited capacity (Anderson, 1981, 1983; Ellis, 1970; Schneider & Shiffrin, 1977; Shiffrin & Schneider, 1977). In short, individual differences in knowledge, strategies, and automaticity interact in any given situation to influence problem-solving and the system's functional working memory capacity (Ferretti & Cavalier, 1991).

Stimulus Trace Theory

Since the ancients, people with intellectual disabilities have been distinguished from and accorded different treatment than people with typically developing intellectual and adaptive skills (Wehmeyer, 2013). One of the earliest definitions of intellectual disabilities (Esquirol, 1838) recognized that "idiocy… is a condition in which the intellectual faculties are never manifested or have never developed sufficiently… to acquire such amount of knowledge as persons of his own aged reared in similar circumstances are capable of receiving." Esquirol's focus on the adverse effects on learning of limited intellectual skills is especially relevant in this context. Much of the empirical research conducted since the mid-twentieth century about the memory and problem-solving of people with intellectual disabilities was designed to identify the psychological and contextual factors that are responsible for these learning difficulties.

Since the earliest psychometric studies of intelligence (e.g., Galton, 1887), memory span has been taken as a diagnostic sign of intellectual disabilities. However, the mechanisms that gave rise to these memory deficiencies and associated behavioral difficulties of people with intellectual disabilities were largely unexplored until Ellis's (1963) pioneering research. He hypothesized that intelligence-related behavioral differences were attributable to a central nervous system (CNS) deficiency that resulted in an impoverished stimulus trace. A very simple memory model based on Hull's (1952) theory of associationism and other neurophysiological theories that were current at the time was used to represent the hypothesized memory deficiency. In brief, an environmental stimulus elicited a transitory stimulus trace that mediated a behavioral response. The amplitude and duration of the stimulus trace were thought to be dependent upon the integrity of the person's CNS. People with intellectual disabilities, whose poor performance on intelligence tests was taken as prima facie evidence of CNS dysfunction, were hypothesized to have a stimulus trace deficit, that is, a trace that was less stable and decayed more quickly than that of people of typical ability. As a result, the duration of stimulus control would be less for people with intellectual disabilities compared to people of average intellectual ability. This theory was intended to account for unlearned behavior. However, Ellis recognized that prior learning was involved, especially when the person's response was delayed, and, furthermore, that future learning was affected by the duration of the stimulus trace.

Ellis (1963) predicted that behaviors that were dependent upon short-term memory would be differentially impacted by intelligence-related differences in the stimulus trace. For example, he reported data about intelligence-related differences in performance on tasks that required a delayed response as compared to those that required an immediate response. He also described findings that suggested that, compared to people without intellectual disabilities, the reaction time of people with intellectual disabilities was more negatively affected by the imposition of a delay between a warning signal and the signal to respond. Furthermore, he reported data that suggested that the paired associates learning of people with intellectual disabilities were differentially and adversely affected by the imposition of a delay between the presentations of the paired stimuli compared to people without intellectual disabilities. He also expected intelligence-related differences in serial-position errors during verbal learning, but the data were equivocal, so he called for further research. Data such as these were generally consistent with the hypothesized stimulus trace deficit in people with intellectual disabilities.

Serious questions about the evidence for the stimulus trace theory were raised by Belmont and Butterfield's (1969) critical review of research about the relations of short-term memory to age and intelligence. Because memory is determined by how much information is initially acquired, the rate with which it is forgotten, and the accuracy with which it is retrieved, Belmont and Butterfield focused their review on empirical studies that varied intelligence and age while controlling for differences in acquisition, retrieval, and other methodological confounds and measurement issues. They found no evidence for the hypothesis that short-term memory varied with age and virtually no credible evidence that it varied with intelligence. The few studies that appeared to show intelligence-related differences in short-term memory employed stimuli for which verbal labels could be used to mediate a delay. At that time, there was little research about the effects of intelligence-related differences in verbal mediation on short-term memory. However, Belmont and Butterfield's analysis raised the possibility that intelligence-related differences in short-term memory could be due to strategic factors influencing the acquisition, maintenance, and retrieval of information from short-term memory.

The Rehearsal Deficit Hypothesis

As I mentioned earlier, the stimulus trace theory of short-term memory was rooted in a simple model of associationism (Ellis, 1970). Subsequent to the publication of the stimulus trace theory (Ellis, 1963), more sophisticated models of human memory were proposed that represented memory processes in terms of stages of information processing involving the sequential storage, retrieval, and transformation of information. These models (e.g., Atkinson & Shiffrin, 1968; Waugh & Norman, 1965) included memory structures that stored information for specific durations (e.g., very short-term memory, short-term memory), had strategic processes that operated on that information (e.g., rehearsal and the articulatory loop), and transferred it to more permanent storage (long-term memory), where the information was represented in the form of abstract knowledge structures (i.e., schemata). These processes were thought to be initiated by sensory input that evoked a sequence of information processes and, as we discussed earlier, were under the control of executive functions that managed the person's limited information-processing resources.

Influenced by these models, Ellis (1970) undertook a series of experiments that focused on intelligence-related differences in short-term and long-term memory. Participants first viewed a sequence of numbers after which they were presented a probe that required the identification of the position in which the probe was last seen. This task allowed Ellis to study differences in memory across each of the positions in the sequence, i.e., Ebbinghaus's serial-position effect. This effect refers to the observation that items presented first and last in a list are remembered best, i.e., the primacy and recency portions of the memory curve. In the information-processing models current at that time (e.g., Atkinson & Shiffrin, 1968), the primacy effect was thought to reflect the transfer of information from short-term to long-term memory by the use of rehearsal strategies. In contrast, the recency effect was thought to reflect the capacity of short-term memory without rehearsal. The information in short-term memory was immediately retrievable but was lost quickly unless it was rehearsed.

The serial probe task also allowed Ellis (1970) to experimentally manipulate a number of factors that had been shown to influence rehearsal processes and memory performance, including item exposure duration, inter-item interval, and the interval between the last item and the probe. In a series of studies, Ellis showed that experimental manipulations that increased the amount of rehearsal time (e.g., a slower presentation rate of the to-be-remembered information) differentially benefitted the memory performance of college students as compared to their peers with intellectual disabilities but only in the primacy portion of the serial-position curve. Similarly, permitting participants to control the amount of time spent between the presentations of each item differentially benefited college students in the primary but not the recency portion of the curve. These findings are consistent with the conclusion that a rehearsal deficit was responsible for the short-term memory deficits of people with intellectual disabilities.

In addition, Ellis (1970) conducted studies that were designed to assess the rate of forgetting from short-term memory under conditions that were intended to preclude rehearsal. This was done by interpolating a variable delay between the presentation of the last item and the memory probe that was either unfilled or filled with a task designed to preclude rehearsal. If rehearsal was responsible for maintaining information in short-term memory, then the filled delay should have more adversely affected the performance of people without intellectual disabilities. Furthermore, if the stimulus trace was responsible for short-term memory, conditions designed to preclude rehearsal should have resulted in comparable decay rates for people with intellectual disabilities and their peers without disabilities.

Ellis (1970) reported that the imposition of a delay appeared to result in more rapid decay for people with intellectual disabilities compared to their peers without disabilities in both the unfilled and filled conditions. While the filled delay resulted in poorer short-term memory performance in both groups, it did not appear to have a differential effect related to intelligence. Given the latter findings and other apparent anomalies in these data, Ellis (1970) was reluctant to draw a strong conclusion about intelligence-related differences in decay rates in short-term memory. He called for a more definitive study that carefully controlled rehearsal strategies. Nevertheless, his data provided tentative support for a rehearsal deficit explanation of the poor memory performance of people with intellectual disabilities.

Ferretti's (1982) study of intelligence-related differences in decay rates from short-term memory was designed to answer Ellis's (1970) call for research that carefully controlled the use of rehearsal strategies. People with intellectual disabilities and their peers without disabilities recalled sequences of consonants after retention intervals of varying durations. In contrast to previous studies of intelligence-related differences in short-term memory, Ferretti (1982) controlled participants' acquisition levels by equating the amount of information they were required to recall with no delay. In addition, participants performed a tonal detection task that was designed to preclude the use memory strategies during the retention interval. Differences in the accuracy and latency of tonal detection with and without memory loads were used to identify people who rehearsed. Ferretti found that the decay rates from short-term memory for people who did not rehearse were unrelated to intelligence. By controlling the level of acquisition and the use of rehearsal strategies, he was able to demonstrate comparable forgetting rates for people with intellectual disabilities and their nondisabled peers. These findings provided empirical support for Belmont and Butterfield's (1969) conclusion about the status of the stimulus trace theory and indirect evidence for the importance of strategic processes in intelligence-related differences in memory performance.

Ellis's (1970) studies provided indirect evidence for the rehearsal deficit hypothesis because inferences about intelligence-related differences in rehearsal were drawn from participants' recall data under conditions that were designed to support or discourage its use. Direct evidence about intelligence-related differences in rehearsal was obtained by Belmont and Butterfield (1969), who designed a variant of the serial learning task that allowed for the explicit measurement of rehearsal strategies. Participants paced themselves through a list of consonants for the purpose of identifying the position of a probe that appeared at the end of the list. In addition to collecting information about their response accuracy, they also collected information about the distribution of participants' pauses across serial positions as they prepared to respond to the probe.

Belmont and Butterfield (1969) found that college students showed distinctive pause patterns that were indicative of different rehearsal strategies. For example, some students used a building strategy during acquisition that was associated with pauses that grew increasingly long for the first few items and then rapid pausing for the final items. This building pause pattern suggested that participants were cumulatively rehearsing information that was distal from the probe and capitalizing on the availability of information in short-term memory that was proximal to it, i.e., cumulative rehearsal fast-finish strategy (Pinkus & Laughery, 1970). In contrast, the pause patterns of many children without disabilities and children and adults with intellectual disabilities were flat and marked by rapid responding throughout the list. In short, the participants' pause patterns were used to infer the use of rehearsal strategies. Furthermore, pause patterns that were indicative of the use of rehearsal strategies were associated with better recall accuracy.

Belmont and Butterfield's (1969) findings about the relationship between intelligence-related strategic behavior and memory performance were confirmed and extended in future studies. For example, Belmont and Butterfield (1971) had people with intellectual disabilities and their normally achieving peers pace themselves through the serial-position task without any instruction. Under self-paced conditions, the abovementioned intelligence-related differences in strategy use and memory performance were observed. Participants then performed the task under two different conditions. In the active rehearsal condition, people with intellectual disabilities and their normally achieving peers were given an instruction to rehearse the earliest presented letters and then proceed quickly through the last few letters. In the passive strategy condition, participants were told to label each item as it appeared and then proceed immediately to the next letter. They found that compared to the self-paced condition, the active rehearsal condition resulted in an increased use of rehearsal in people with intellectual disabilities and improved memory performance in the primacy portion of the memory curve. In addition, the passive strategy condition adversely affected the use of rehearsal strategies in people without intellectual disabilities and depressed their memory performance in the primacy portion. These and other studies (e.g., Brown, 1974; Butterfield, Wambold, & Belmont, 1973) provided compelling experimental evidence for the rehearsal deficit explanation of intelligence-related differences in short-term memory.

Executive Process and Self-Management

Studies of the rehearsal deficit hypothesis established three important findings that needed explanation. First, instruction could be used to test hypotheses about the cognitive mechanisms that underlie intelligence- and age-related differences in memory performance (Belmont & Butterfield, 1977; Butterfield et al., 1980; Campione & Brown, 1977). Second, intelligence-related differences in short-term memory were ubiquitous, but they could be mitigated by the provision of instruction that supported strategy production (Bray & Turner, 1986; Ferretti & Cavalier, 1991). Third, people with intellectual disabilities often failed to transfer strategies that they learned and had improved their memory performance (Borkowski & Cavanaugh, 1979; Campione & Brown, 1984). Said differently, people with intellectual disabilities were strategically passive, and they could be taught to produce strategies that positively impacted memory performance. However, they did not seem to spontaneously generalize these strategies. This raises a fundamental question: Why were they unable to transfer strategies that had positively impacted their memory performance?

Belmont, Butterfield, and Ferretti (1982) reviewed instructional studies that were reported to promote strategy generalization. The instructional protocols used in these studies targeted subordinate strategies as well as superordinate executive processes and metacognitive knowledge that were thought to influence strategy production, and transfer (Butterfield & Ferretti, 1987). Belmont, Ferretti, and Mitchell (1982) referred to superordinate knowledge and processes as self-management skills because they could be used to regulate goal-directed planning and strategic problem-solving. For example, Kendall, Borkowski, and Cavanaugh (1980) taught students with mild intellectual disabilities to use a self-interrogation strategy to generate images that linked items during paired-associate learning. In addition, students were provided feedback about the effectiveness of the self-interrogation strategy. The feedback was interpreted by Belmont, Ferretti, and Mitchell (1982) to indirectly influence the students' superordinate processing. Kendall et al. (1980) reported that students who received feedback performed better on a triadic-associates task than those who did not receive feedback. This finding was taken as evidence by Belmont, Ferretti, and Mitchell (1982) for the influence of self-management skills. Similar conclusions about intelligence-related differences in strategy transfer and self-management skills were drawn by other researchers (see Ferretti, 1989; Ferretti & Cavalier, 1991).

I would be remiss if I failed to note that instructional studies designed to validate a process explanation of intelligence-related performance differences have not satisfied the inferential conditions needed to draw an irrefutable conclusion about the self-management hypothesis (Belmont & Butterfield, 1971; Brown & Campione, 1978; Ferretti & Cavalier, 1991). In brief, proponents of the instructional approach believe that instruction can be used to validate a theoretically motivated analysis of the processes underlying task performance (Ferretti & Cavalier, 1991). Predictions should then follow from this analysis about the processes that are putatively responsible for individual or group differences in performance. Experimental tests of these predictions should show that teaching people who did not spontaneously use these processes improved their performance. In contrast, the performance of people who already use these processes should be unaffected by the same instruction. To my knowledge, these conditions have never been met in the instructional literature.

Furthermore, proponents of the self-management hypothesis may have underestimated the strategic proclivities of people with intellectual disabilities (Bray, Fletcher, & Turner, 1997; Ferretti & Cavalier, 1991). The literature shows that people with intellectual disabilities can be strategic without self-management support when the task is not too difficult (Bray & Turner, 1987; Spitz & Borys, 1984), they understand the task requirements (Bray & Turner, 1987; Bray et al., 1997), and they possess the content knowledge about the domain in which the problem-solving occurs (Bray et al., 1997; Brown, Bransford, Ferrara, & Campione, 1983). Each of these factors is known to affect functional working memory capacity (Ferretti & Cavalier, 1991; Ferretti & Butterfield, 1992; Spitz & Borys, 1984).

Questions remain about the status of the self-management hypothesis as represented by advocates of the instructional approach to validating process theories of intelligence. The literature shows that people with intellectual disabilities can be spontaneously strategic when the task requirements do not overtax their functional memory capacity (Ferretti and Cavalier, 1991). However, questions about the self-management hypothesis should not raise doubts about the practical benefits of training self-management skills. The literature clearly shows that self-management training can have a salutary effect on the performance of people with intellectual disabilities (Ferretti & Cavalier, 1991; Ferretti, Cavalier, Murphy, & Murphy, 1993; Wehmeyer, 2007). This finding is important because the inclusion of people with intellectual disabilities in normalized environments presupposes their ability to set personal goals, monitor performance, and be less reliant on external conditions for controlling their behavior (Ferretti & Cavalier, 1991; Ferretti et al., 1993). In addition, the capacity of people with intellectual disabilities to manage their behavior accords dignity, reduces the behavioral and economic costs of dependency, and increases their potential to live and work in settings with limited supervision (Ferretti & Cavalier, 1991). These highly desirable outcomes are consistent with the core values that undergird our system of representative democracy (Wehmeyer, 2001).

Self-Determination

Much of the theoretical work about individual differences in intelligence has been concerned with identifying the cognitive processes that distinguish people with intellectual disabilities and their normally achieving peers. As I mentioned earlier, this was a natural extension of psychometricians' conjectures about the psychological processes that were responsible for individual and group differences in intelligence (Ferretti & Belmont, 1983). This research resulted in the development of new inferential methods for studying intelligence-related differences in those processes, and while the findings were not always directly applicable, they gave impetus to the development of instructional techniques for improving students' self-regulatory skills in educational contexts (e.g., Hacker et al., 2009).

Studies of self-management in the behavioral tradition (Ferretti & Cavalier, 1991; Ferretti et al., 1993) have been less concerned about uncovering the underlying psychological processes that account for intelligence-related differences in academic performance. Rather, most of this work was done to establish the efficacy of training that mitigated the adaptive difficulties experienced by people with intellectual disabilities (Ferretti et al., 1993). The focus on the practical aspects of intelligent adaptation was driven by parental opposition to the institutional segregation of students with intellectual disabilities and the ethical and legal imperatives to provide normalized experiences in inclusive settings (Wehmeyer & Shalock, 2013). As more normalized opportunities were made available and students were expected to participate in transition planning (Wehmeyer, 2001), the notion of a disability was reconceptualized as a fit between the person's capacities and the environmental demands rather than as an intrinsic property of the person (Ferguson, Ferguson, & Wehmeyer, 2013). Researchers and practitioners began to focus on abilities instead of disabilities. As we have seen, people with intellectual disabilities can demonstrate surprising competencies under supportive conditions. In short, there was a pressing need to identify interventions that empowered people with intellectual disabilities to realize their self-determined goals (Wehmeyer, 2007).

Self-determination is a construct that implies that people cause themselves to act by internal forces instead of being compelled to act by external agents (Wehmeyer, 2001). Self-determined actions are volitional in the sense that they are the result of conscious choices. Choice making is an important aspect of self-determination because it involves exercising control over one's fate (Wehmeyer, 2001). Viewed this way, self-determination is a form of self-regulation in which self-determined people choose to act to reduce the discrepancy between their goals and current state (Mithaug, 1993). This conception is remarkably similar to the view espoused by information-processing theorists (e.g., Newell & Simon, 1972). However, it has a motivational aspect because it accounts for the psychological effects of failure on expectations about the likely success of future choices. Unfortunately, the evidence shows that people with intellectual disabilities have had relatively fewer opportunities to exercise choice (Wehmeyer, 2001), and when they have, they often experience failure (Wehmeyer, 2007). The antidote to low expectations is to create frequent opportunities for choice making in situations that that have a high likelihood of success and encourage realistic assessments of likely outcomes (Mithaug, 1993). In short, self-determined decision-making depends on self-management skills (Wehmeyer, 2001).

Wehmeyer's (Wehmeyer, 2001, 2007; Wehmeyer, Agran, & Hughes, 1998) functional model identifies the conditions that influence the development of self-determination and the functions that self-determination serve. According to the model, self-determination develops as people acquire attitudes and skills that promote a sense of causal agency. The development of self-determination depends upon opportunities to experience success when people act as causal agents. As a result of these experiences, people develop expectations and beliefs about their causal efficacy. Of course, supports are needed to ensure that causal actions are met with success and also to develop the self-regulatory skills needed to set and achieve realistic goals. When people experience success as causal agents, they develop a greater sense of autonomy, empowerment, and self-realization. Wehmeyer's functional model has been used to develop self-determination interventions for students with intellectual disabilities. These interventions have been shown to have a causal influence on students' self-determination (Wehmeyer, 2015), their participation in educational planning (Wehmeyer, Palmer, Lee, Williams-Diehm, & Shogren, 2011), and employment and community inclusion outcomes (Powers et al., 2012). In sum, there is a substantial and growing body of evidence about the heuristic value of self-determination theory and the efficacy of self-determination interventions.

Conclusions

I began this paper by providing a brief overview of problem-solving theory to situate Norman Ellis's (1963, 1970) contributions to the study of intelligence-related differences in short-term memory. Ellis was the first scholar to experimentally investigate the structural characteristics of short-term memory in persons with intellectual disabilities, and he was among the first to provide evidence about strategic aspects on memory. In short, Ellis (1963, 1970) conducted some of the earliest and most influential studies of structural and strategic aspects of intelligence-related differences in memory performance. Subsequent studies provided evidence that was inconsistent with the predictions of stimulus trace theory, and investigators that followed Ellis developed more refined methods that allowed for the direct measurement of variables that influence strategic behavior. While Ellis recognized the theoretical existence of executive processes, he did not investigate their influence on intelligence-related differences in memory and problem-solving. Nevertheless, his work inaugurated decades of research about the instructional and contextual conditions that influence the strategic self-regulation of memory processes and, indirectly through the influence of future researchers, the conditions that affect the self-determination of people with intellectual disabilities.

References

Aben, B., Stapret, S., & Blokland, A. (2012). About the distinction between working memory and short-term memory. *Frontiers in Psychology, 3*, 301.

American Association on Intellectual and Developmental Disabilities. (2010). *Intellectual disability: Definition, classification, and systems of support* (11th ed.). Washington, DC: AAIDD.

Anderson, J. R. (1981). *Cognitive skills and their acquisition*. Hillsdale, NJ: Erlbaum.

Anderson, J. R. (1983). *The architecture of cognition*. London: Harvard University Press.

Atkinson, R. C., & Shiffrin, R. M. (1968). Human memory: A proposed system and its control processes. In K. W. Spence & J. T. Spence (Eds.), *The psychology of learning and motivation: II*. Oxford, England: Academic Press.

Baddeley, A. (1998). The central executive: A concept and some misconceptions. *Journal of the International Neuropsychological Society, 4*, 523–526.

Belmont, J. M., & Butterfield, E. C. (1969). The relations of short-term memory to development and intelligence. In L. Lipsitt & H. Reese (Eds.), *Advances in child development and behavior* (Vol. 4, pp. 29–82). New York: Academic Press.

Belmont, J. M., & Butterfield, E. C. (1971). Learning strategies as determinants of memory deficiencies. *Cognitive Psychology, 2*, 411–420.

Belmont, J. M., & Butterfield, E. C. (1977). The instructional approach to developmental cognitive research. In R. V. Kail & J. W. Hagen (Eds.), *Perspectives on the development of memory and cognition* (pp. 437–481). Hillsdale, NJ: Erlbaum.

Belmont, J. M., Butterfield, E. C., & Ferretti, R. P. (1982). To secure transfer of training, instruct self-management skills. In D. K. Detterman & R. J. Sternberg (Eds.), *How and how much can intelligence be increased* (pp. 147–154). Norwood, NJ: Ablex.

Belmont, J. M., Ferretti, R. P., & Mitchell, D. W. (1982). Memorizing: A test of untrained mildly retarded children's problem solving. *American Journal of Mental Deficiency, 87*, 197–210.

Borkowski, J. G., & Cavanaugh, J. C. (1979). Maintenance and generalization of skills by the mentally retarded. In N. R. Ellis (Ed.), *Handbook of mental deficiency: Psychological theory and research* (2nd ed., pp. 569–617). Hillsdale, NJ: Erlbaum.

Borkowski, J. G., Reid, M. K., & Kurtz, B. E. (1984). Metacognition and retardation: Paradigmatic, theoretical, and applied perspectives. In P. H. Brooks, R. Sperber, & C. McCauley (Eds.), *Learning and cognition in the mentally retarded* (pp. 55–75). Hillsdale, NJ: Erlbaum.

Bray, N. W., & Turner, L. A. (1986). The rehearsal deficit hypothesis. In N. R. Ellis & N. W. Bray (Eds.), *International review of research in mental retardation* (Vol. 14, pp. 47–51). Orlando, FL: Academic Press.

Bray, N. W., & Turner, L. A. (1987). Production anomalies (not strategic deficiencies) in mentally retarded individuals. *Intelligence, 11*, 49–60.

Bray, N. W., Fletcher, K. L., & Turner, L. A. (1997). Cognitive competencies and strategy use in individuals with mental retardation. In W. E. MacLean Jr. (Ed.), *Ellis' handbook of mental deficiency, psychological theory and research* (3rd ed., pp. 197–217). Mahwah, NJ: Lawrence Erlbaum Associates.

Brown, A. L. (1974). The role of strategic behavior in retardate memory. In N. R. Ellis (Ed.), *International review of research in mental retardation* (Vol. 7, pp. 55–111). New York: Academic Press.

Brown, A. L., Bransford, J. D., Ferrara, R. A., & Campione, J. C. (1983). Learning, remembering, and understanding. In J. H. Flavell & E.M. Markman (Eds.) *Handbook of child psychology* (4th ed., pp. 77–166). New York: Wiley.

Brown, A. L., & Campione, J.C. (1978). Permissible inferences from cognitive training studies in developmental research. In W. S. Hall & M. Cole (Eds.), Quarterly Newsletter of the Institute for Comparative Human Development 2, pp. 46–53.

Butterfield, E. C., & Belmont, J. M. (1977). Assessing and improving the cognition of mentally retarded people. In I. Bialer & M. Sternlicht (Eds.), *Psychology of mental retardation: Issues and approaches* (pp. 277–318). New York: Psychological Dimensions.

Butterfield, E. C., & Ferretti, R. P. (1984). Some extensions of the instructional approach to the study of cognitive development and a sufficient condition for transfer of training. In P. H. Brooks, C. McCauley, & R. Sperber (Eds.), *Learning and cognition in the mentally retarded* (pp. 311–332). Hillsdale, NJ: Erlbaum.

Butterfield, E. C., & Ferretti, R. P. (1987). Toward a theoretical integration of cognitive hypotheses about intellectual differences among children. In J. G. Borkowski & J. D. Day (Eds.), *Cognition in special children: Comparative approaches to retardation, learning disabilities, and giftedness* (pp. 195–233). Norwood, NJ: Ablex.

Butterfield, E. C., Siladi, D., & Belmont, J. M. (1980). Validating theories of intelligence. In H. W. Reese & L. P. Lipsitt (Eds.), *Advances in child development and behavior* (Vol. 15, pp. 95–162). New York: Academic Press.

Butterfield, E. C., Wambold, C., & Belmont, J. M. (1973). On the theory and practice of improving short-term memory. *American Journal of Mental Deficiency, 77*, 654–669.

Campione, J. C., & Brown, A. (1977). Memory and metamemory development in educable retarded children. In R. V. Kail & J. W. Hagen (Eds.), *Perspectives on the development of memory and cognition* (pp. 367–406). Hillsdale, NJ: Erlbaum.

Campione, J. C., & Brown, A. L. (1984). Learning ability and transfer propensity as sources of individual differences in intelligence. In P. H. Brooks, R. Sperber, & C. McCauley (Eds.), *Learning and cognition in the mentally retarded* (pp. 265–293). Hillsdale, NJ: Erlbaum.

Ebbinghaus, H. (1964). *Memory: A contribution to experimental psychology (H. A. Ruger, C. E. Bussenius Trans.)*. New York: Dover. (Original work published 1885).

Ellis, N. R. (1963). The stimulus trace and behavioral inadequacy. In N. R. Ellis (Ed.), *Handbook of mental deficiency* (pp. 134–158). New York: McGraw-Hill.

Ellis, N. R. (1970). Memory processes in retardates and normals. In N. R. Ellis (Ed.), *International review of research in mental retardation* (Vol. 4, pp. 1–32). New York: Academic Press.

Ellis, N. R. (1978). Do the retarded have poor memory? *Intelligence, 2*, 41–54.

Esquirol, É. (1838). (original French edition).). *Mental maladies; a treatise on insanity. Translated from the French by Ebenezer Kingsbury Hunt*. Philadelphia, PA: Lea and Blanchard.

Ferguson, D. L., Ferguson, P. M., & Wehmeyer, M. L. (2013). The self-advocacy movement: Late modern times (1980 to present). In M. L. Wehmeyer (Ed.), *The story of intellectual disability: An evolution of meaning, understanding, and public perception* (pp. 233–227). Baltimore, MD/London/Sydney: Paul H. Brookes.

Ferretti, R. P. (1982). An analysis of passive memory in normal and mentally retarded persons. *Intelligence, 6*, 69–87.

Ferretti, R. P. (1989). Problem solving and strategy production in mentally retarded persons. *Research in Developmental Disabilities, 10*, 19–31.

Ferretti, R. P., & Belmont, J. M. (1983). Intelligence, adaptation, and problem solving. In K. T. Kernan, M. J. Begab, & R. B. Edgerton (Eds.), *Environments and behavior: The adaptation of mentally retarded persons* (pp. 55–69). Baltimore, MD: University Park Press.

Ferretti, R. P., & Butterfield, E. C. (1989). Intelligence as a correlate of children's problem solving. *American Journal on Mental Retardation, 93*, 424–433.

Ferretti, R. P., & Butterfield, E. C. (1992). Intelligence-related differences in the learning, maintenance, and transfer of problem-solving strategies. *Intelligence, 16*, 207–223.

Ferretti, R. P., & Cavalier, A. R. (1991). Constraints on the problems solving of persons with mental retardation. In N. W. Bray (Ed.), *International review of research in mental retardation* (Vol. 17, pp. 153–192). New York: Academic Press.

Ferretti, R. P., Cavalier, A. R., Murphy, M., & Murphy, R. (1993). The self-management of skills by persons with mental retardation. *Research in Developmental Disabilities, 14*, 189–205.

Galton, F. (1887). Supplementary notes on "Prehension" in idiots. *Mind, 12*, 79–82.

Hacker, D. J., Dunlosky, J., & Graesser, A. C. (2009). *Handbook of metacognition in education*. New York: Routledge.

Hull, C. L. (1952). *A behavior system; An introduction to behavior theory concerning the individual organism*. New Haven, CT: Yale University Press.

Kendall, C. R., Borkowski, J. G., & Cavanaugh, J. C. (1980). Metamemory and the transfer of an interrogative strategy by EMR children. *Intelligence, 4*, 255–270.

Mithaug, D. E. (1993). *Self-regulation theory: How optimal adjustment maximizes again*. Westport, CT: Praeger.

Newell, A., & Simon, H.A. (1972). *Human problem solving*. Englewood Cliffs, NJ: Prentice-Hall.

Pinkus, A. L., & Laughery, K. R. (1970). Recoding and grouping processes in short-term memory: Effects of subject-paced presentation. *Journal of Experimental Psychology, 85*, 335–341.

Powers, L. E., Geenen, S., Powers, J., Pommier-Satya, S., Turner, A., Dalton, L., … and other members of the Research Consortium to increase the Success of Youth in Foster Care. (2012). My Life: Effects of a longitudinal, randomized study of self-determination enhancement on the transition outcomes of youth in foster care and special education. *Children and Youth Services Review, 34*, 2179–2187.

Schneider, W., & Shiffrin, R. M. (1977). Controlled and automatic information processing: I. Detection, search, and attention. *Psychological Review, 84*, 1–66.

Shiffrin, R. M., & Schneider, W. (1977). Controlled and automatic information processing: II. Perceptual learning, automatic attending and a general theory. *Psychological Review, 84*, 127–190.

Spitz, H. H., & Borys, S. V. (1984). Depth of search: How far can the retarded search through an internally represented problem space? In P. H. Brooks, R. Sperber, & C. McCauley (Eds.), *Learning and cognition in the mentally retarded* (pp. 333–358). Hillsdale, NJ: Erlbaum.

Waugh, N. C., & Norman, D. A. (1965). Primary memory. *Journal of Experimental Psychology, 72*, 89–104.

Wehmeyer, M. L. (2001). Self-determination and mental retardation. In L. M. Glidden (Ed.), *International review of research and mental retardation* (Vol. 24, pp. 1–48). New York: Academic Press.

Wehmeyer, M. L. (2007). *Promoting self-determination in students with developmental disabilities*. New York/London: The Guilford Press.

Wehmeyer, M. L. (2013). *The story of intellectual disability: An evolution of meaning, understanding, and public perception*. Baltimore, MD/London/Sydney: Paul H. Brookes.

Wehmeyer, M. L. (2015). Framing for the future: Self-determination. *Remedial and Special Education, 36*, 20–23.

Wehmeyer, M. L., Agran, M., & Hughes, C. (1998). *Teaching self-determination to students with disabilities: Basic skills for successful transition*. Baltimore, MD: Paul H. Brookes.

Wehmeyer, M. L., Palmer, S. B., Lee, Y., Williams-Diehm, K., & Shogren, K. A. (2011). A randomized-trial evaluation of the effect of *Whose Future is it Anyway?* On self- determination. *Career Development for Exceptional Individuals, 34*, 45–56.

Wehmeyer, M. L., & Shalock, R. L. (2013). The parent movement: Late modern times (1950 to 1980). In M. L. Wehmeyer (Ed.), *The story of intellectual disability: An evolution of meaning, understanding, and public perception* (pp. 187–231). Baltimore, MD: Paul H. Brookes.

Part II

Philosophy of Care

5

Philosophy of Care

Maya Matheis

Philosophy of Care

Individuals with intellectual disabilities have been one of the most marginalized and disadvantaged groups within society. Historically, individuals with intellectual disability have been shunned, considered "mentally ill" or "deviant," and treated as less than human (Kanner, 1967; Karten, 2015; Wolfensberger, 1972). In the past century, considerable advances have been made to improve treatment and care for individuals with intellectual disability. The deinstitutionalization movement in Europe and North America has made significant strides in reducing the number of individuals with intellectual disability confined to residential facilities (Mansell & Ericsson, 2013). Our understanding of intellectual disability has evolved from a medical model, in which it is viewed as a "static and rather hopeless condition," to a developmental model, in which individuals are considered capable of learning and developing (Horejsi, 1975, p. 26). Attitudes and policies have shifted toward emphasizing social integration and inclusion for individuals with disabilities. This chapter discusses major philosophical shifts in the field of intellectual disability since the twentieth century, including reviews of concepts related to deinstitutionalization, normalization, social inclusion, self-determination, and person-centered planning.

Deinstitutionalization

Deinstitutionalization has been one of the most influential and important concepts in the field of intellectual disability in the past 50 years. The movement away from models of long-term residential institutional care toward community-based services is at varying stages around the world. Scandinavian countries have been at the forefront of the movement, with laws in Sweden and Norway protecting the right to community-based services (Tøssebro, 2004, 2016). Deinstitutionalization is in its late stages in the United States, the United Kingdom, Ireland, Canada, and Australia (Mansell, 2006; Mansell & Ericsson, 2013). In the United States, the number of individuals with intellectual disability or other developmental disabilities living in state institutions decreased from 131,345 in 1980 to 29,768 in 2016—a 77% reduction (Coucouvanis, Lakin, Prouty, & Webster, 2006; Larson et al., 2018). Deinstitutionalization is in its early stages, with community-based placements becoming more commonplace, in European countries such as Germany, Spain, Austria, Belgium, Italy, Greece, and Portugal (Beadle-Brown, Mansell, & Kozma, 2007; Buchner, 2009; Mansell & Ericsson, 2013;

M. Matheis, Ph.D. (✉)
MIND Institute, University of California Davis,
Sacramento, CA, USA

J. L. Matson (ed.), *Handbook of Intellectual Disabilities*, Autism and Child Psychopathology
Series, https://doi.org/10.1007/978-3-030-20843-1_5

Martinez-Leal et al., 2011), as well as countries in Asia such as Singapore, Japan, and South Korea (Jeevanandam, 2009). However, institutional care remains the norm in many countries across the world (Jeevanandam, 2009; Lin, Hsu, Kuo, & Lin, 2013; Lin & Lin, 2011; Martinez-Leal et al., 2011; Mercadante, Evans-Lacko, & Paula, 2009; Vann & Šiška, 2006).

The deinstitutionalization of services for individuals with intellectual disability and other developmental disabilities has had a very different process and timeline compared to the deinstitutionalization of residential facilities for individuals with mental illness (Landesman & Butterfield, 1987). The movement to dismantle mental asylums and residential state hospitals in Europe and North America began following World War II, as such facilities came under scrutiny for poor living conditions and treatment of patients (Fakhoury & Priebe, 2007; Paulson, 2012). In the following decades, the civil rights movement, changes in the conceptualization of mental illness, and advances in psychopharmacology contributed toward a shift in mental health service delivery (Paulson, 2012). The process of deinstitutionalizing services for individuals with developmental disabilities did not start until the late 1960s and early 1970s, with the publication of *Changing patterns in residential services for the mentally retarded* (Kugel & Wolfensberger, 1969) representing a significant change in the field's conceptualization of services (Landesman & Butterfield, 1987; Mansell & Ericsson, 2013). The movement was heavily influenced by normalization ideology, which posited that social integration would be in the best interest of individuals with intellectual disability (Culham & Nind, 2003; Landesman & Butterfield, 1987).

In the United States, the process of establishing alternatives to residential institutions was begun in a handful of states during the 1960s, including Connecticut, Nebraska, and California (Horejsi, 1975). However, state institutions didn't begin to close until the 1970s, with the majority of closures and development in community services occurring in the 1980s (Horejsi, 1975; Landesman & Butterfield, 1987; Mansell & Ericsson, 2013). The Developmentally Disabled Assistance and Bill of Rights Act (P.L. 94-103, , 1975), which introduced the requirement of protection and advocacy services for individuals with developmental disabilities, required that states develop plans to "support the establishment of community programs as alternatives to institutionalization" and to "eliminate inappropriate placement in institutions of persons with developmental disabilities" (89 Stat. 481).

Defining Deinstitutionalization

While a large focus within deinstitutionalization is the closure of long-term residential facilities for individuals with developmental disabilities, it also emphasizes the development and sustainment of alternative forms of care within community settings. Horejsi (1975) described deinstitutionalization as a conglomeration of four interrelated activities:

1. Preventing the admission of persons to institutional facilities by finding and developing alternative community methods of care and training
2. Returning to the community all institutional residents who have been prepared through programs of rehabilitation and training to function adequately in appropriate local settings
3. Establishing and maintaining a responsive residential environment which protects human and civil rights and which contributes to the expeditious return of the individual to normal community living whenever possible
4. Promoting public acceptance of retarded persons as neighbors, employees, and citizens possessing civil and human rights (p. 10)

This definition underscores the importance of community-based care, the continuing role that institutions and service systems play in caring for this population, and an emphasis on human rights within the deinstitutionalization process. In his model of deinstitutionalization, Horejsi (1975) described a comprehensive support system for individuals with developmental disabilities beyond residential services, including child development services, public education, family support services, and vocational services. He also called for the involvement of parent/family advocates as well as individuals with intellectual disability in designing and improving community-based programs, proposing that individuals with intellectual disability hold places on advisory boards. The exact configuration of services, Horejsi (1975) maintained, would need to be individualized to the needs of specific regions, stating that "deinstitutionalization does not occur in a vacuum. A whole host of economic, political, cultural, and social factors affect what can or cannot be done at a particular point in time in a particular area" (p. 40).

Horejsi (1975) also distinguished deinstitutionalization from institutional reform and decentralization, two similar but distinct concepts. He described "institutional reform" as referring to the improvement of institution-based care and "decentralization" as referring to the dissolution of large state facilities in place of smaller facilities dispersed across a region. As can be seen by these definitions, both institutional reform and decentralization retain the model of long-term residential institutions and lack the emphasis on community-based services that characterizes deinstitutionalization.

Associated Outcomes and Costs

A foundational premise of deinstitutionalization is that community-based services provide individuals with developmental disabilities with a better standard of living and improved functioning (Chowdhury & Benson, 2011; Mansell & Ericsson, 2013). As the process of deinstitutionalization has progressed around the world, research has examined the associated outcomes for individuals with intellectual disability. A comprehensive review of studies examining outcomes associated with deinstitutionalization published in English between 1997 and 2008 found that, compared to institutions, community-based services were associated with superior community participation, social networks, family contact, quality of life, adaptive behavior, self-determination/choice, and user/family satisfaction (Kozma, Mansell, & Beadle-Brown, 2009). This review also found that these findings were similar in different countries, across variations in service structures and socioeconomic factors. Chowdhury and Benson (2011) conducted a review of literature examining the specific construct of quality of life, with results indicating that community-based living following deinstitutionalization was associated with better quality of life for individuals with intellectual disability. Another comprehensive review examined health-related outcomes in 14 European Union countries, finding that late-stage deinstitutionalization was associated with the lower presence of certain health conditions (e.g., chronic bronchitis, osteoporosis, gastric ulcers) among individuals with intellectual disability compared to early-stage deinstitutionalization.

Individual factors have been found to influence outcomes related to deinstitutionalization. In their comprehensive review of deinstitutionalization research, Kozma et al. (2009) found that better adaptive skills are associated with better outcomes, particularly in relation to community participation, social networks, and self-determination. A general finding from the literature indicates that individuals with higher support needs, such as challenging behavior, experience worse outcomes (Kozma et al., 2009; Mansell, 2006). Individuals with challenging behavior have been found to be more likely to remain in institutional settings and to be re-institutionalized after the transition to community-based services (Beadle-Brown et al., 2007; Mansell, 2006).

As deinstitutionalization has progressed, research has also focused on comparing the associated costs of institutional and community-based services. Findings from the United States have consistently demonstrated that community-based services are less costly than state institutions (Campbell & Heal, 1995; Knobbe, Carey, Rhodes, & Horner, 1995; Stancliffe & Lakin, 1997). Research in the United Kingdom has found the cost of community-based services for individuals with intellectual disability to be higher compared to institutional care; however, the associated costs of community-based services were observed to be decreasing over time (Hallam et al., 2006).

Opposition and Challenges

Much of the opposition and resistance to deinstitutionalization stemmed from concerns regarding the transition in service delivery systems rather than disagreement with the concept of deinstitutionalization (Landesman & Butterfield, 1987; Mansell & Ericsson, 2013). During the early stages of deinstitutionalization in the United States, Scandinavia, and the United Kingdom, many parent and family advocates expressed concerns that alternative services were insufficient to meet the needs of their relatives with intellectual disability and that the transition in residential setting would have negative effects on their family member's functioning (Landesman & Butterfield, 1987; Mansell & Ericsson, 2013). Parent advocacy groups voiced concerns that individuals with developmental disabilities would be vulnerable in community settings and that community-based residences would have inadequate support and continuity of care (Landesman & Butterfield, 1987). In the decades following, parental and familial attitudes regarding deinstitutionalization have become more positive, though concerns still remain regarding the availability of alternative resources (Mansell & Ericsson, 2013). These concerns are also shaped by the changing political and economic climates in Europe and the United States, as fiscal conservatism has broadly resulted in the reduction of social services (Mansell & Ericsson, 2013).

Related to these concerns is de-differentiation, a common consequence of deinstitutionalization (Sandvin, 1996). De-differentiation describes "the loss of special, separate policies and service structures for people with intellectual disabilities and their replacement by general policies and structures" (Mansell, 2006, p. 11). The replacement of specialized services with those that serve a range of populations creates competition for these resources with other client groups, such as the elderly, individuals with physical disabilities, and individuals with mental illness. De-differentiation is also a result of the social model of disability, which perceives specialized services as discriminatory (Bickenbach, Chatterji, & Badley, 1999; Mansell, 2006). The combined effect is that the specific needs of individuals with intellectual disability may go unmet. As noted by Mansell (2006):

> A critical question for proponents of better-quality services for people with intellectual disabilities will be how they can achieve the benefits of social integration for individuals and for the whole group while at the same time retaining enough knowledge and recognition to ensure that special needs are noticed. (p. 13)

The challenges of deinstitutionalization lie thusly in the development and maintenance of a service system that simultaneously allows individuals with intellectual disabilities to be socially integrated within communities and that provides services specifically designed to address their needs and help them develop, while also ensuring the availability of sufficient resources.

Normalization

The ideology of normalization was a dominant philosophy in the field of intellectual disability for several decades and was a driving force behind deinstitutionalization. The concept of normalization originated in Scandinavia with efforts to reform services for individuals with intellectual disability in the late 1950s and 1960s (Bank-Mikkelsen, 1969; Nirje, 1970). The first definition of normalization was developed by Bank-Mikkelsen (1969), the head of the Danish Mental Retardation Service, who described it as "letting the mentally retarded obtain an existence as close to the normal as possible" (p. 227). This concept was further developed by Nirje (1970), who was the first to develop it into an ideology with accompanying principles and who applied it to policies pertaining to services for individuals with intellectual disability. His work emphasized providing individuals with disabilities equal rights and opportunities, defining the normalization principle as "making available to the mentally retarded patterns and conditions of everyday life which are as close as possible to the norms and patterns of the mainstream of society" (Nirje, 1969, p. 181). Nirje (1985) also introduced the concept of social integration, which he described as the opportunity "to be yourself—to be able and to be allowed to be yourself among others" (p. 67).

Wolfensberger (1972) later introduced normalization to the United States along with an accompanying set of guidelines for applying the ideology to a broad array of human services.

Wolfensberger's definition of normalization described it as the "utilization of means which are as culturally normative as possible, in order to establish and/or maintain personal behaviors and characteristics which are as culturally normative as possible" (Wolfensberger, 1972, p. 28). This definition clarifies "normal" to be relative to one's social context and also describes normalization as both a continuous process and a goal. By supporting individuals marginalized within society to adopt culturally typical behaviors, Wolfensberger (1972) argued that social stigma could be decreased, leading to increased social acceptance and social value. His model of human services and human management called for action within interpersonal interactions and at a societal level, with supports for both physical (e.g., location, access) and social (e.g., programming, public perception) integration within the community.

Criticism and Controversy

As pointed out by Landesman and Butterfield (1987) in their analysis of normalization, this ideology is built on the assumption that the environment plays a large role in an individual's functioning, with access to typical activities and settings increasing quality of life. When applied to service design and delivery, this philosophy promotes deinstitutionalization and community integration for living, employment, and recreational activities. While this ideology received widespread support and was deeply entrenched within the field of intellectual disabilities for some time (Culham & Nind, 2003; Landesman & Butterfield, 1987), it was not without its critics. Direct opposition to normalization centered upon concern that individuals with intellectual disability would be at risk in mainstream community settings and that they would benefit more from environments designed to be protective and nurturing (Landesman & Butterfield, 1987). Given that Wolfensberger's guidelines for implementing normalization action in service delivery outlined multiple levels of support, these early concerns were largely centered on the practical implementation of normalization and the risks of deinstitutionalization rather than the theoretical underpinnings of normalization.

Criticism of the ideology of normalization gained traction with the rise of the social model of disability (Culham & Nind, 2003). Within the social model of disability, disability is viewed as a social construct, as opposed to the medical model, in which disability is viewed as a deficit or defect within an individual (Bickenbach et al., 1999; Oliver, 1990). The social model of disability was embraced by disability advocates and civil rights activists, who argued that the difficulties experienced by individuals with disabilities are due to social attitudes about their abilities and needs. Critics questioned the expectation that individuals with intellectual disability should act "normal," raising concerns that normalization emphasized assimilation with societal standards, and placed the responsibility on individuals with disabilities to conform in order to receive social acceptance and respect (Culham & Nind, 2003; Northway, 2002; Perrin & Nirje, 1985; Shaddock & Zilber, 1991). Advocates argued that individuals with disabilities should embrace their differences, following in the footsteps of civil rights movements and empowerment strategies used by other marginalized groups. Culham and Nind (2003) noted that individuals with intellectual disabilities were one of the last marginalized social groups to which this consideration of human rights and empowerment of diversity has been applied and that the ideology of normalization insulated this population from empowerment. They commented that "while other groups have come to regard themselves as strong…the assimilation of normalization has led only to a begrudging acceptance for people with an intellectual disability" (Culham & Nind, 2003, p. 71).

Wolfensberger and Nirje held that these criticisms stemmed from "misconceptions" and "preconceived associations" of normalization (Perrin & Nirje, 1985; Wolfensberger, 1983). Perrin and Nirje (1985) commented that:

> Probably the most common misinterpretation of the normalisation principle is the mistaken belief that it means mentally handicapped people must be expected to, indeed be forced to, act 'normal,' ... normalisation does not mean normalcy; it does not mean that people should be normalised ... normalisation means the acceptance of a person with their handicap within 'normal society.' (pp. 69–70)

Wolfensberger (1983) attributed much of the controversy to the connotations invoked by the term "normalization," describing its nomenclature as "unfortunate" (p. 234). As such, he sought to distance his model from such criticism and to clarify its original intentions by discarding the term "normalization" and replacing it with the term "social role valorization."

Social Role Valorization Theory

Wolfensberger (1983) proposed that the term "social role valorization" (SRV) replace "normalization," believing that this new term better encapsulated his ideology and his applied model. He further contextualized the new term within the "recent insight that the most explicit and highest goal of normalization must be the creation, support, and defense of valued social roles for people who are at risk of social devaluation" (Wolfensberger, 1983, p. 234). In his work introducing the new term, Wolfensberger also re-emphasized the cultural relativity of his original definition of normalization and reiterated the elements of his model pertaining to actions directed at reducing social stigma and changing public perception of marginalized groups. Like his original conceptualization of normalization, Wolfensberger's SRV theory extended to all socially marginalized groups and was nonspecific to individuals with disabilities.

While Wolfensberger first introduced SRV as a refinement of an existing set of principles, it is largely viewed as a unique theory due to the extent to which it has since been developed (Cocks, 2001; Kumar & Singh, 2015; Lemay, 1995; Wolfensberger, 2000). Wolfensberger's later writing on SRV situates it within complex sociological theory about social roles and needs (Cocks, 2001; Kumar & Singh, 2015). The focus of SRV is on the socially devalued individual, who is likely to be socially excluded, treated poorly, and denied autonomy (Cocks, 2001; Lemay, 1995). As described by Wolfensberger (2000):

> The key premise of SRV is that people's welfare depends extensively on the social roles they occupy… This implies that in the case of people whose life situations are very bad, and whose bad situations are bound up with occupancy of devalued roles, then if the social roles they are seen as occupying can somehow be upgraded in the eyes of perceivers, their life conditions will usually improve. (p. 105)

Within this framework, service systems influence the way their clients are viewed by the larger community and thus are called upon to go beyond rehabilitation to address the devaluation experienced by these individuals. Two main processes work to place individuals in positive social roles with the goal of preventing and mitigating social devaluation: enhancing the social image of vulnerable groups and enhancing the competencies of vulnerable individuals (Cocks, 2001; Lemay, 1995; Wolfensberger, 2000). Critics of SRV note that this framework still places the burden on individuals with disabilities to engage in certain behaviors and to develop certain competencies in order to be accepted and valued by society (Culham & Nind, 2003; Kumar & Singh, 2015).

The Legacy of Normalization

Since its introduction, the ideology of normalization has continued to be refined and expanded, with its principles heavily influencing social and educational policy in Europe, the United States, and Australia. Normalization was a major impetus for deinstitutionalization in Europe and the United States, and its focus on social integration is at the foundation of many of the service structures aimed at supporting individuals with intellectual disability (Buell & Minnes, 2006; Landesman & Butterfield, 1987). Its emphasis on social integration and human rights for individuals with intellectual disability helped pave the way for philosophies of social inclusion, empowerment, and self-determination (Culham & Nind, 2003; Kumar & Singh, 2015).

Inclusion

The philosophy of inclusion became dominant in the field of intellectual and developmental disabilities in the late 1990s and early 2000s and is now a central part of our contemporary conceptualization of social policy (Power, 2013). Inclusion broadly describes increased participation in community-based activities and social networks (Abbott & McConkey, 2006). The major goals of inclusion for individuals with intellectual disability are to ensure that they "have full and equal access to health care, social roles, and relationships that are equal to their non-disabled peers" (Bollard, 2009, p. 5).

Social inclusion can trace its theoretical roots back to normalization, which laid the foundation for conceptualizing social integration for individuals with cognitive disabilities (Culham & Nind, 2003; Renzaglia, Karvonen, Drasgow, & Stoxen, 2003). However, inclusion differs from integration due to its emphasis on participation, as well as the foci of change. Within normalization, individuals are encouraged to adopt socially typical behaviors to gain access to community activities and social networks (Culham & Nind, 2003). The approach of inclusion places the responsibility on society to ensure that all individuals are able to participate, with inclusion conceptualized as a basic human right (Culham & Nind, 2003). The movement toward inclusion was influenced by growing awareness that individuals with intellectual disability continue to experience social isolation even when living in community settings, which has led to the description "asylum without walls" (Hall, 2005). This shift in thinking looked beyond integration to focus on creating an inclusive environment, rather than bringing individuals with intellectual disability into environments to which they must learn to adapt and conform (Culham & Nind, 2003; Renzaglia et al., 2003).

The Philosophy of Inclusion

While social inclusion has been conceptualized in a myriad of ways, most definitions emphasize meaningful interaction with the community, use of community facilities, and large social networks (Simplican, Leader, Kosciulek, & Leahy, 2015). Independent living (Duggan & Linehan, 2013), participation in economic systems, and employment (Hall, 2005; Hall & McGarrol, 2012) have been highlighted as important aspects of inclusion. Literature on inclusion has also emphasized the significance of "belonging" in a community, with belonging defined as "meaningful engagement and reciprocal relationships within local neighborhoods or networks between people with and without disabilities" (Power, 2013, p. 69).

An ecological model developed by Simplican et al. (2015) focuses on the functional aspects of social inclusion, conceptualizing it as the interaction between the domains of interpersonal relationships and community participation. These domains are theorized to interact and strengthen each other: by developing meaningful relationships with other people, community participation will increase, and, in turn, increased community participation will lead to the broadening of one's social network. Within this model, independent living and employment can be considered as factors that promote and facilitate the domains of interpersonal relationships and community participation, highlighting the ways in which various processes build upon each other to create social inclusion.

The primary aim of inclusive philosophy is to improve the lives of individuals marginalized and excluded from society. As described by Renzaglia et al. (2003), inclusion works to improve the quality of life of individuals with intellectual disabilities through three processes: "a) empowering individuals to have control over their own lives, b) providing individuals with the opportunity to select the lives of their choosing, and c) conferring individuals with the sociopolitical power to defend their choices" (p. 141). Inclusion also aims to better society as a whole by maximizing individual productivity and contribution through the process of making communities welcoming and accepting of differences (Renzaglia et al., 2003). The concept of "universal design," borrowed from architecture, refers to the creation of physical and social spaces designed to accommodate the needs of special populations (Renzaglia et al., 2003). By designing and planning for a diverse array of needs and abilities, inclusion aims to ensure universal participation.

Barriers and Challenges

The path to social inclusion for individuals with intellectual disabilities has been hindered by a number of social and structural barriers. A review of research examining environmental factors affecting community participation among individuals with intellectual disability identified lack of transportation and negative attitudes in the community as two major impediments toward participation, particularly in regard to participation in leisure activities (Verdonschot, de Witte, Reichrath, Buntinx, & Curfs, 2009). A qualitative study conducted by Abbott and McConkey (2006) surveyed individuals with intellectual disability on their perceived barriers to inclusion, identifying four main barriers: lack of necessary knowledge and skills (e.g., literacy, numeracy, knowledge of area), role of support staff and service managers (e.g., limited independence, short staffed), location of residence (e.g., lack of transport, few community activities/facilities in area), and community factors (e.g., negative attitudes, lack of advocacy/volunteer groups).

Limited opportunities for paid employment has also been highlighted as a significant barrier (Hall, 2005; Hall & McGarrol, 2012). Unwelcoming employment settings and shortage of services providing vocational training and/or support contribute to this, along with policies that disassociate social benefits and employment (Hall & McGarrol, 2012). These factors limit economic inclusion and choice-making, hindering participation in economic as well as social spheres. Hall and McGarrol (2012) have commented that our binary conceptualization of employment and care contributes toward the difficulty that individuals with intellectual disabilities experience in this regard and that reconceptualizing the contributions and positions for this population in the workplace will be necessary for inclusion. Similarly, Kittay (2001) has suggested that the emphasis in the literature on independent living creates a barrier to inclusion, as many individuals with intellectual disability require some degree of support and assistance, noting that "perhaps a way of including the severely intellectually impaired person and bestowing the dignity of citizenship is to interpret what it means to live independently, to be employed, and to be productive, in very broad terms" (p. 569).

Self-Determination

The concept of self-determination is prominent in multiple disciplines, including political science, philosophy, and social psychology (Wehmeyer, 1999). Broadly, self-determination refers to one's ability to determine one's own fate (McCorquodale, 1994; Quane, 1998). Many human and civil rights groups, including the United Nations, have declared self-determination a basic human right and consider it a precondition for other human rights (McCorquodale, 1994; Wehmeyer, 2005). Self-determination theory, which is concerned with understanding the motivations behind the behavior of individuals, became prominent in the area of personality psychology in the 1970s (Deci & Ryan, 2008). However, it wasn't until the 1990s that self-determination became an important construct in the fields of education and disability advocacy, following the funding of several projects aimed at promoting self-determination in children and youth with disabilities in the United States by the Office of Special Education Programs (Wehmeyer, 1999). Currently, self-determination is at the core of many policies related to education and disability.

The Philosophy of Self-Determination

Self-determination has been defined in numerous ways in disability and education literature, with models and definitions emphasizing the human rights aspect of the concept, personal choice, and empowerment (Malian & Nevin, 2002; Wehmeyer, 1999). Turnbull and Turnbull (2001) described self-determination as "living one's life consistent with one's own values, preferences, strengths, and needs and implicitly doing so free from external influences" (p. 58). Although several measures have been developed to measure constructs and skills related to self-determination, it itself is conceptualized as an unmeasurable heuristic used to describe behavior or a state characteristic (Wehmeyer, 2005). For individuals with intellectual disability, self-determination is an important component of social inclusion, as it allows them to navigate social environments and become active participants (Renzaglia et al., 2003).

Wehmeyer, Kelchner, and Richards (1996) developed a definitional framework for self-determination for individuals with intellectual disability through a combination of review of the relevant literature and interviews with self-advocates. This framework defines self-determination as "acting as the primary causal agent in one's life and making choices and decisions regarding one's quality of life free from undue external influence or interference" (Wehmeyer et al., 1996, p. 632). As such, self-determination describes when an individual brings about events in his or her life without an unacceptable level of interference from others. The model developed by Wehmeyer et al. (1996) also identified four essential characteristics of self-determination: autonomy, self-regulation (e.g., self-monitoring, self-reinforcement), psychological empowerment (i.e., perceived control), and self-realization (i.e., self-awareness of strengths and limitations).

Self-Determination for Individuals with Severe and Profound Intellectual Disability

Self-determination is often misinterpreted as being synonymous with independence or self-reliance, which limits the degree to which this construct can be applied to individuals with severe and profound intellectual disability, as individuals with severe cognitive impairments often require support in daily life activities as well as decision-making (Turnbull & Turnbull, 2001; Wehmeyer, 2005). A survey conducted with teachers in the United States found that teachers working with students with severe cognitive impairments considered self-determination to be significantly less important for future success than teachers working with students with mild impairments (Wehmeyer, 2005; Wehmeyer, Agran, & Hughes, 2000). Wehmeyer (2005) has commented that misperceptions in the field conflating self-determination with self-sufficiency and control have created a barrier to self-determination for individuals with severe disabilities. To address this issue, he developed a refined definition of self-determination to specify that "it refers to people acting volitionally, based on their own will" and that individuals with severe impairments can "be supported to act more volitionally" (p. 117).

Turnbull and Turnbull (2001) proposed a framework for conceptualizing the application of self-determination principles for individuals with significant cognitive disabilities. Their framework emphasizes the role of trusted allies, who often have an established relationship with the individual, in supporting individuals with severe disabilities in acting volitionally. Two steps are recommended: (1) individuals with severe impairments should develop an awareness of their functional limitations and build alliances with allies to help support them; and (2) allies should collaborate with individuals with disabilities to support them in decision-making and, when necessary, make decisions for them, taking into consideration their "values, preferences, strengths, and needs" if they were able to make the decision independently (Turnbull & Turnbull, 2001, p. 57).

Teaching Self-Determination

Self-determination has become an important aspect of education for individuals with intellectual disability. The special education system of the United States involves students with disabilities in the development of goals for their own individualized education programs (IEPs) and transition plans (Martin & Marshall, 1996; Wood, Karvonen, Test, Browder, & Algozzine, 2004). Educational policies for students with disabilities also emphasize the development of self-determination as a critical component of the educational process (Test, Karvonen, Wood, Browder, & Algozzine, 2000; Wehmeyer, 1999).

Self-determination is understood as a trait that emerges through interactions with the environment (Malian & Nevin, 2002). As such, self-determination is considered teachable (Malian & Nevin, 2002; Martin & Marshall, 1996;Renzaglia et al., 2003 ; Test et al., 2000). Wehmeyer (1999) developed a function model of self-determination to guide interventions aimed at promoting self-determination for individuals with and without disabilities. This model identified the following component elements of self-determined behavior: choice-making, decision-making, and problem-solving skills; goal-setting and attainment skills; self-observation, self-evaluation, self-reinforcement, and self-instruction skills; self-advocacy and leadership skills; internal locus of control; positive attributions of efficacy and outcome expectancy; and self-awareness and self-knowledge (Wehmeyer, 1999).

The development of self-determination should be a lifelong process, with instruction beginning at young ages and following a developmental trajectory (Renzaglia et al., 2003; Test et al., 2000). Numerous instructional strategies and materials have been developed to support educators in teaching students skills related to self-determination (Renzaglia et al., 2003; Test et al., 2000; Wehmeyer, 1999). The most common method of instruction in teaching self-determination is direct instruction, in which a skill is introduced, practiced, and then applied (Renzaglia et al., 2003). Curricula have been developed for individuals with disabilities across the lifespan, as well as measures to assess the effectiveness of such curricula. In selecting a curriculum to teach self-determination, Test et al. (2000) recommend reviewing materials for fit in regard to intended audience, relevancy of skills, prerequisite skills, ease of use, types of materials, supporting evidence, and time/financial cost. Generalization of skills to natural environments is a particularly important aspect of instruction, as this is necessary for skills to manifest into self-determined behavior. In working toward the goal of generalization, educators should aim to maximize opportunities to learn and practice skills.

Self-Determination and Inclusion

For individuals with intellectual disability, self-determination is necessary for achieving full inclusion. Components of self-determination, such as problem-solving and decision-marking skills, allow individuals to navigate the environment and actively participate in their communities. Self-advocacy is also critical, as it allows individuals with disabilities to communicate their needs as individuals and as a group. Unlike among other historically marginalized groups, self-advocates have largely been absent from the movement toward social inclusion for individuals with intellectual disabilities (Abbott & McConkey, 2006). While individuals with cognitive impairments may require supports, qualitative research has demonstrated that they have insight into their experiences of exclusion and their needs (Abbott & McConkey, 2006). Educators, caregivers, and service providers can support individuals with intellectual disabilities in developing self-advocacy skills and by promoting opportunities for decision-making and leadership. Creating environments that are receptive toward active participation of individuals with intellectual disability and that provide opportunities for self-determined behavior is necessary in working toward inclusion (Renzaglia et al., 2003).

Person-Centered Planning

Person-centered planning (PCP) describes an approach to assisting individuals with disabilities in planning their futures and organizing their support structures (Clark, Knab, & Kincaid, 2005; Sanderson, 2000). The process of PCP aims to empower individuals with disabilities by including them in discussions and decision-making pertaining to their care, with the main goal to "aid an individual in developing meaningful life goals based on his or her strengths and talents, utilizing individual, natural, and creative supports and services" (Clark et al., 2005, p. 429). The approach of PCP developed in the 1980s out of the normalization movement within the United States and was quickly embraced within the United Kingdom (Clark et al., 2005; Holburn, 2002). PCP has become widespread in the field of intellectual and developmental disabilities, with educational and service policies in the United States, the United Kingdom, and Australia requiring PCP practices (Holburn, 2002; Ratti et al., 2016).

The Philosophy of PCP

In its development, PCP was greatly informed by the social inclusion movement and theories pertaining to self-determination (Holburn, 2002; Sanderson, 2000). The central tenant of PCP is that an individual is viewed as a person first and foremost and that his or her diagnosis, disability, or areas of weakness are not a focus of plan development (Clark et al., 2005; Holburn, 2002; Sanderson, 2000). Emphasis is placed on personal choice and empowering individuals to envision their desired future. Self-advocacy is developed by encouraging individuals with disabilities to voice their questions and interests. While PCP is focused on planning for the future, the process of developing goals inherently involves the evaluation of current quality of life and conditions, giving individuals with disabilities opportunities to address their unmet needs. The person-centered and strength-based aspects of PCP result in an approach that is focused on individual capacities and possibilities rather than deficits and available service supports.

Components of PCP

The term PCP is used to describe a broad approach rather than a specific technique. Many manualized interventions based on the PCP framework have been developed with widespread implementation, including Personal Futures Planning (Mount, 1994), Planning Alternative Tomorrows with Hope (PATH; Pearpoint, O'Brien, & Forest, 1993), the McGill Action Planning System (MAPS; Vandercook, York, & Forest, 1989), Individual Service Design (O'Brien & Lovett, 1993), Essential Lifestyle Planning (Smull & Harrison, 1992), and Whole Life Planning (Butterworth et al., 1993). These various interventions have been designed for application within specific organizational structures but share the overarching structure and philosophy of PCP.

The basic process of PCP involves the formation of a team consisting of people committed to the well-being of the individual (e.g., family members, staff, teachers, community members), encouraging the individual to describe the life he or she wants, and the development of a plan with short- and long-term goals in collaboration with the team and the individual (Clark et al., 2005; Holburn, 2002). Sanderson (2000) identified five key features of PCP:

1. The person is at the center.
2. Family members and friends are partners in planning.
3. The plan reflects what is important to the person, their capacities, and the support they require.
4. The plan results in actions that are about life, not just services, and reflect what is possible, not just what is available.
5. The plan results in ongoing listening, learning, and further action (pp. 3–7).

These five features emphasize the importance of taking a strengths-based and community-centered approach to developing an individual's plan. To promote the inclusion of individuals with intellectual disability in the planning process, PCP commonly involves the replacement of professional jargon with everyday language as well as pictures and graphic displays (Clark et al., 2005).

Challenges and Evidence

Empirical evaluation of PCP is complicated by the broadness of the approach, implementation variations, and challenges in measuring related constructs (Holburn, 2002). A recent systematic review found that while there are discrepancies across research studies, there is some evidence to indicate that PCP has a positive impact on outcomes across the lifespan (Ratti et al., 2016). PCP was found to have significant positive impact on community participation, as well as moderate positive impact on quality of life, participation in activities, and everyday choice-making. However, the review found no evidence that PCP increased self-determination, improved adaptive functioning, or increased decision-making related to care. These findings suggest that while evidence supporting PCP is limited, it has been demonstrated to help individuals with intellectual disability in certain domains. Further research is necessary to examine long-term and large-scale outcomes.

Some have voiced concerns that PCP is simply a "paper planning exercise" that has little impact on outcomes for individuals with intellectual disabilities (Claes, Van Hove, Vandevelde, van Loon, & Schalock, 2010; Ratti et al., 2016). Researchers have noted the tendency for PCP practices to fade after initial plan development, without updates or progress monitoring (Holburn, 2002; Menchetti & Garcia, 2003; Ratti et al., 2016). The implementation gap is likely to be larger in contexts lacking sufficient resources. Kaehne and Beyer (2014) noted that, in order for PCP to be successful, it must be embraced at the institutional level by all stakeholders. Further attention and research should be focused on PCP implementation fidelity, individual and programmatic evaluation, and plan progress monitoring in an effort to decrease the implementation gap.

Conclusion

Since the 1950s, considerable shifts have occurred in our conceptualization of care for individuals with intellectual disability. The principle of normalization paved the way for the deinstitutionalization and social inclusion movements. Philosophies of self-determination and person-centered planning currently have widespread influence on service structures related to education, employment, and community-based supports. Despite these advances, individuals with intellectual disability still face considerable stigma, prejudice, and social ostracization. Institutionalization remains a reality for many individuals with intellectual disability across the globe, including in the United States. Policies and interventions designed to promote social inclusion face many structural and social barriers. Continued effort is needed to advocate for the rights of individuals with intellectual disabilities, to empower and support self-advocates, and to refine our strategies of creating a society in which individuals with intellectual disabilities are welcomed, valued, and treated with respect.

References

Abbott, S., & McConkey, R. (2006). The barriers to social inclusion as perceived by people with intellectual disabilities. *Journal of Intellectual Disabilities, 10*(3), 275–287. https://doi.org/10.1177/1744629506067618

Bank-Mikkelsen, N. E. (1969). A metropolitan area in Denmark: Copenhagen. In R. Kugel & W. Wolfensberger (Eds.), *Changing patterns in residential services for the mentally retarded* (pp. 227–254). Washington, DC: President's Committee on Mental Retardation.

Beadle-Brown, J., Mansell, J., & Kozma, A. (2007). Deinstitutionalization in intellectual disabilities. *Current Opinion in Psychiatry, 20*(5), 437. https://doi.org/10.1097/YCO.0b013e32827b14ab

Bickenbach, J. E., Chatterji, S., & Badley, E. M. (1999). Models of disablement, universalism and the International Classification of Impairments, Disabilities and Handicaps. *Social Science, 48*(9), 1173–1187.

Bollard, M. R. (2009). *Intellectual disability and social inclusion: A critical review*. New York: Elsevier Health Sciences.

Buchner, T. (2009). Deinstitutionalisation and community living for people with intellectual disabilities in Austria: History, policies, implementation and research. *Tizard Learning Disability Review, 14*(1), 4–13. https://doi.org/10.1108/13595474200900002

Buell, K., & Minnes, P. M. (2006). An acculturation perspective on deinstitutionalization and service delivery. *Journal on Developmental Disabilities*, 7–17.

Butterworth, J., Hagner, H., Heikkinen, B., Faris, S., DeMello, S., & McDonough, K. (1993). *Whole Life Planning: A guide for organizers and facilitators*. Boston, MA: Training and Research Institute for People with Learning Disabilities.

Campbell, E. M., & Heal, L. W. (1995). Prediction of cost, rates, and staffing by provider and client characteristics. *American Journal on Mental Retardation, 100*(1), 17–35.

Chowdhury, M., & Benson, B. A. (2011). Deinstitutionalization and quality of life of individuals with intellectual disability: A review of the international literature. *Journal of Policy and Practice in Intellectual Disabilities, 8*(4), 256–265.

Claes, C., Van Hove, G., Vandevelde, S., van Loon, J., & Schalock, R. L. (2010). Person-centered planning: Analysis of research and effectiveness. *Intellectual and Developmental Disabilities, 48*(6), 432–453. https://doi.org/10.1352/1934-9556-48.6.432

Clark, H. B., Knab, J. T., & Kincaid, D. (2005). Person-centered planning. In *Encyclopedia of behavior modification and cognitive behavior therapy* (Vol. 1–3, pp. 429–430). Thousand Oaks: SAGE Publications, Inc. https://doi.org/10.4135/9781412950534

Cocks, E. (2001). Normalisation and social role valorisation: Guidance for human service development. *Hong Kong Journal of Psychiatry, 11*(1), 12–16.

Coucouvanis, K., Lakin, K. C., Prouty, R., & Webster, A. (2006). Reductions continue in average daily populations of large state facilities; Nearly 70% decrease between 1980 and 2005. *Mental Retardation, 44*(3), 235–238. https://doi.org/10.1352/0047-6765(2006)44[235:RCIADP]2.0.CO;2

Culham, A., & Nind, M. (2003). Deconstructing normalisation: Clearing the way for inclusion. *Journal of Intellectual & Developmental Disability, 28*(1), 65–78. https://doi.org/10.1080/1366825031000086902

Deci, E. L., & Ryan, R. M. (2008). Self-determination theory: A macrotheory of human motivation, development, and health. *Canadian Psychology/Psychologie Canadienne, 49*(3), 182–185. https://doi.org/10.1037/a0012801

Developmentally Disabled Assistance and Bill of Rights Act, Pub. L. No. 94–103 (1975).

Duggan, C., & Linehan, C. (2013). The role of 'natural supports' in promoting independent living for people with disabilities; a review of existing literature. *British Journal of Learning Disabilities, 41*(3), 199–207. https://doi.org/10.1111/bld.12040

Fakhoury, W., & Priebe, S. (2007). Deinstitutionalization and reinstitutionalization: Major changes in the provision of mental healthcare. *Psychiatry, 6*(8), 313–316. https://doi.org/10.1016/j.mppsy.2007.05.008

Hall, E. (2005). The entangled geographies of social exclusion/inclusion for people with learning disabilities. *Health & Place, 11*(2), 107–115. https://doi.org/10.1016/j.healthplace.2004.10.007

Hall, E., & McGarrol, S. (2012). Bridging the gap between employment and social care for people with learning disabilities: Local Area Co-ordination and in-between spaces of social inclusion. *Geoforum, 43*(6), 1276–1286. https://doi.org/10.1016/j.geoforum.2012.03.015

Hallam, A., Beecham, J., Knapp, M., Carpenter, J., Cambridge, P., Forrester-Jones, R., … Wooff, D. (2006). Service use and costs of support 12 years after leaving hospital. *Journal of Applied Research in Intellectual Disabilities, 19*(4), 296–308. https://doi.org/10.1111/j.1468-3148.2006.00278.x

Holburn, S. (2002). How science can evaluate and enhance person-centered planning. *Research and Practice for Persons with Severe Disabilities, 27*(4), 250–260. https://doi.org/10.2511/rpsd.27.4.250

Horejsi, C. R. (1975). Deinstitutionalization and the development of community based services for the mentally retarded: An overview of concepts and issues. Retrieved from https://eric.ed.gov/?id=ED112606

Jeevanandam, L. (2009). Perspectives of intellectual disability in Asia: epidemiology, policy, and services for children and adults. *Current Opinion in Psychiatry, 22*(5), 462. https://doi.org/10.1097/YCO.0b013e32832ec056

Kaehne, A., & Beyer, S. (2014). Person-centred reviews as a mechanism for planning the post-school transition of young people with intellectual disability: Person-centred transition reviews. *Journal of Intellectual Disability Research, 58*(7), 603–613. https://doi.org/10.1111/jir.12058

Kanner, L. (1967). Medicine in the history of mental retardation: 1800–1965. *American Journal of Mental Deficiency, 72*(2), 165–170.

Karten, T. J. (2015). Historical background of disabilities. In *Embracing disabilities in the classroom: Strategies to maximize students assets* (pp. 2–32). New York: Skyhorse Publishing, Inc..

Kittay, E. F. (2001). When caring is just and justice is caring: Justice and mental retardation. *Public Culture, 13*(3), 557–579.

Knobbe, C. A., Carey, S. P., Rhodes, L., & Horner, R. H. (1995). Benefit-cost analysis of community residential versus institutional services for adults with severe mental retardation and challenging behaviors. *American Journal on Mental Retardation, 99*(5), 533–541.

Kozma, A., Mansell, J., & Beadle-Brown, J. (2009). Outcomes in different residential settings for people with intellectual disability: A systematic review. *American Journal on Intellectual and Developmental Disabilities, 114*(3), 193–222. https://doi.org/10.1352/1944-7558-114.3.193

Kugel, R. B., & Wolfensberger, W. (Eds.). (1969). *Changing patterns in residential services for the mentally retarded*. Washington, DC: President's Committee on Mental Retardation.

Kumar, D. A., & Singh, D. R. R. (2015). Normalization vs. social role valorization: Similar or different? *International Journal of Special Education, 30*(3), 1–7.

Landesman, S., & Butterfield, E. C. (1987). Normalization and deinstitutionalization of mentally retarded individuals. *American Psychologist, 42*(8), 809–816.

Larson, S. A., Eschenbacher, H. J., Anderson, L. L., Taylor, B., Pettingell, S., Sowers, M., & Bourne, M. L. (2018). *In-home and residential long-term supports and services for persons with intellectual or developmental disabilities: Status and trends through 2016*. Minneapolis, MN: University of Minnesota, Research and Training Center on Community Living, Institute on Community Integration.

Lemay, R. (1995). Social role valorization theory and the principle of normalization. In *Encyclopedia of disability and rehabilitation* (pp. 515–521). New York: Simon & Schuster Macmillan.

Lin, L.-P., Hsu, S.-W., Kuo, F.-Y., & Lin, J.-D. (2013). Geographic differences in social-welfare oriented institutional care for people with disabilities in Taiwan, 2002–2009. *Research in Autism Spectrum Disorders, 7*(1), 43–48. https://doi.org/10.1016/j.rasd.2012.07.008

Lin, L.-P., & Lin, J.-D. (2011). Perspectives on intellectual disability in Taiwan: Epidemiology, policy and services for children and adults. *Current Opinion in Psychiatry, 24*(5), 413–418. https://doi.org/10.1097/YCO.0b013e328346176e

Malian, I., & Nevin, A. (2002). A review of self-determination literature: Implications for practitioners. *Remedial and Special Education, 23*(2), 68–74. https://doi.org/10.1177/074193250202300202

Mansell, J. (2006). Deinstitutionalisation and community living: Progress, problems and priorities. *Journal of Intellectual & Developmental Disability, 31*(2), 65–76. https://doi.org/10.1080/13668250600686726

Mansell, J., & Ericsson, K. (2013). *Deinstitutionalization and community living: Intellectual disability services in Britain, Scandinavia and the USA*. London: Chapman and Hall.

Martin, J. E., & Marshall, L. H. (1996). Infusing self-determination instruction into the IEP and transition process. In *Self-determination across the lifespan: Independence and choice for people with disabilities*. Baltimore, MD: Paul H. Brookes. Retrieved from http://journals.sagepub.com/doi/pdf/10.1177/088572889301600105

Martinez-Leal, R., Salvador-Carulla, L., Linehan, C., Walsh, P., Weber, G., Van Hove, G., … Kerr, M. (2011). The impact of living arrangements and deinstitutionalisation in the health status of persons with intellectual disability in Europe. *Journal of Intellectual Disability Research, 55*(9), 858–872.

McCorquodale, R. (1994). Self-determination: A human rights approach. *International & Comparative Law Quarterly, 43*(4), 857–885. https://doi.org/10.1093/iclqaj/43.4.857

Menchetti, B. M., & Garcia, L. A. (2003). Personal and employment outcomes of person-centered career planning. *Education and Training in Developmental Disabilities, 38*(2), 145–156.

Mercadante, M. T., Evans-Lacko, S., & Paula, C. S. (2009). Perspectives of intellectual disability in Latin American countries: Epidemiology, policy, and services for children and adults. *Current Opinion in Psychiatry, 22*(5), 469. https://doi.org/10.1097/YCO.0b013e32832eb8c6

Mount, B. (1994). Benefits and limitations of Personal Futures Planning. In V. J. Bradley, J. W. Ashbaugh, & B. C. Blaney (Eds.), *Creating individual supports for people with developmental disabilities: A mandate for change at many levels* (pp. 97–108). Baltimore, MD: Paul H. Brookes.

Nirje, B. (1969). The normalization principle and its human management implications. In R. Kugel & W. Wolfensberger (Eds.), *Changing patterns in residential services for the mentally retarded* (pp. 179–195). Washington, DC: President's Committee on Mental Retardation.

Nirje, B. (1970). The normalisation principle-implications and comments. *Journal of Mental Subnormality, 16*, 62–70.

Nirje, B. (1985). The basis and logic of the normalization principle. *Australia and New Zealand Journal of Developmental Disabilities, 11*(2), 65–68. https://doi.org/10.3109/13668258509008747

Northway, R. (2002). Integration and inclusion: Illusion or progress in services for disabled people? *Social Policy & Administration, 31*(2), 157–172. https://doi.org/10.1111/1467-9515.00046

O'Brien, J., & Lovett, H. (1993). Finding a way toward everyday lives: The contribution of person centered planning. Retrieved from https://eric.ed.gov/?id=ED356596

Oliver, M. (1990). *The politics of disablement: A sociological approach*. Basingstoke: Palgrave Macmillan.

Paulson, G. (2012). *Closing the asylums: Causes and consequences of the deinstitutionalization movement*. Jefferson, NC: McFarland & Co. Publishers.

Pearpoint, J., O'Brien, J., & Forest, M. (1993). *PATH: A workbook for planning positive possible futures and planning alternative tomorrows with hope for schools, organizations, businesses, and families* (2nd ed.). Toronto: Inclusion Press.

Perrin, B., & Nirje, B. (1985). Setting the record straight: A critique of some frequent misconceptions of the normalization principle. *Australia and New Zealand Journal of Developmental Disabilities, 11*(2), 69–74. https://doi.org/10.3109/13668258509008748

Power, A. (2013). Making space for belonging: Critical reflections on the implementation of personalised adult social care under the veil of meaningful inclusion. *Social Science & Medicine, 88*, 68–75. https://doi.org/10.1016/j.socscimed.2013.04.008

Quane, H. (1998). The United Nations and the evolving right to self-determination. *International & Comparative Law Quarterly, 47*(3), 537–572. https://doi.org/10.1017/S0020589300062175

Ratti, V., Hassiotis, A., Crabtree, J., Deb, S., Gallagher, P., & Unwin, G. (2016). The effectiveness of person-centred planning for people with intellectual disabilities: A systematic review. *Research in Developmental Disabilities, 57*, 63–84. https://doi.org/10.1016/j.ridd.2016.06.015

Renzaglia, A., Karvonen, M., Dragsow, E., & Stoxen, C. C. (2003). Promoting a lifetime of inclusion. *Focus on Autism and Other Developmental Disabilities, 18*(3), 140–149. https://doi.org/10.1177/10883576030180030201

Sanderson, H. (2000). *Person Centred Planning*. York: Joseph Rowntree Foundation.

Sandvin, J. (1996). The transition to community services in Norway. *Deinstitutionalization and Community Living* (pp. 185–196). Springer, Boston, MA. https://doi.org/10.1007/978-1-4899-4517-4_12

Shaddock, A. J., & Zilber, D. (1991). Current service ideologies and responses to challenging behaviour: Social role valorization or vaporization? *Australia and New Zealand Journal of Developmental Disabilities, 17*(2), 169–175. https://doi.org/10.1080/07263869100034381

Simplican, S. C., Leader, G., Kosciulek, J., & Leahy, M. (2015). Defining social inclusion of people with intellectual and developmental disabilities: An ecological model of social networks and community participation. *Research in Developmental Disabilities, 38*, 18–29. https://doi.org/10.1016/j.ridd.2014.10.008

Smull, M. W., & Harrison, S. B. (1992). *Supporting people with severe retardation in the community*. Alexandria, VA: National Association of State Mental Retardation Program Directors.

Stancliffe, R. J., & Lakin, K. C. (1997). Analysis of expenditures and outcomes of residential alternatives for persons with developmental disabilities. *American Journal on Mental Retardation, 102*(6), 552–568. https://doi.org/10.1352/0895-8017(1998)1022.0.CO;2

Test, D. W., Karvonen, M., Wood, W. M., Browder, D., & Algozzine, B. (2000). Choosing a self-determination curriculum: Plan for the future. *TEACHING Exceptional Children, 33*(2), 48–54. https://doi.org/10.1177/004005990003300207

Tøssebro, J. (2004). Policy evaluation and national surveys of the living conditions of people with intellectual disabilities in Norway. *Journal of Intellectual Disability Research, 48*(4), 486–507.

Tøssebro, J. (2016). Scandinavian disability policy: From deinstitutionalisation to non-discrimination and beyond. *ALTER - European Journal of Disability Research/Revue Européenne de Recherche Sur Le Handicap, 10*(2), 111–123. https://doi.org/10.1016/j.alter.2016.03.003

Turnbull, A., & Turnbull, R. (2001). Self-determination for individuals with significant cognitive disabilities and their families. *Journal of the Association for Persons with Severe Handicaps (JASH), 26*(1), 56–62.

Vandercook, T., York, J., & Forest, M. (1989). The McGill Action Planning System (MAPS): A strategy for building the vision. *Journal of the Association for Persons with Severe Handicaps, 14*(3), 205–215. https://doi.org/10.1177/154079698901400306

Vann, B. H., & Šiška, J. (2006). From 'cage beds' to inclusion: The long road for individuals with intellectual disability in the Czech Republic. *Disability & Society, 21*(5), 425–439. https://doi.org/10.1080/09687590600785811

Verdonschot, M. M. L., de Witte, L. P., Reichrath, E., Buntinx, W. H. E., & Curfs, L. M. G. (2009). Community participation of people with an intellectual disability: a review of empirical findings. *Journal of Intellectual Disability Research, 53*(4), 303–318. https://doi.org/10.1111/j.1365-2788.2008.01144.x

Wehmeyer, M. L. (1999). A functional model of self-determination: Describing development and implementing instruction. *Focus on Autism and Other Developmental Disabilities, 14*(1), 53–61. https://doi.org/10.1177/108835769901400107

Wehmeyer, M. L. (2005). Self-determination and individuals with severe disabilities: Re-examining meanings and misinterpretations. *Research and Practice for Persons with Severe Disabilities, 30*(3), 113–120. https://doi.org/10.2511/rpsd.30.3.113

Wehmeyer, M. L., Agran, M., & Hughes, C. (2000). A national survey of teachers' promotion of self-determination and student-directed learning. *The Journal of Special Education, 34*(2), 58–68. https://doi.org/10.1177/002246690003400201

Wehmeyer, M. L., Kelchner, K., & Richards, S. (1996). Essential characteristics of self-determined behavior of individuals with mental retardation. *American Journal on Mental Retardation, 100*(6), 632–642.

Wolfensberger, W. (1972). *The principle of normalization in human services*. Toronto, ON: National Institute on Mental Retardation.

Wolfensberger, W. (1983). Social role valorization: A proposed new term for the principle of normalization. *Mental Retardation, 21*(6), 234–239.

Wolfensberger, W. (2000). A brief overview of social role valorization. *Mental Retardation, 38*(2), 105–123. https://doi.org/10.1352/0047-6765(2000)0382.0.CO;2

Wood, W. M., Karvonen, M., Test, D. W., Browder, D., & Algozzine, B. (2004). Promoting student self-determination skills in IEP planning. *TEACHING Exceptional Children, 36*(3), 8–16. https://doi.org/10.1177/004005990403600301

6

Quality of Life

Haris Memisevic and Mirjana Djordjevic

Introduction

Quality of life (QOL) has become a major topic of many scientific disciplines in the last couple of decades. The concept of QOL has been studied extensively from many perspectives and within many scientific fields such as medicine, economics, and the social sciences (Cummins, 2005). As an indication of the scientific interest in QOL, we will mention that there were (up to December 31, 2017) 74,069 documents in the SCOPUS database that contained the term QOL in its title. Out of this number, there were 59,234 scientific articles, 5610 review articles, and 926 book chapters. In Fig. 6.1, we present the number of articles containing QOL in their titles in the last 5 years.

There are even three journals indexed in the Web of Science that, in its title, have the words QOL. In addition to this, there are numerous books and monographs on the QOL topic. From all this, it is quite obvious that the interest in this topic has not diminished.

Increased interest in QOL research can also be reflected in the number of theses containing QOL in their titles. By examining the Open Access Theses and Dissertations database, for the period of 2014–2017, there were 1,254,251 defended theses (PhD, MS, and MA) in different languages (English, Portuguese, Spanish, Swedish, Japanese, Finnish, French, German, Czech, Hungarian, Greek, Norwegian, etc.) and within different departments (psychology, mechanical engineering, chemistry, education, electrical engineering, physics English, computer science, electrical and computer engineering, history, medicine, economics , civil engineering, sociology).

The topic of QOL has been investigated widely in the context of human development. For example, how does QOL change through different age stages? It has also been studied cross-culturally. Are there differences in the QOL among nations and cultures? Next, QOL has been compared across disability categories (Memisevic, Hadzic, Ibralic Biscevic, & Mujkanovic, 2017). Obviously the topic is very broad, and many approaches and perspectives can be taken in studying the QOL.

In writing this chapter, the authors were faced with many dilemmas. What should be our main focus in this chapter? QOL in general, as it applies to all human beings and universally conceptualized, different parts of QOL? Or should we write about it specifically in terms of QOL for people with intellectual disability? Although the major part of QOL is universal or universally

---

H. Memisevic (✉)
Faculty of Educational Sciences, University of Sarajevo, Sarajevo, Bosnia and Herzegovina

M. Djordjevic
Faculty of Special Education and Rehabilitation, University of Belgrade, Belgrade, Serbia

J. L. Matson (ed.), *Handbook of Intellectual Disabilities*, Autism and Child Psychopathology Series, https://doi.org/10.1007/978-3-030-20843-1_6

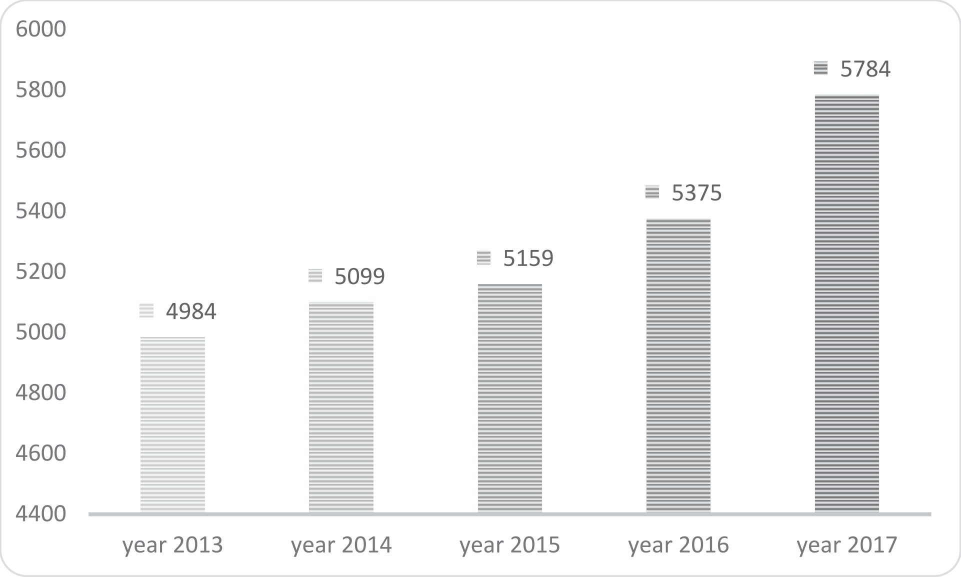


Fig. 6.1 Number of documents indexed in SCOPUS containing quality of life in their titles in the last 5 years

applied, there are, however, some aspects of it that pertain to people with disabilities only. Further on, there are some issues that pertain only to people with disabilities in certain parts of the world. Thus, we attempted to cover as many different topics as possible within this chapter, with the main focus on universal dimensions of QOL in people with ID and its general principles regardless of culture and identity factors

There is almost no medical condition, disability, nor group for which there is no QOL study. This voluminous work in the QOL field best reflects its importance in human life. QOL is a shared goal, one we would all like to improve, however elusive the goal may be. The field of intellectual disability (ID) was not spared from this increased general interest in the QOL concept.

In relation to the field of ID, the concept of QOL was embraced during the 1980s as a sensitizing notion and an overarching principle for service delivery (Schalock, 2000). In fact, the relationship between QOL and ID is a reciprocal one. Schalock, Verdugo, and Gomez (2017) propose that QOL has affected the field of intellectual disability in five different ways. It fostered the development of individualized supports, evidence-based practices, personal outcomes, quality improvement, and a catalyst for organization and system-level transformation. Given the fact that the QOL concept changed with time, it is unsurprising that it has not always been used for benefitting people with ID. For example, not so long ago, the very concept of QOL was used in argumentation to deprive persons with ID some basic rights to people with intellectual disability. There were several court cases in the United States where fundamental rights to life were denied to people with ID on the grounds of QOL. All this led to Baby Doe Law that stated that a child's quality of life is not a valid reason for withholding medical care.

Unfortunately, the issue of human rights still dominates the field of ID in many developing countries. As an example of this, we can take the numbers of institutionalized persons with ID in developing countries or the issue of legal capacity; in some countries, it is still a common practice to deprive legal capacity to people with ID. As we can see, many issues that people with ID are facing are region-specific. However, there are many universal topics that are relevant to people with ID throughout the world. All individuals with ID need to have the access to appropriate health services, support services, rehabilitation, education, employment, recreation, etc. Obviously there is a hierarchical nature of QOL dimensions. For example, before any talk on support services, people with ID need to have their basic human rights fulfilled, such as choosing their residence. Much research has been devoted to etic (universal) and emic (culture-bound) properties of the QOL concept. In relation to people with ID, besides having in mind etic and emic properties of the construct, we also need to consider disability-bound factors. Social support, social integration, internal control, autonomy, self-determination, and independent decision-making are all factors that need to be taken into account when talking about the QOL in people with ID (Schalock, 2004a, 2004b). Identity factors such as age, gender, etiology, and level of intellectual disability are yet another group of factors that might have an impact on the QOL.

Human rights movements and conventions stemming from their work have a significant role in improving the QOL. Currently, there are more than 40 worldwide treaties and conventions dealing with the human rights of people with disabilities (Schalock & Verdugo, 2012). One of the most important conventions is certainly the UN Convention on the Rights of Persons with Disabilities (UNCRPD, 2006). The UNCRPD is currently signed by 153 states. Given its role in promoting the rights of people with disabilities, it definitely warrants a short description. The objective of the UNCRPD, as stated in the Article 1, is to promote, protect, and ensure the full and equal enjoyment of all human rights and fundamental freedoms by all persons with disabilities and to promote respect for their inherent dignity. UNCRPD has been a major quantitative and qualitative step forward in that it considers both the environmental supports and individual needs (Verdugo, Navas, Gómez, & Schalock, 2012).

As we have already pointed, studies on the QOL of people with intellectual disability follow the general incremental trend. In addition to this, QOL outcomes are becoming the target for intervention and a measure of success of different support programs (Memisevic, Hadzic, Zecic, & Mujkanovic, 2016). This is an indicator of the human rights approach that is currently dominating the field of ID.

Prior to embarking on the task of defining the QOL and describing individual QOL indicators, let us try to trace down the historical development of QOL concept. Then we will offer some contemporary QOL definitions and consider the breadth of the QOL concept.

History

The very phrase QOL was first used after the World War II, and since then it has been overused and infrequently defined (Meeberg, 1993). In the beginning, the term was equated with material goods: a house, a car, money to travel, and money to retire. Only later was the term broadened to capture the concepts such as education, health, and welfare (Farquhar, 1995). A “good life” is a life filled with happiness and meaning (Wolf, 1997).

The phrase “quality of life” was popularized in the 1970s as a global taxonomy used in the context of healthcare where it has been linked with sociology, medicine, psychology, economics, geography, social history, and nursing (Milton, 2013). However, we can trace back the origins of writings on QOL to the ancient Greek philosophers. Back in those days, QOL was referred to as a good life, happiness, well-being, and satisfaction. These were viewed as civic virtues that demanded cultivation (Schoch, 2006).

Ancient philosopher Aristotle (384–322BC) spoke about “good life” and the impact of public politics on its improvement. Philosophers following Aristotle were also interested in “the good life” and how to achieve it. Maybe most pertinent to this chapter is the view of Epicurean philosophy which states that happiness and a good life can be achieved through the possibility of making one’s own choices – self-determination (Cheung Chung, 1997). However, philosophers have never agreed upon the final and all-encompassing definition of QOL.

The history of research regarding QOL in people with ID is significantly shorter than that of the general population. This is a result of the priorities in the civil rights movements: certain preconditions had to be fulfilled before dealing with QOL.

One such issue is existential – right to life! People with intellectual disability have been oppressed throughout history. As a result the QOL for this category of people was not regarded as important.

This attitude originated from the widespread deprivation of human rights to people with ID in the past. This included rights such as forming intimate relationships, marriage, education, employment, self-determination, etc. Negative societal attitudes were best reflected in inhumane treatment practices for persons with ID such as institutionalization, asylums, involuntary sterilization, overmedication, and cruel punishments (Griffiths et al., 2006).

In the last four decades, there has been a significant improvement in societal attitudes toward persons with ID, and QOL becomes a major topic in the discourse in the field of ID. Many reasons contributed to this significant focus on QOL. Schalock, Verdugo, and Braddock (2002) identified four such reasons on the emphasis on QOL. The first reason deals with the change in attitudes on how QOL can be improved. Earlier views strongly associated the QOL concept with the advances in medical field. However, other aspects of a person's life are equally important, such as personal and environmental aspects. The second reason on the focus on QOL comes from the normalization principle. This principle postulates the need to provide same life conditions to all people regardless of the presence of disabilities. The third reason is the increased self-determination and empowerment of persons with ID. The fourth and last reason is the reconceptualization of QOL construct. This new view (concept) of QOL views both QOL components, subjective and objective, as equally important.

QOL Definitions and Conceptualizations

Defining the QOL and the answer as to what it constitutes is not as simple as it might seem. QOL can be conceptualized either as conditions of life or as experiences in life. It can also be considered for individuals or for a society as a whole (McCall, 1975, in Meeberg, 1993). In addition to having subjective and objective dimensions, the QOL construct can be viewed as either unidimensional or multidimensional. The fact that everybody uses the term QOL on an everyday basis, from advertising executives to politicians, thus contributes to multiple interpretations and poor definitions of the QOL concept (Haas, 1999).

In spite of all abovementioned difficulties in defining and conceptualizing QOL, there is consensus in the QOL literature about three things: First, QOL is subjective by nature; second, the various core dimensions are valued by persons differently; lastly, the value attached to each core dimension varies across one's life (Schalock, 2000).

Independently of the consensus in the literature, the fact is that QOL concept can be viewed from many perspectives. From the possible perspectives, we will focus briefly on the medical and social.

Medical Concept

The initial conceptualizations of QOL were related to the health status of the individuals. Most research on the medical perspective regarding the QOL has been done by the World Health Organization (WHO). Health, defined by WHO, is a state of complete physical, mental, and social well-being. Defined in such a way, health can easily be used as a synonym for QOL. The World Health Organization work group (WHO, 1995) defined QOL as the "individual's perception of their position in life in the context of the culture and value systems in which they live and in relation to their goals, expectations, standards and norms." It is obvious that this definition puts a strong emphasis on the subjective perception of QOL of the person.

According to the WHO, the QOL domains are physical health, psychological health, social relationships, and environment. Some equate it with well-being. In the context of health, most would agree that QOL decisions should be oriented toward improving and benefitting patients' lives (Keyserlingk, 1979; Meeberg, 1993).

However, the use of QOL term has diverse and inconsistent meanings in the healthcare literature (Milton, 2013). Very rarely do researchers define the concept at all, which leaves us unclear about what they are specifically referring to when speaking about the QOL (Meeberg, 1993). The WHO model of QOL has been employed widely in studies examining certain medical conditions but not so much with people with intellectual disability.

Social Concept

The field of ID has embraced the social view on QOL (Cummins, 2005). Within the social perspective, the concept of QOL reflects the following principles: (1) QOL consists of the same factors for all people, (2) QOL is achieved when a person's needs have been met, (3) QOL is a multidimensional construct that is affected by individual and environmental factors, and (4) QOL has both subjective and objective components (Verdugo et al., 2012). It is important to note that subjective and objective indicators do not carry the same weight in overall QOL. Authors mainly agree that the subjective perception is more important in assessing the overall QOL than objective (Day & Jankey, 1996). The objective side of QOL mainly deals with the socioeconomic status of the individual. As the very name indicates, the objective side could be assessed through someone's health status and functional ability.

One of the most widely used models of QOL in people with ID is the one formulated by Schalock and Verdugo (2012). This model includes eight core QOL domains: (1) emotional well-being, (2) interpersonal relations, (3) material well-being, (4) personal development, (5) physical well-being, (6) self-determination, (7) social inclusion, and (8) rights. Again, as is the case with WHO model, each of these domains has its own indicators and descriptors. However, the QOL domains most often associated with policy and practice guidelines for services for people with intellectual disability are inclusion, independence (personal development), and choice (Beadle-Brown, 2006).

General Indicators of QOL

QOL domains are universal for all people. In order to assess the QOL, each domain has its own indicators that need to be sensitive to personal and cultural specificities. In addition to this, indicators need to fulfill some basic criteria, such as importance for the individual, relevance, and to contribute to the overall QOL improvement (Schalock et al., 2005). There are numerous indicators of QOL. When asked what is a good QOL, most people will readily list elements such as health, happiness, family, friends, job satisfaction, marriage and intimate relations satisfaction, and so on. How people rank these QOL indicators in terms of importance is a different story. Most people rate health as the most important factor in QOL. However some people are healthy and lonely and do not feel they have a good QOL. The reason for this is that the QOL indicators differ in their universality in relation to geographic region and they also differ within micro, meso, and macro systems.

Lastly, indicators differ among individuals. Many authors point to the fact that there must be a difference made between subjective and objective QOL indicators (Headey & Wearing, 1992; Pacione, 1982; Schneider, 2005). These two groups of factors are not mutually exclusive however, and if we are to measure QOL validly and reliably, both subjective and objective indicators must be used in the QOL assessment (Das, 2008; Diener & Suh, 1997). In an additional effort to conceptualize QOL more clearly, Cummins (2005) suggests introducing a clear hierarchical organization of indicators from general to specific while simultaneously controlling for the so-called "causal" variables. These variables, together with the environmental characteristics, exert their complex influence on the indicators. According to Cummins (2005), general QOL indicators are related to specific QOL domains' indicators. Specific domains' indicators are influenced by objective (goods and services) and causal variables (perceived deficit in goods and services). Complexity of the relations that indicators have with different factors can be summarized in three-element system consisting of life conditions, subjective well-being, and personal values and aspiration (Felce & Perry, 1995).

Objective indicators can easily be observed by others and are thus called life conditions or life events. Life events include economic, political, cultural, familial, or natural events outside of the direct control of the individual (Goode, 1990). It also encompasses some characteristics of the individual such as health and socioeconomic status. Objective indicators are much easier to define and measure than subjective indicators. Subjective indicators represent personal perceptions and individual assessment of objective indicators and thus are harder to measure. Although there is no universally accepted list of QOL indicators, there are several available examples.

Many of the examples of QOL were specified in the European Conference in Rio de Janeiro (Quarrie, 1992). The conference committee came to the conclusion that QOL indicators can be divided into four basic groups of indicators – social, economic, environmental, and institutional-political. Each of these groups was further divided into smaller subgroups, resulting in a total of 76 indicators. In the European Commission final report, nine key indicators are mentioned: material living conditions, productive or main activity, health, education, leisure and social interactions, economic and physical safety, governance and basic rights, natural and living environment, and overall experience of life (Eurostat, 2017).

Similarly extensive, the division of indicators provided by Henderson and Lickerman (2000) consists of 12 groups of indicators: education, employment, energy, environment, health, human rights, income, infrastructure, nationals, publics, recreation, and shelter indicator.

QOL Indicators for Persons with ID

In her inaugural talk as the president of the American Association on Intellectual and Developmental Disabilities (AAIDD), Susan M. Havercamp (2017) emphasized the notion that health and well-being are critical to QOL. Knowing what constitutes a good QOL for an individual might help us in designing programs for its improvement. A great role for improving QOL in persons with ID has been placed on support service providers. One way of improving the QOL is to employ person-centered planning and systems of support for individuals with ID (Verdugo et al., 2012). QOL can be seen as a relevant outcome measure for education, health, and social service programs (Schalock, 2004a, 2004b). In addition to this, knowing the challenges to improve the QOL resulted in many productive and innovative changes in service delivery principles and practices, evaluation strategies, and identification of personal outcomes reflecting the QOL (Schalock, 2004a, 2004b).

Although, as we have seen, there are many domains and indicators pertaining to the QOL, we will not systematically embrace any of these concepts but instead will write about what we believe are the most important aspects/dimensions/domains of QOL in persons with ID. As QOL, as a social construct, is measured through its indicators, we will focus our attention now on its most important constituents. As a result of numerous studies aimed at QOL assessment in persons with ID, 3 key factors, 8 domains, and 23 indicators stood out (Schalock & Verdugo, 2012; Verdugo, Arias, Gómez, & Schalock, 2010). This is shown in Fig. 6.2.

Besides these established QOL domains and indicators, we cannot speak about QOL in people with ID without mentioning some constructs that are closely related to their QOL. In the following text, we will describe some of these main constructs including empowerment, choice/self-determination, self-advocacy, inclusion, independent living, and person-centered planning and their relationship with QOL in persons with ID.

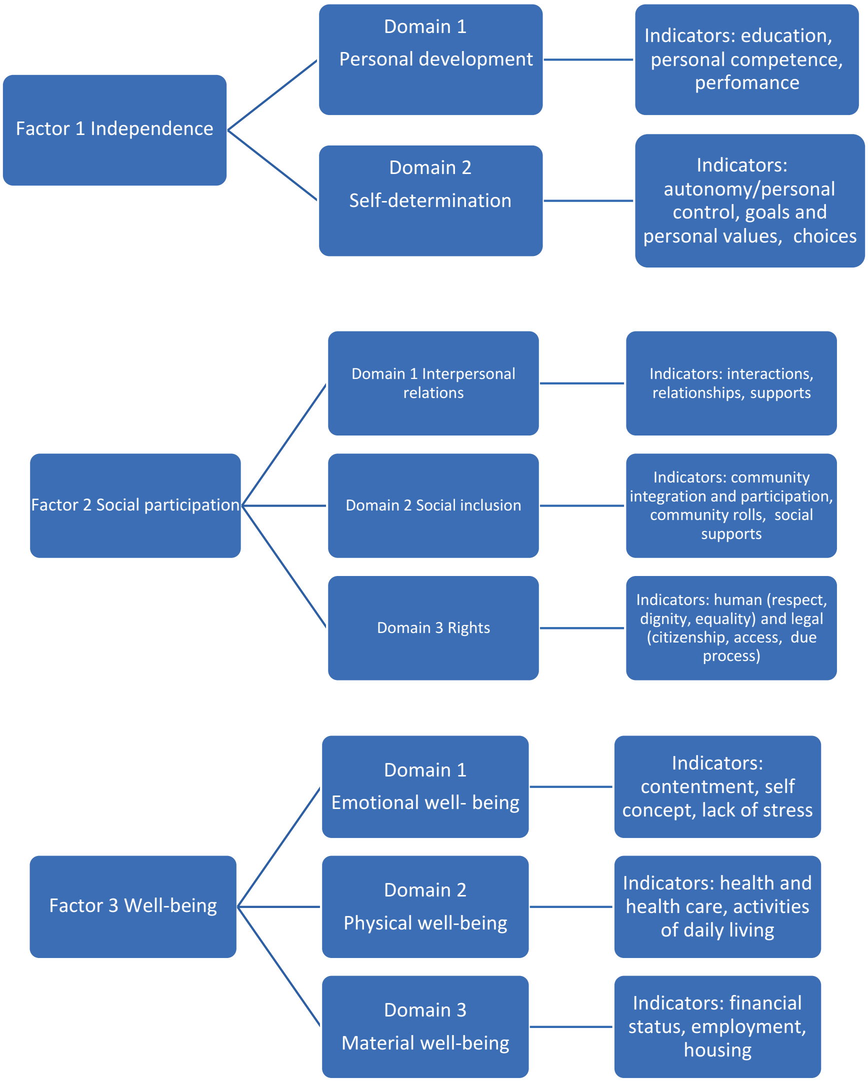


Fig. 6.2 Key factors, domains, and indicators of QOL for people with ID

Empowerment

Empowerment, as a term, has been used since the 1970s of the last century. It was first used by social service providers and researchers in relation to marginalized groups (Calvès, 2009). One of the first empowerment theories is the one developed by Freire (1974, in Calvès, 2009), which postulates that the duty and obligation of the educators are to help individuals develop a "critical consciousness," awareness of themselves and their situation, and the ability to make independent choices. In the 1990s, empowerment became a basic premise in rehabilitation of persons with disability. Special importance is given to independent decision-making, risk-taking of people with disabilities, as well as taking responsibility for the choices they made, and taking control of one's life (Emener, 1991, in Niesz, Koch, & Rumrill, 2008).

This empowerment concept allows people with disabilities to reject the role of the patient – a passive role imposed on them by the medical model of disability – and to accomplish the sense of personal power (Hayes & Hannold, 2007). The very concept of autonomy can be perceived in two ways – autonomy of action and autonomy as a capacity of persons (Miller, 1985, in Clark, 1988). Autonomy of action means that the person voluntarily and willingly implements actions he/she considers authentic, in line with her life values and goals. Autonomy as a capacity of persons means personal power of an individual to make decisions themselves, independently from other's opinions judgments and decisions.

Empowerment, according to Spreitzer (1996), is composed of four basic components: meaning, competence, self-determination, and personal control. Meaning is related to the coherence of the roles given to the person and the person's opinions, convictions, value system, and behavior. Competence, as the second component of empowerment, can be defined as an individual's conviction about their skills and abilities. Self-determination means the ability of the person to make their own choices without the influence from others. The last component of empowerment is personal control, which is defined as a level of the person's influence on the outcomes. The purpose of empowerment is to put a person with ID into the focus and prescribes that an individual has a very important role in organizing their life and everyday activities.

QOL is closely related to the concept of empowerment and personal autonomy. Improvement in empowerment contributes to (a) improved levels of information (people with ID need to have more information about the services and options which are at their disposal) and (b) values clarification (in order to make informed decisions, people need to have more information about the criteria and principles which need to be respected in decision-making) (Clark, 1988).

Different life periods carry various roles and options and put the person with ID in situations to make decisions, compare information, assess the outcomes, and then make informed decisions about their life. The transition period from adolescence into adulthood, which is usually between the ages 18 and 25, is especially sensitive as it is the period of many major choices. People with ID need to be especially empowered in this period to decide how they want their lives to look (Curryer, Stancliffe, & Dew, 2015).

An international study (Canada, United States, Belgium, and France) examining the link between empowerment and QOL has shown that autonomous functioning, self-regulation, psychological empowerment, and self-realization are statistically correlated with QOL; the stronger the empowerment, the better the QOL for the individual with ID (Lachapelle et al., 2005). Psychological empowerment predicts QOL outcomes and lower level of financial support for people with ID, and higher levels of autonomy predict independent living of persons with ID (Shogren & Shaw, 2016).

Choice/Self-Determination

One of the fundamental human rights is the ability to make choices regarding one's life, such as place of residence, marriage partner, friends, etc. Ability to choose is considered to be an essential component of QOL. Choice involves the concept of self-determination (Smith, Morgan, & Davidson, 2005). Thus, promoting choice options and self-determination for people with intellectual disability has become a best practice in the field of ID (Wehmeyer & Abery, 2013). According to them, self-determined actions have four essential characteristics: (1) the person acts autonomously, (2) behavior of the person is self-regulated, (3) the person initiates and responds to events in a psychologically empowered manner, and (4) the person acts in a self-realizing manner.

Self-determination influences QOL in many ways, such as through social and vocational integration, rehabilitation, and habilitation goals (Nota, Ferrari, Soresi, & Wehmeyer, 2007). However, as QOL is a complex social construct consisting of many components, self-determination is not equally strongly related to all its constituents. For example, research has shown that self-determination is a good predictor of personal development and personal fulfillment (McDougall, Evans, & Baldwin, 2010). Also of note is the potential susceptibility of self-determination to training. Components of self-determination such as choice-making skills, decision-making skills, goal-setting, and self-awareness skills can all be promoted and improved in individuals with ID (Wehmeyer & Abery, 2013). However, these attempts to improve self-determination skills in persons with ID must be followed by education and training of the support service staff. In addition to this, actions aimed at raising the awareness of wider society should also be undertaken. As an example of this, let us shortly explore the issue of sexuality in persons with ID, as this is viewed as a complex issue. Even parents of persons with intellectual disability express concerns regarding the sexuality of their children and believe that expert support is needed to deal with this issue (Isler, Beytut, Tas, & Conk, 2009). Research has shown that there is a need for the education of caregivers and a need to discuss sexuality issues at societal and policy levels (Mcguire & Bayley, 2011). This in turn will help persons with ID to better deal with their sexuality and make decisions regarding their sexuality.

Self-Advocacy

Self-advocacy is a civil rights movement of and by people with ID which serves as a method of empowering people with ID (Friedman, 2017). This movement originated in Sweden and then gradually expanded to other parts of the world. Self-advocating means the ability and possibility to speak on behalf of yourself. It means that the persons with ID may express themselves freely, make their own decisions, have the right to dignity, and take over control of their own lives and is a means to fight against discrimination.

It is particularly important in transition years, when people make important choices in their lives. This is especially evident when students with ID are finishing their formal schooling. After exiting school, many former special education students cannot plan their future, remain unemployed or underemployed, and experience a quality of life remarkably different from their nondisabled peers (Martin, Huber Marshall, & Maxson, 1993). Fortunately, as with self-determination, self-advocacy skills can be learned. During the school years, students should master self-management skills as they move through the grades; students learn self-advocacy by participating in their IEP meetings, learning to develop the IEP, and then managing it (Martin et al., 1993). Students with ID require intentional training to further develop their self-advocacy skills. Training in self-advocacy for people with ID should encompass strengthening mutual trust among persons with ID, focusing on joint problems and interests, strengthening self-respect, and introducing people with ID to their rights and responsibilities (Bilić, Bratković, & Nikolić, 2004).

Inclusion

There is no universally accepted definition of the term inclusion. The definition changed over time. Some authors perceive inclusion as a movement (Karagiannis, Stainback, & Stainback, 2000), while others see it as a philosophy or belief system (Florian, 2000; Ryndak, Jackson, & Billingsley, 2000). Inclusion as a civil movement has its roots in Western Europe, where it first developed with the aim to improve human rights of people regardless of their nation, religion, sexual orientation, disability, etc.

Four steps (phases) can be identified in the process from exclusion to inclusion: (1) exclusion of people with disability from society; (2) technical inclusion, which means satisfying basic needs of people with ID and support in their least restrictive environment; (3) legal inclusion, the laws and legislation protecting the rights of people with ID and emphasis on their autonomy, independence, and dignity; and (4) ethical inclusion, which stresses fulfilling interpersonal relations and acceptance of differences (Clapton, 2009).

Inclusion of persons with ID is usually viewed through the prism of educational inclusion. We will, however, say something about social inclusion, which is a broader concept and encompassed education inclusion as well. Social inclusion of persons with ID includes total access to activities, social roles, and interpersonal relations (Partington, 2005). Simplican, Leader, Kosciulek, and Leahy (2015) provide an ecological model of defining social inclusion, consisting of subcomponents: individual (characteristics of the person), interpersonal (attitudes, relations with the environment), organizational (socioeconomic characteristics of the family, access to support services), social (availability of services, type of accommodation, self-advocacy groups), and social-political level (legislature). Social inclusion depends on several factors: self-determination, autonomy, participation in policy-making, availability of social supports, and availability of transport (Verdonschot, De Witte, Reichrath, Buntinx, & Curfs, 2009).

Social inclusion is in many ways related to the QOL in persons with ID. Higher levels of social inclusion are related to enhanced personal satisfaction and higher level of QOL (Eliason, 1998). Persons with ID with more social interactions and interpersonal relations have better QOL (Lee McIntyre, Kraemer, Blacher, & Simmerman, 2004; O'Brien, Thesing, Tuck, & Capie, 2001). Piro (2017) pointed to the link between QOL and social inclusion, explaining that society set standards for all persons, and if an individual cannot fulfill these standards and assume social roles, then that individual will have lower QOL.

Independent Living and QOL

Independent living is strongly related to self-determination and the QOL. Not long ago, when the medical concept of disability still dominated the field, the consensus was that the majority of people with ID were not competent to live in the local community. As a result, they were placed in state-run institutions that "cared" for them for the rest of their lives. This process is widely known as institutionalization. Unfortunately, institutionalization of persons with ID is still a widely used model of social care in many parts of the world.

However, in developed countries, the reversed process, called deinstitutionalization, was sparked in the late 1960s and early 1970s by disability movements seeking better QOL for people with disabilities. The policy of deinstitutionalization was based on the notion that the QOL of people with ID will improve when they move from institutions to local communities (Chowdhury & Benson, 2011). Deinstitutionalization is now gaining its momentum throughout the world thanks to the human rights movement and international conventions, in particular the UNCRPD which stressed the right of people with ID to live in their local communities and exercise their rights as full members of the society. It is evident that the process of deinstitutionalization is strongly related to independent living of people with ID. One of the early conceptualizations of independent living was given by Gerben DeJong (1979), who views independent living as a social movement and an "analytic paradigm" that is redirecting the course of disability policy, practice, and research.

There are many benefits of living in the local communities rather than living in institutions, and these benefits are all related to the QOL outcomes. In a study by Emerson and Hatton (1996), the authors state some of these benefits such as increased use of community facilities, increases in adaptive behavior, increased opportunities for choice, increased contact with family and friends, better material standard of living, and increased acceptance by the community. However it is important to note that QOL outcomes depend mainly on the quality of the service offered by community-based residential services. Again, the role of service providers in the community was viewed as crucial component in the participants' QOL. In a review of 15 studies examining QOL of people with ID, authors found that relocation had a positive impact on QOL measures in participants (Chowdhury & Benson, 2011).

There are several types of community-based living options for people with ID. They are group-based homes, semi-independent, and independent living. It seems that a higher level of independence is related to better QOL outcomes. People living semi-independently experience less social dissatisfaction, use community services more frequently, have more participation in domestic tasks, and feel greater empowerment (Stancliffe & Keane, 2000).

It is important to note that the social care system poses many barriers to independent living for people with ID. Social service professionals still focus on the “risk” of living in the community and question the “capacity” of people with ID to live independently (Morris, 2004). It is of paramount importance that the legislature changes to include entitlement to independent living.

Person-Centered Planning

From the service-level perspective, one of the most important segments is person-centered planning. In order to understand the relationship between the disability, community, and organized services, the term person-centered planning (PCP) has been developed. PCP has its origin in Wolfensberger’s principles of normalization theory (Wolfensberger, 1972). Person-centered planning (PCP) can be defined in several ways. It is an individualized approach to organizing assistance and support to people with ID (Mansell & Beadle-Brown, 2004). PCP is an approach in which a person with disabilities designs and manages the support services and is strongly based in the person’s strengths and visions (Flannery et al., 2000). It is especially important due to its use as a vehicle to achieve the following goals for people with ID: rights, independence, choice and inclusion.

PCP is more than a theoretical concept; it is a specific method for bringing about positive changes in the lives of people with ID (Holburn, 2002). It is based on three important principles. First, it emphasizes aspirations and capacities of people with ID (instead of their weaknesses and shortcomings). Secondly, it includes the person’s family and mobilizes the wider social network in order to support the person with ID. Finally, it emphasizes providing the support required to achieve the goal set by the person with ID.

It is evident that PCP is strongly related to support services. What are the ways to make support services more person-centered? The answer to this question is borrowed from Cambridge and Carnaby’s work from Cambridge & Carnaby, 2005. Firstly, PCP should be legally regulated, that is, it should be given legal weight. As often as possible, persons with ID should have direct payments and control of their budgets. The focus should be shifted from *person-centered planning* to person-centered action.

The goal of PCP is to allow a person with ID to lead a meaningful life in a way that he/she wants, with the support of their circle of friends and their community. Principles of PCP are greatly affecting the support services provided to people with ID and thus improving them constantly. NGOs that provide such services need to be aware of PCP; fortunately, this approach is expanding widely throughout the world. Now the support services need to ensure that certain standards are in place if PCP is used in the field. For example, indicators of PCP can be assessed through the following questions (Parley, 2001):

- Does the person make everyday choices?
- Are they treated with respect?
- Do they take part in everyday activities?
- Do they have friendships and relationships?
- Are they part of their local community?
- Do they get a chance to work?
- Do they take part in important decisions about their life?
- Are their families’ views listened to?
- Are they safe from harassment and abuse?
- Do they get help to stay healthy?

Measuring QOL

> “If something exists, then it can be measured.” (Thorndike, 1904)

We are not sure how the concept of measuring and assessing the QOL can be applied meaningfully given that each individual has their own perspective on what it means to have a good QOL. Nevertheless, the instruments, questionnaire, and scales purporting to “measure” the QOL are on the constant rise. They are in line with the task to better understand the social construct of QOL in relation to many factors such as health status, disability, severity, aging, etc. Schalock et al. (2002) stated that the task of measuring QOL should take into account five basic principles, and within each principle, there are certain directions that need to be followed:

1. Measurement should encompass the level of achievement of individually valued life experiences. Within this principle it is necessary to take into account the theoretical basis of measurement, cultural and time differences, precise terminology, and categories stemming from the measurement itself.
2. Measurement should have a positive impact on the individual in the sense that he/she directs their will toward activities that are meaningful and pleasant for them. Measurement is focused on key life aspects that can be improved and is focused to identify non-adequately satisfied needs and creation of an action plan to satisfy these needs, and the results of QOL measurement are interpreted in relation to contextual factors, such as age, gender, etc.
3. Measurement's purpose is to determine the extent to which different QOL domains are related to QOL in general. It should be ensured that domains relevant to all people are encompassed, while the critical indicators can be different in relation to the age, culture, disability, etc.
4. Measurement should be performed in the context that is important for the individual – where he lives and spends time. Within this principle, it may be necessary to engage a proxy who would assess the QOL from the perspective of the person who cannot speak for themselves and choose the experience that is most appropriate for that person.
5. Measurement is based on both universal life experiences and unique personal experiences. It is necessary to measure both objective and subjective QOL aspects, encompassing both cognitive and affective components and by using quantitative and qualitative techniques.

Many individuals with ID are willing and able to discuss their QOL. Consequently, researchers should use all available and efficacious methods to enable a person with ID to report about their life experiences. This may include simplifying questions for individuals with ID (by using simple and concrete examples) as well as simplifying response methods (by using assistive technologies, e.g., pictures). Researchers should be aware of bias, given the fact that persons with ID are susceptible to giving socially desired responses. In order to ensure this, prior to any measurement of QOL, individuals with ID should be asked how well they understand items (perhaps through the use of pretest questions or formal tasks – as in the case of ComQol-ID, Cummins, 1993) (White-Koning et al., 2005).

In extreme cases, when the person with ID is not able to report about their life experiences, even with accommodations (as in the case of people with severe or profound ID), the most common way to assess their life situation is through the use of proxies (parents, caretakers, legal guardians) (Verdugo, Schalock, Keith, & Stancliffe, 2005). But, these data obtained from proxies should not replace self-reporting of the persons with ID themselves.

Respecting all these principles and direction in QOL measurement, Claes, Van Hove, van Loon, Vandevelde, and Schalock (2010) stated that the measurement results of QOL in persons with ID should reflect their personal, organizational, and macro-level situation. On personal plan, QOL results should provide information on each of the eight QOL domains. Results of QOL measurement on organizational level can be used to determine the predictors of QOL and their effect in predicting the outcomes. QOL measurement has also great impact on macro level, in shaping, creating, and planning the policies and in financing the services for persons with ID.

In order to find a perfect instrument for QOL assessment in persons with ID, one that has excellent psychometric properties, which is culturally specific and economic, Li et al. conducted a theoretical review of 309 studies (Li, Tsoi, Zhang, Chen, & Wang, 2013). Their research determined nine instruments that satisfied all criteria and were used to assess QOL in people with different levels of severity of ID. The number of items varied from 7 to 58, and the time needed to fill out the questionnaires was between 20 and 45 min. Only three instruments were used to assess both subjective and objective aspects of QOL (ComQol-ID, Cummins, 1993; QOLQ, Schalock & Keith, 1993; POS, Van Loon, Van Hove, Schalock, & Claes, 2009), while the remaining six assessed subjective components only (CQ, Stancliffe & Parmenter, 1999; HSIS, Ruddick & Oliver, 2005; LSS, Heal & Chadsey-Rusch, 1985; MLSS, Harner & Heal, 1993; PWI-ID, Cummins & Lau, 2005; MAMP, Bonham et al., 2004). Similar theoretical research was conducted by Townsend-White, Pham, and Vassos (Townsend-White, Pham, & Vassos, 2012), with the aim to find the most appropriate instrument for QOL assessment in persons with ID and comorbid behavioral issues. The review encompassed studies published between 1980 and 2008 and resulted in six instruments (MLSS, Harner & Heal, 1993; ComQol-ID, Cummins, 1993; QOLQ, Schalock & Keith, 1993; PWI-ID, Cummins & Lau, 2005; QUOLIS, Ouellette-Kuntz, 1990; EQLI, Nota, Soresi, & Perry, 2006). Analyzing these instruments, authors conclude that there is no specialized instrument that would measure QOL in persons with ID and comorbid behavioral difficulties. In order to increase variety and the number of instruments aimed at measuring QOL in persons with ID, authors emphasize the need to develop instruments based on the theoretical concepts of QOL.

Short Overview of the QOL Research in People with ID

Many studies point to the fact that people with ID have lower QOL than people without ID. In a study conducted in Taiwan, authors found that 42.9% of people with ID do not use local community services and do not have much interaction with education, employment, and social inclusion. In the study, adaptive behavior skills and educational level were identified as predictors of higher QOL. The place of residence was the strongest predictor for utilization of community services (Chou et al., 2007).

Results of the research comparing the QOL in adults with ID in Italy and Australia found no differences between them, but both groups achieved lower scores in the domains of material well-being, health, productivity, and community. The users in both countries were most dissatisfied with the “community living” domain (Verri et al., 1999). Other authors report similar results. For example, Bramston et al. reported that people with ID are best satisfied with the health domain and less satisfied with the domains of privacy and social inclusion (Bramston, Chipuer, & Pretty, 2005).

Investigators have also examined some demographic variables related to the QOL and found that male gender, lower severity of ID, and family environment contribute to better QOL (Simões & Santos, 2017). The type of residence frequently plays a role in QOL. Tamaš (2015) reported that the best QOL was found in adults with ID who are living independently with support, while the lower QOL was found for persons with ID living in families and institutions. A plethora of research supports the notion that living in local community improves the QOL in persons who were previously institutionalized (Chowdhury & Benson, 2011; Felce & Emerson, 2001; Kozma, Mansell, & Beadle-Brown, 2009; O’Brien et al., 2001; Walsh et al., 2010).

Age and gender effects were also examined in relation to the QOL. Older women reported significantly lower subjective well-being (SBW) and less positive self-concept than men on all measures, except subjective age, although gender accounted for less than 1% of the variance in well-being and self-concept. Smaller gender differences in SWB were found in younger than in older groups. Statistically controlling for gender differences, marriage status, health, and socioeconomic status decreased gender differences in SWB. Cohort differences in SWB are reported as well (Pinquart & Sörensen, 2001). Friedman and Rizzolo (2018) emphasize the importance of interactions with friends in adults with ID as a significant variable in predicting the QOL. They reported that satisfaction with friends increases the odds of being included in community and fulfillment of various social roles by four times.

Duvdevany and Arar (2004) reported similar results, pointing that the quality of friendships is closely related to the subjective well-being in persons with ID.

Mothers of adult individuals with severe ID reported that for the QOL of their children the most crucial factors are participation in recreation activities and hobbies. After these factors, in importance, is the belonging to local community and satisfaction of their needs (Lee McIntyre et al., 2004). A survey of experts and parents of adults with severe ID living in the community indicated that various social roles, participation in hobbies, and vocational activities might have a positive impact on the QOL of people with severe ID (Hartnett et al., 2008).

Concluding Remarks

Quality of life has become a topic of major importance in the field of intellectual disability. And rightly so. In this chapter we described different models of QOL as well as its different components. We can confidently say that QOL of people with ID is steadily improving all over the world. Many factors contribute to this widespread improvement in QOL for people with ID. These factors are related to improved legislature supporting human rights of people with ID in many countries. In addition to this, numerous technical and medical advances have also contributed to the better QOL. In order to further advance QOL in people with ID, it is necessary to empower them to fully participate in their local communities. The role of support service providers is also very important in this regard. These providers need to incorporate the concepts of empowerment, self-determination, inclusion, and independent living, in their policies and practices in order to enhance person-referenced QOL outcomes. In conclusion, it is obvious that the concept of QOL is complex and many questions regarding it remain unanswered. However, we should be optimistic. The emphasis in QOL research has shifted from human rights issues to identifying factors that will reduce the gap in QOL regardless of the presence of intellectual disability. “

References

Beadle-Brown, J. (2006). Person-centred approaches and quality of life. *Tizard Learning Disability Review, 11*(3), 4–12.

Bilić, M., Bratković, D., & Nikolić, B. (2004). Evaluacija programa osposobljavanja osoba s mentalnom retardacijom za samozastupanje u području samopoštovanja [*Evaluation of training program for persons with intellectual disability for self-advocacy and self-respect*]. *Hrvatska revija za rehabilitacijska istraživanja, 40*(1), 1–12.

Bonham, G. S., Basehart, S., Schalock, R. L., Marchand, C. B., Kirchner, N., & Rumenap, J. M. (2004). Consumer-based quality of life assessment: The Maryland Ask Me! Project. *Mental Retardation, 42*, 338–355.

Bramston, P., Chipuer, H., & Pretty, G. (2005). Conceptual principles of quality of life: An empirical exploration. *Journal of Intellectual Disability Research, 49*(10), 728–733.

Calvès, A. E. (2009). Empowerment: The history of a key concept in contemporary development discourse. *Revue Tiers Monde*, (4), 735–749.

Cambridge, P., & Carnaby, S. (2005). *Person centred planning and care management with people with learning disabilities*. London: Jessica Kingsley Publishers.

Cheung Chung, M. (1997). A critique of the concept of quality of life. *International Journal of Health Care Quality Assurance, 10*(2), 80–84.

Chou, Y. C., Schalock, R. L., Tzou, P. Y., Lin, L. C., Chang, A. L., Lee, W. P., & Chang, S. C. (2007). Quality of life of adults with intellectual disabilities who live with families in Taiwan. *Journal of Intellectual Disability Research, 51*(11), 875–883.

Chowdhury, M., & Benson, B. A. (2011). Deinstitutionalization and quality of life of individuals with intellectual disability: A review of the international literature. *Journal of Policy and Practice in Intellectual Disabilities, 8*(4), 256–265.

Claes, C., Van Hove, G., van Loon, J., Vandevelde, S., & Schalock, R. L. (2010). Quality of life measurement in the field of intellectual disabilities: Eight principles for assessing quality of life-related personal outcomes. *Social Indicators Research, 98*(1), 61–72.

Clapton, J. (2009). *A transformatory ethic on inclusion: Rupturing concepts of disability and inclusion*. Boston, MA: Sense Publishers.

Clark, P. G. (1988). Autonomy, personal empowerment, and quality of life in long-term care. *Journal of Applied Gerontology, 7*(3), 279–297.

Cummins, R. A. (1993). *Comprehensive quality of life scale* (4th ed.). Melbourne, VIC: Deakin University.

Cummins, R. A. (2005). Moving from the quality of life concept to a theory. *Journal of Intellectual Disability Research, 49*(10), 699–706.

Cummins, R. A., & Lau, A. D. L. (2005). *Personal wellbeing index-intellectual disability* (3rd ed.). Melbourne, VIC: Deakin University.

Curryer, B., Stancliffe, R. J., & Dew, A. (2015). Self-determination: Adults with intellectual disability and their family. *Journal of Intellectual and Developmental Disability, 40*(4), 394–399.

Das, D. (2008). Urban quality of life: A case study of Guwahati. *Social Indicators Research, 88*(2), 297–310.

Day, H., & Jankey, S. G. (1996). Lessons from the literature: Toward a holistic model of quality of life. In R. Renwick, I. Brown, & M. Nagler (Eds.), *Quality of life in health promotion and rehabilitation* (pp. 39–62). Thousand Oaks, CA: Sage.

Diener, E., & Suh, E. (1997). Measuring quality of life: Economic, social, and subjective indicators. *Social Indicators Research, 40*(1-2), 189–216.

Duvdevany, I., & Arar, E. (2004). Leisure activities, friendships, and quality of life of persons with intellectual disability: Foster homes vs community residential settings. *International Journal of Rehabilitation Research, 27*(4), 289–296.

Eliason, S. L. (1998). Social integration and satisfaction among individuals with developmental disabilities: A sociological perspective. *Education and Training in Mental Retardation and Developmental Disabilities, 33*(2), 162–167.

Emerson, E., & Hatton, C. (1996). Deinstitutionalization in the UK and Ireland: Outcomes for service users. *Journal of Intellectual and Developmental Disability, 21*(1), 17–37.

Eurostat. (2017). *Final report of the expert group on quality of life indicators*. Luxembourg: Publications Office of the European Union.

Farquhar, M. (1995). Elderly people's definitions of quality of life. *Social Science & Medicine, 41*(10), 1439–1446.

Felce, D., & Emerson, E. (2001). Living with support in a home in the community: Predictors of behavioral development and household and community activity. *Developmental Disabilities Research Reviews, 7*(2), 75–83.

Felce, D., & Perry, J. (1995). Quality of life: Its definition and measurement. *Research in Developmental Disabilities, 16*(1), 51–74.

Flannery, K. B., Newton, S., Horner, R. H., Slovic, R., Blumberg, R., & Ard, W. R. (2000). The impact of person centered planning on the content and organization of individual supports. *Career Development for Exceptional Individuals, 23*(2), 123–137.

Florian, L. (2000). Inclusive practice: What, why and how? In C. Tilstone, L. Florian, & R. Rose (Eds.), *Promoting inclusive practice*. London/New York: Routledge Falmer.

Friedman, C. (2017). Self-advocacy services for People with intellectual and developmental disabilities: A national analysis. *Intellectual and Developmental Disabilities, 55*(6), 370–376.

Friedman, C., & Rizzolo, M. C. (2018). Friendship, quality of life, and people with intellectual and developmental disabilities. *Journal of Developmental and Physical Disabilities, 30*(1), 39–54.

Gerben Dejong, M. P. A. (1979). Independent living: From social movement to analytic paradigm. *Archives of Physical Medicine and Rehabilitation, 60*(10), 435–436.

Goode, D. A. (1990). Thinking about and discussing quality of life. *Quality of life: Perspectives and issues*, 41–57.

Griffiths, D. M., Owen, F., Gosse, L., Stoner, K., Tardif, C. Y., Watson, S., … Vyrostko, B. (2006). Human rights and persons with intellectual disabilities: An action research approach for community based organizational self-evaluation. *Journal of Developmental Disabilities, 12*, 165–181.

Haas, B. K. (1999). A multidisciplinary concept analysis of quality of life. *Western Journal of Nursing Research, 21*(6), 728–742.

Harner, C. J., & Heal, L. W. (1993). The multifaceted lifestyle satisfaction scale (MLSS): Psychometric properties of an interview schedule for assessing personal satisfaction of adults with limited intelligence. *Research in Developmental Disabilities, 14*, 221–236.

Hartnett, E., Gallagher, P., Kiernan, G., Poulsen, C., Gilligan, E., & Reynolds, M. (2008). Day service programmes for people with a severe intellectual disability and quality of life: Parent and staff perspectives. *Journal of Intellectual Disabilities, 12*(2), 153–172.

Havercamp, S. M. (2017). Presidential address: Promoting health and wellness. *Intellectual and Developmental Disabilities, 55*(6), 365–369.

Hayes, J., & Hannold, E. L. M. (2007). The road to empowerment: A historical perspective on the medicalization of disability. *Journal of Health and Human Services Administration, 30*(3), 352–377.

Headey, B., & Wearing, A. J. (1992). *Understanding happiness: A theory of subjective well-being*. Melbourne, VIC: Longman Cheshire.

Heal, L. W., & Chadsey-Rusch, J. (1985). The lifestyle satisfaction scale (LSS): Assessing individuals' satisfaction with residence, community settings an associated services. *Applied Research in Mental Retardation, 6*, 470–495.

Henderson, H., & Lickerman, J. (2000). In P. Flynn (Ed.), *Calvert-Henderson quality of life indicators*. Bethesda, MD: Calvert Group.

Holburn, S. (2002). How science can evaluate and enhance person-centered planning. *Research and Practice for Persons with Severe Disabilities, 27*(4), 250–260.

Isler, A., Beytut, D., Tas, F., & Conk, Z. (2009). A study on sexuality with the parents of adolescents with intellectual disability. *Sexuality and Disability, 27*, 229–237.

Karagiannis, A., Stainback, W., & Stainback, S. (2000). Historical overview of inclusion. In W. Stainback & S. Stainback (Eds.), *Inclusion: A guide for educators*. Baltimore, MD: Brookes Publishing Co.

Keyserlingk, E. W. (1979). *Sanctity of life or quality of life in the context of ethics, medicine and law*. Ottow: Law Reform Commission of Canada.

Kozma, A., Mansell, J., & Beadle-Brown, J. (2009). Outcomes in different residential settings for people with intellectual disability: A systematic review. *American Journal on Intellectual and Developmental Disabilities, 114*(3), 193–222.

Lachapelle, Y., Wehmeyer, M. L., Haelewyck, M. C., Courbois, Y., Keith, K. D., Schalock, R., … Walsh, P. N. (2005). The relationship between quality of life and self-determination: An international study. *Journal of Intellectual Disability Research, 49*(10), 740–744.

Lee McIntyre, L., Kraemer, B. R., Blacher, J., & Simmerman, S. (2004). Quality of life for young adults with severe intellectual disability: Mothers' thoughts and reflections. *Journal of Intellectual and Developmental Disability, 29*(2), 131–146.

Li, C., Tsoi, E. W., Zhang, A. L., Chen, S., & Wang, C. J. (2013). Psychometric properties of self-reported quality of life measures for people with intellectual disabilities: A systematic review. *Journal of Developmental and Physical Disabilities, 25*(2), 253–270.

Mansell, J., & Beadle-Brown, J. (2004). Person-centred planning or person-centred action? Policy and practice in intellectual disability services. *Journal of Applied Research in Intellectual Disabilities, 17*(1), 1–9.

Martin, J. E., Huber Marshall, L., & Maxson, L. L. (1993). Transition policy: Infusing self-determination and self-advocacy into transition programs. *Career Development for Exceptional Individuals, 16*(1), 53–61.

McDougall, J., Evans, J., & Baldwin, P. (2010). The importance of self-determination to perceived quality of life for youth and young adults with chronic conditions and disabilities. *Remedial and Special Education, 31*(4), 252–260.

McGuire, B. E., & Bayley, A. A. (2011). Relationships, sexuality and decision-making capacity in people with an intellectual disability. *Current Opinion in Psychiatry, 24*(5), 398–402.

Meeberg, G. A. (1993). Quality of life: A concept analysis. *Journal of Advanced Nursing, 18*(1), 32–38.

Memisevic, H., Hadzic, S., Ibralic Biscevic, I., & Mujkanovic, E. (2017). Quality of life of people with disabilities in Bosnia and Herzegovina–is there a moderating effect of the health status? *Scandinavian Journal of Disability Research*, 1–8.

Memisevic, H., Hadzic, S., Zecic, S., & Mujkanovic, E. (2016). Predictors of quality of life in people with intellectual disability in Bosnia and Herzegovina. *International Journal on Disability and Human Development, 15*(3), 299–304.

Milton, C. L. (2013). The ethics of defining quality of life. *Nursing Science Quarterly, 26*(2), 121–123.

Morris, J. (2004). Independent living and community care: A disempowering framework. *Disability & Society, 19*(5), 427–442.

Niesz, T., Koch, L., & Rumrill, P. D. (2008). The empowerment of people with disabilities through qualitative research. *Work, 31*(1), 113–125.

Nota, L., Ferrari, L., Soresi, S., & Wehmeyer, M. (2007). Self-determination, social abilities and the quality of life of people with intellectual disability. *Journal of Intellectual Disability Research, 51*(11), 850–865.

Nota, L., Soresi, S., & Perry, J. (2006). Quality of life in adults with an intellectual disability: The evaluation of quality of life instrument. *Journal of Intellectual Disability Research, 50*, 371–385.

O'Brien, P., Thesing, A., Tuck, B., & Capie, A. (2001). Perceptions of change, advantage and quality of life for people with intellectual disability who left a long stay institution to live in the community. *Journal of Intellectual and Developmental Disability, 26*(1), 67–82.

Ouellette-Kuntz, H. (1990). A pilot study in the use of the quality of life interview schedule. *Social Indicators Research, 23*, 283–298.

Pacione, M. (1982). The use of objective and subjective measures of life quality in human geography. *Progress in Geography, 6*(4), 495–514.

Parley, F. F. (2001). Person-centred outcomes: Are outcomes improved where a person-centred care model is used? *Journal of Learning Disabilities, 5*(4), 299–308.

Partington, K. (2005). What do we mean by our community? *Journal of Intellectual Disabilities, 9*, 241–251.

Pinquart, M., & Sörensen, S. (2001). Gender differences in self-concept and psychological well-being in old age: A meta-analysis. *The Journals of Gerontology Series B: Psychological Sciences and Social Sciences, 56*(4), P195–P213.

Piro, B. (2017). Understanding the influence of social networks and social roles on individuals with developmental disabilities: Providing opportunities in order to assess quality of life.

Quarrie, J. (1992). Earth Summit'92. The United Nations Conference on Environment and Development, Rio de Janeiro 1992.

Ruddick, L., & Oliver, C. (2005). The development of a health status measure for self-report by people with intellectual disabilities. *Journal of Applied Research in Intellectual Disabilities, 18*, 143–150.

Ryndak, D. L., Jackson, L., & Billingsley, F. (2000). Defining school inclusion for students with moderate to severe disabilities: What do experts say? *Exceptionality, 8*(2), 101–116.

Schalock, R. L. (2000). Three decades of quality of life. *Focus on Autism and Other Developmental Disabilities, 15*(2), 116–127.

Schalock, R. L. (2004a). The concept of quality of life: What we know and do not know. *Journal of Intellectual Disability Research, 48*(3), 203–216.

Schalock, R. L. (2004b). The concept of quality of life: What we know and do not know. *Journal of Intellectual Disability ResearchJournal of intellectual disability research, 48*(3), 203–216.

Schalock, R. L., & Keith, K. D. (1993). *Quality of life questionnaire manual*. Worthington, OH: IDS Publishing Corporation.

Schalock, R. L., & Verdugo, M. A. (2012). A conceptual and measurement framework to guide policy development and systems change. *Journal of Policy and Practice in Intellectual Disabilities, 9*(1), 63–72.

Schalock, R. L., Verdugo, M. A., & Braddock, D. L. (2002). *Handbook on quality of life for human service practitioners*. Washington, DC: American Association on Mental Retardation.

Schalock, R. L., Verdugo, M. A., & Gomez, L. E. (2017). Translating the quality of life concept into practice. In *Handbook of positive psychology in intellectual and developmental disabilities* (pp. 115–126). Cham: Springer.

Schalock, R. L., Verdugo, M. A., Jenaro, C., Wang, M., Wehmeyer, M., Jiancheng, X., & Lachapelle, Y. (2005). Cross-cultural study of quality of life indicators. *American Journal on Mental Retardation, 110*(4), 298–311.

Schneider, M. (2005). The quality of life in large American cities: Objective and subjective social indicators. In *Citation classics from social indicators research* (pp. 101–115). Dordrecht: Springer.

Schoch, R. (2006). *The secrets of happiness: Three thousand years of searching for the good life*. New York: Simon and Schuster.

Shogren, K. A., & Shaw, L. A. (2016). The role of autonomy, self-realization, and psychological empowerment in predicting outcomes for youth with disabilities. *Remedial and Special Education, 37*(1), 55–62.

Simões, C., & Santos, S. (2017). The impact of personal and environmental characteristics on quality of life of people with intellectual disability. *Applied Research in Quality of Life, 12*(2), 389–408.

Simplican, S. C., Leader, G., Kosciulek, J., & Leahy, M. (2015). Defining social inclusion of people with intellectual and developmental disabilities: An ecological model of social networks and community participation. *Research in Developmental Disabilities, 38*, 18–29.

Smith, R. B., Morgan, M., & Davidson, J. (2005). Does the daily choice making of adults with intellectual disability meet the normalisation principle? *Journal of Intellectual and Developmental Disability, 30*(4), 226–235.

Spreitzer, G. M. (1996). Social structural characteristics of psychological empowerment. *Academy of Management Journal, 39*(2), 483–504.

Stancliffe, R. J., & Keane, S. (2000). Outcomes and costs of community living: A matched comparison of group homes and semi-independent living. *Journal of Intellectual and Developmental Disability, 25*(4), 281–305.

Stancliffe, R. J., & Parmenter, T. R. (1999). The choice questionnaire: A scale to assess choices exercised by adults with intellectual disability. *Journal of Intellectual & Developmental Disability, 24*, 107–132.

Tamaš, D. M. (2015). *Tip stanovanja i kvalitet života osoba sa intelektualnom ometenošću* (Doctoral dissertation, Univerzitet u Beogradu-Fakultet za specijalnu edukaciju i rehabilitaciju).

Thorndike, E. L. (1904). The newest psychology. *Educational Review, 28*, 217–227.

Townsend-White, C., Pham, A. N. T., & Vassos, M. V. (2012). A systematic review of quality of life measures for people with intellectual disabilities and challenging behaviours. *Journal of Intellectual Disability Research, 56*(3), 270–284.

UNCRPD (2006). Convention on the rights of persons with disabilities. Retrieved from http://www.un.org/disabilities

Van Loon, J., Van Hove, G., Schalock, R., & Claes, C. (2009). *Personal outcomes scale: Administration and standardization manual*. Ghent, Netherlands: Stichting Arduin.

Verdonschot, M. M., De Witte, L. P., Reichrath, E., Buntinx, W. H. E., & Curfs, L. M. (2009). Community participation of people with an intellectual disability: A review of empirical findings. *Journal of Intellectual Disability Research, 53*(4), 303–318.

Verdugo, M. Á., Arias, B., Gómez, L. E., & Schalock, R. L. (2010). Development of an objective instrument to assess quality of life in social services: Reliability and validity in Spain. *International Journal of Clinical and Health Psychology, 10*(1).

Verdugo, M. A., Navas, P., Gómez, L. E., & Schalock, R. L. (2012). The concept of quality of life and its role in enhancing human rights in the field of intellectual disability. *Journal of Intellectual Disability Research, 56*(11), 1036–1045.

Verdugo, M. A., Schalock, R. L., Keith, K. D., & Stancliffe, R. J. (2005). Quality of life and its measurement: Important principles and guidelines. *Journal of Intellectual Disability Research, 49*(10), 707–717.

Walsh, P. N., Emerson, E., Lobb, C., Hatton, C., Bradley, V., Schalock, R. L., & Moseley, C. (2010). Supported accommodation for people with intellectual disabilities and quality of life: An overview. *Journal of Policy and Practice in Intellectual Disabilities, 7*(2), 137–142.

Wehmeyer, M. L., & Abery, B. H. (2013). Self-determination and choice. *Intellectual and Developmental Disabilities, 51*(5), 399–411.

White-Koning, M., Arnaud, C., Bourdet-Loubère, S., Bazex, H., Colver, A., & Grandjean, H. (2005). Subjective quality of life in children with intellectual impairment–how can it be assessed? *Developmental Medicine and Child Neurology, 47*(4), 281–285.

WHOQoL Group. (1995). The World Health Organization quality of life assessment (WHOQOL): Position paper from the World Health Organization. *Social Science & Medicine, 41*(10), 1403–1409.

Wolf, S. (1997). Happiness and meaning: Two aspects of the good life. *Social Philosophy and Policy, 14*(1), 207–225.

Wolfensberger, W. (1972). *The principle of normalization in human services*. Toronto, ON: National Institute on Mental Retardation.

7

Service Delivery Models

Anne M. Bowers

Introduction

People with intellectual disabilities may require an array of services to meet their multifaceted health, daily living, and social needs. The implications of these vast needs indicate that a multidisciplinary, person-centered approach is necessary for comprehensive service delivery. There are a variety of ways services can be coordinated and provided. This chapter discusses the development of service provision and reviews the types of services individuals may utilize over time. Four conceptual models demonstrate paths of service delivery for persons with I/DD and illustrate differing mechanisms of coordination and opportunities for self-direction within each framework. These models are considered in context of individual care needs, life stage, and sources of natural support, as well as significant system-level constraints that impact the delivery of services to people with I/DD.

Evolution of Service Delivery

Until the late 1970s, large residential care facilities were the primary source of daily care and long-term services for people with I/DD in the USA. In large congregated institutions, care and services are available around the clock and delivered to residents on-site. Although there are still persons with I/DD in large residential care facilities, these institutions have historically provided costly but often inadequate services and care, giving too little consideration to the respect, dignity, quality of life, and holistic needs of residents. Disability advocates opposed institutionalization for its infringement upon the human rights and autonomy of persons with disabilities. Deinstitutionalization and the Independent Living Movement (ILM) led to increases in the number of individuals living in noninstitutional settings (White, Lloyd Simpson, Gonda, Ravesloot, & Coble, 2010). Today, more people with I/DD are living in communities of their choice. The ILM and disability rights activism has also stimulated the self-advocacy of people with I/DD and furthered the availability and accessibility of education, employment, and social opportunities for community members with disabilities.

Systems of long-term services and supports (LTSS) have shifted from institutionally based care to less restrictive care and services provided in home- and community-based settings (Lind & Archibald, 2013; Turygin, Matson, & Adams, 2014). This transition in the values of service provision moves away from the "medical model, with a strong custodial component" toward a "developmental model, in which enhancement of

A. M. Bowers (✉)
National Council of State Boards of Nursing,
Chicago, IL, USA

J. L. Matson (ed.), *Handbook of Intellectual Disabilities*, Autism and Child Psychopathology Series, https://doi.org/10.1007/978-3-030-20843-1_7

skills and the promotion of social independence is the main goal" (Alvarez, 2016). The shift reflects the increased application of person-centered approaches in service delivery for people with I/DD. Person-centered delivery approaches provide services in context of an individual's care requirements, personal preferences, and environmental circumstances and position users as equal partners in the management and monitoring of their care (Bowers, Owen, & Heller, 2017). In person-centered approaches, services are specifically structured to meet a person's unique health and social needs and actively facilitate the individual's input and involvement with their care planning and coordination. Many contemporary service delivery models for people with I/DD are based in person-centered approaches that promote independent living, community participation, and individual development.

Service Domains

A person-centered approach to service delivery recognizes that people with I/DD need more than just medical-related care. Individuals should be expected to vary in the type and number of services and supports they require. From early childhood to the late stages of aging, individuals with I/DD should engage with a variety of services to maintain their health, fully participate in their communities, and realize their personal aspirations. Service areas (also called domains) can include medical, support, education, social, employment, community living, and long-term care (Table 7.1).

Medical Care

Medical care services include the primary specialty, preventive, rehabilitative, inpatient, outpatient, and emergency health care that an individual may need. Regular medical care services are important for maintaining the health of people with I/DD who are more likely to have call co-occurring medical conditions and complex health profiles. Medical professionals provide diagnostic testing, functional assessment, and evaluation of the disability condition in early life, adolescence, and young adulthood. Clinicians monitor the health of adults with I/DD and provide necessary care, preventive screening, and appropriate medical intervention as these individuals age (Peacock & Rhodes, 2016). People with I/DD may also require more specialized care than other patient groups in every stage of life. Specialists in psychiatry, cardiology, podiatry, neurology, dentistry, occupational therapy, and behavioral health, among others, can all be a part of the multidisciplinary provider team involved in the lifelong care of a person with I/DD.

Direct Support

Direct support services refer to the provision of individual support and general care for persons with I/DD and their families, both in the home and in community settings. The purpose of these services is to support people with I/DD in completing tasks and in achieving their short- and long-term personal goals. Direct support workers may help individuals with basic activities of daily living, such as eating, dressing, grooming, bathing, and getting around house. Direct support workers can also help individuals with I/DD in instrumental activities of daily living, such as cooking, cleaning, shopping, exercising, transportation, taking medication, and managing money. Direct support care help people with I/DD live independently and participate in the community. Support services can also provide respite care services to relieve family caregivers, who provide a sizable portion of direct support for people with I/DD in the USA. Depending on an individual's functional ability, medical complexity, life stage, and level of family involvement, individuals with I/DD may need direct support services at all times, while others may need only part time or intermittent support services.

Table 7.1 Service domains for individuals with I/DD

Domain	Service description	Examples
Medical care	Primary, specialty, preventive, rehabilitative, inpatient, outpatient, and emergency health care that an individual with I/DD may need over the life course	• Diagnostic testing and evaluation • Preventive screenings • Multidisciplinary health care
Direct support	Direct support and general care for individuals with I/DD in home- and community-based settings, including assistance with activities of daily living	• In-home direct support care • Respite services for family caregivers
Community living	Noninstitutional housing, community-integrated dwellings, and residential services for individuals with I/DD	• Independent living • Supported community living arrangements
Education	Special education, transition services, vocational training, and post-secondary education for children and adults with I/DD	• Special education • Transition planning • Vocational training
Employment	Services for adults with I/DD related to finding job opportunities and maintaining regular, paid work	• Independent employment • Supported employment • Job coaching
Social and leisure	Services providing recreational activities, socialization, travel, continuing education, and personal development opportunities for community members with I/DD	• Sports leagues • Social clubs • Volunteering
Long-term care	Institutionally based residential services and care for people with I/DD who may need continuous, intensive medical treatment	• Nursing homes • ICF/MRs

Community Living

Community living services refer to noninstitutional housing and residential services for individuals with I/DD. Noninstitutional residential settings include living at home with family, living independently in an apartment or house (with or without direct support services), or supported community living arrangements. Supported community living arrangements are typically group homes, family homes, and apartments where 2–8 adults with I/DD reside and receive oversight from continuously present direct care providers and support staff. Direct care workers in such group living arrangements are typically employed by a not for profit (NFP) organization or a community developmental services agency. Supported community living can provide more intensive direct care to people with I/DD as needed, as well as assist in the coordination and organization of the array of services utilized by residents. These noninstitutional residential settings serve to promote the community integration and independent living of persons with I/DD.

Education

Educational services include special education, transition services, vocational training, and post-secondary education for people with I/DD. Educators contribute to the diagnosis and evaluation of the disability condition, especially in early life stages of people with I/DD. In primary and secondary school-based settings, students with I/DD and their families and caregivers can participate with teachers and school administrators in developing an individualized education program (IEP) to establish learning accommodations and optimize curriculum delivery and classroom supports for a student. Many students with I/DD are educated in integrated classrooms with the appropriate accommodations. Others may be enrolled in special education programs throughout their primary and secondary school years. Transition services are another type of educational service that assists individuals in planning and preparing for life after school. Transition services help individuals with I/DD identify their long-term goals and plan for housing, employment, and options for further education. Students with I/DD may elect to receive vocational training to gain skills in a specific career area. Additionally, a growing number of students with I/DD continue with postsecondary education, including college degree and certificate programs.

Employment

Employment services are services for adults with I/DD related to finding job opportunities and maintaining regular, paid work. Employment services are important for the productive community living of adults with I/DD. Many people with I/DD work independently in integrated settings with coworkers who typically do not have disabilities. Employers can provide job accommodations to increase the accessibility of the workplace for employees with I/DD, including plain language training and modified work arrangements. Individuals and their families and caregivers may also partner with local state agencies or NFP organizations that offer supported employment services for community members with I/DD. These organizations can help arrange customized employment opportunities, as well as provide job coaching services for persons with I/DD to assist them in interviewing, performing work-related activities, and engaging with coworkers.

Social and Leisure

Social and leisure services provide opportunities for recreational activities, socialization, travel, and personal development to community members with I/DD. Such services may include intramural and competitive sports leagues; creative art, music, theater, and hobby workshops and classes; adult day programs; summer camps; exercise groups and gym memberships; organized travel and outings; social clubs; religious fellowship; and volunteer activities. Leisure and social services give people with I/DD ways to engage with friends, meet new people, participate in community life, and develop their interests and skill sets. Some social and leisure services provide activities exclusively for individuals with I/DD, while others may be social and leisure activities open to the public that can allow persons with I/DD to integrate with more members of the community.

Long-Term Care

Long-term care services are intensive, institutionally based residential services and care for people with I/DD, including Intermediate Care Facilities for Individuals with Developmental Disabilities (ICF/MR) and nursing homes. Nursing homes are a common long-term care option for persons with I/DD in the advanced stages of aging or who have serious illness. However, younger individuals and adults who have more severe disabilities may sometimes receive their services from in ICF/MRs if they require consistent, active treatment that cannot be received in a community-based setting, like complex medical care, skilled nursing, or multi-provider behavioral health intervention (Turygin et al., 2014). Both ICF/MRs and nursing homes are considered large, residential facilities that are very costly and often inappropriate for members of the I/DD population who can live at home and in the community with the appropriate supports.

Contexts of Provision

The delivery of an array of services to a person with I/DD will be influenced by a variety of factors. Individual health status, medical complexity, life stage, and level of family support may also play a role in how services are provided and coordinated. These contextual elements and their interplay help define an appropriate framework of service delivery.

Health Status and Medical Complexity

Intellectual disabilities are lifelong problems which are often accompanied by co-occurring health issues such as seizures, dementia, and motor disorders (Courtenay, Jokinen, & Strydom, 2010; Matson & Shoemaker, 2009; Turygin et al., 2014). Additionally, comorbid psychopathology and challenging behaviors are much more common among people with I/DD relative to the general population (Matson & Shoemaker, 2009). Complex medical conditions and dual diagnosis can impact the physical mobility, personal capacity, and participation of individuals with I/DD (Matson & Shoemaker, 2009), especially those in their late senior years and those with serious illness. The functional ability and health status of a person with I/DD will determine the number and type of services needed and their degree of application.

The capacity to thrive in community-based settings will be impacted by the healthcare needs and medical complexity of an individual with I/DD. For certain individuals, an appropriate level of care may not be reasonably provided by family members or through community-based programs (Enquist, Johnson, & Johnson, 2012). As more intensive care and services become necessary, increased supports for daily living and decision-making, or long-term care services, may be appropriate (Turygin et al., 2014). These and other health-related factors can dictate an individual's level of need and, in turn, impact the model of service delivery best suited for a person with I/DD.

Changing Needs with Age

As a person with I/DD ages, their medical, education, direct support, social, and employment service needs should also be expected to change. In childhood and early adult life stages, education, employment, and community living services will have greater importance for people with I/DD. Individuals with I/DD are more likely to have co-occurring health conditions that will increase the importance of medical care and direct support services (Peacock & Rhodes, 2016). Older adults with I/DD have earlier onset dementia and are more likely to exhibit psychiatric symptoms with age. As an individual with I/DD enters their senior years, long-term care services may be needed when an individual is no longer able to live independently in their home (Turygin et al., 2014).

Transitions from childhood to adulthood and beyond will affect personal circumstances, health status, and the number and type of services required for a person with I/DD (Peacock & Rhodes, 2016). The fluctuating importance of certain care and services in the various stages of life may impact the model of service delivery experienced by an individual with I/DD as their needs change over time. For example, as persons with I/DD grow older, their ability to self-direct and independently coordinate their services could decline.

Family Involvement

Family involvement has become a significant source of support for people with I/DD, especially as services and care were made more available in home and community settings. The shift to community living has enabled more individuals with I/DD to receive support from family members throughout their lives. An increasing amount of people with I/DD are living at home with family members or are living independently with the support of nearby family members than in previous decades.

The involvement of family members can influence the model of service delivery for an individual with I/DD. Support from family can impact how services are delivered to a person with I/DD, as family members may provide direct care, service coordination, finance management, supported decision-making, or assistance with individual and future planning as needed on an ongoing, often informal basis (Heller, Arnold, McBride, & Factor, 2012). Family members may play the role of primary service coordinator for an individual with I/DD. Advocacy from family members can help individuals with I/DD have more input and control over their service planning and care (Heller et al., 2012). A family member or caregiver may also petition for legal guardianship of an adult with I/DD, where the guardian is appointed to make decisions on behalf of the person with I/DD (Wood, Edwards, & Hennen, 2016). The degree of family involvement is contingent on the availability and willingness of family members to provide support, as well as their ability to provide the level of care required to meet a person's needs.

Models of Service Delivery

Engagement with multiple service domains, along with the contextual factors affecting delivery, determines the appropriate service delivery model for an individual with I/DD. Four conceptual models—uncoordinated, coordinator-supported, residence-based, and self-directed—represent common frameworks of service delivery for persons with I/DD in the USA. Each model has implications for comprehensive service delivery, as well as the level of self-determination and involvement of the individual receiving care. These model frameworks vary in their locus of control and method of coordination to demonstrate the spectrum of possibilities for service delivery to the I/DD population (Henman & Foster, 2015).

Uncoordinated Service Delivery Models

In an uncoordinated delivery model, a variety of services are delivered to a person with I/DD as available and deemed necessary. Arrows pointing inward from service domains toward the individual with I/DD, as shown in Fig. 7.1, position the person with I/DD as a passive recipient of services across domains in uncoordinated delivery models. Such a model depicts service delivery in its most rudimentary form; services are provided through multiple independent channels and are oftentimes sourced through different funding streams. Siloed service delivery creates multiple points of contact, leaving opportunities for cross-domain coordination deeply fragmented (Henman & Foster, 2015).

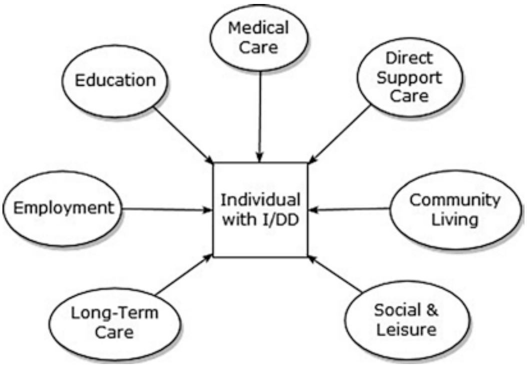


Fig. 7.1 Uncoordinated delivery model

Persons with I/DD essentially have services done to them in uncoordinated service delivery (Henman & Foster, 2015). Recipients are subject to inadequate, imposed, and likely disorganized service delivery in these types of frameworks. Consequently, uncoordinated models often transpire in service delivery to people with I/DD who cannot participate in planning and coordination activities or who have limited self-agency due to the nature of their disability condition, terms of guardianship, or otherwise. User engagement may also depend on residential setting, age, as well as program availability, eligibility, and accessibility, which can impact the achievement of comprehensive service provision in uncoordinated models of delivery for people with I/DD.

Coordinator-Supported Service Delivery Models

In the coordinator-supported delivery model, one or more case workers or service coordinators initiate, organize, and coordinate the care of an individual, as indicated by the arrows pointing outward from coordinators (shaded in gray) toward various, discrete services in Fig. 7.2. Coordinators then ensure the services are received, as illustrated by the arrows pointing from coordinators inwards toward the person with I/DD. A single case worker or coordinator may plan and organize all a person's services across domains (sometimes known as a "case manager"). Alternatively, multiple case workers or service coordinators may be involved in the delivery of services from separate domains; this

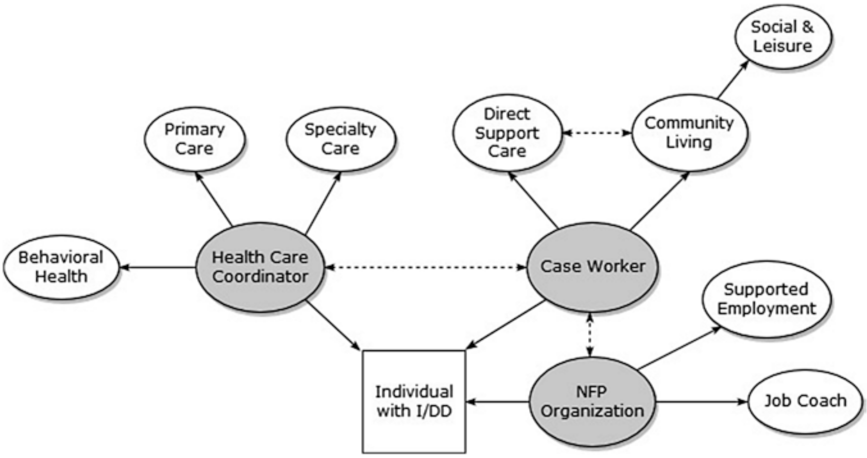


Fig. 7.2 Coordinator-supported delivery model

can be more common when service domains are administered under different funding arrangements (Henman & Foster, 2015).

The individual with I/DD's centralized position in the framework illustrates how coordinator-supported models can allow for better, more comprehensive service delivery. In addition to organizing multiple (and often complexly delivered) discrete services, a defining feature of the coordinator-supported model is linkages of cross-domain service coordination. The dotted-line arrows represent how coordinating entities within and between various domains should communicate regularly and work together to arrange and monitor the rendering of services based on a person's care plan and evolving needs. Improved accessibility, quality, and person-centeredness of service delivery are benefits of enhanced coordination in coordinator-supported models for people with I/DD. However, there is risk of model underperformance if coordinators are inexperienced or have an excessive caseload, have underdeveloped relationships with their clients with I/DD, or organize and direct care without eliciting input and feedback from the individual. If coordinators act as service "gatekeepers," their "role can undermine individual choice, autonomy and control" (Henman & Foster, 2015). Continuously updated individualized service planning is an important key to the success of coordinator-supported models.

Residence-Based Service Delivery Models

Residence-based delivery models share similar features to coordinator-supported delivery models, where a single case manager organizes all services across domains for an individual. However, unlike coordinator-supported delivery, residence-based models are characterized by the centralized coordination of services across domains as an aspect of a person's supported community living arrangement, such as a community-based group home for adults with I/DD (Henman & Foster, 2015). All services are typically organized by one to three residential coordinators who, along with other direct support staff members, are usually employed by NFP organizations or social service agencies that provide community living services for people with disabilities. Residential coordinators frequently provide service coordination for other members of the home.

Residence-based models can streamline the delivery of comprehensive services and supports, as illustrated by the arrow pointing from the residential coordinator toward the person with I/DD in Fig. 7.3. This type of service delivery model can be useful for higher need individuals who need daily direct support care, community living assistance, as well as help with arranging medical care, maintaining supported employment, and

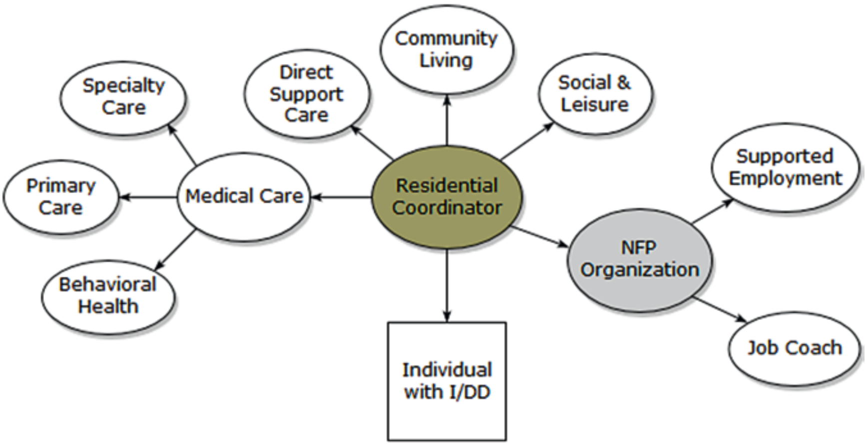


Fig. 7.3 Residence-based delivery model

accessing social activities. Residence-based models can help enhance coordination and delivery when the same services, providers, or community programs are needed and received by more than one member of the household. Much like in coordinator-supported models, impeded choice, autonomy, and control can result from residential coordinators acting as “gatekeepers” to services. This underscores the need for deliberate and continuous person-centered planning with persons who have I/DD and their family members to ensure resident participation, care preferences, and feedback are documented and reflected in provision.

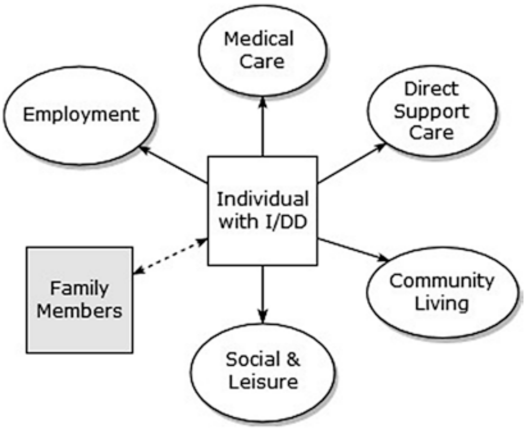


Fig. 7.4 Self-directed delivery model

Self-Directed Service Delivery Models

Self-directed service delivery arrangements are increasingly common since the ILM. In self-directed models, all services across domains are planned and organized by the person with I/DD (Heller et al., 2012). Self-directed delivery epitomizes an ideal model where the individual with I/DD exercises full autonomy and control over their service structure and management (Henman & Foster, 2015). The individual is considered to have the greatest understanding of their needs and wishes for their own care (Henman & Foster, 2015). Accordingly, such models position the individual with I/DD as the central authority of service delivery. The arrows pointing outward from the individual toward their services (Fig. 7.4) show how this model enables the greatest degree of person-centered delivery, with users selecting and scheduling providers and directing services in the way that best meets their individual needs and preferences.

When users coordinate their own care, they assume responsibility for the delivery and payment of services. This may hinder the adoption of self-directed models, especially if accounting for the quality and payment of services is unduly burdensome on an individual. If service availability is sparse, or individuals lack the appropriate funding resources to pay for care, the self-directed model may be ineffective (Henman & Foster, 2015). Familial involvement can be an asset to individuals self-directing their own services. Family members can provide support with navigating delivery systems and transitions, as represented by the dotted-line arrows between the person with I/DD and their family members in Fig. 7.4. Individuals with I/DD may also receive support with self-directed services from a local Center for Independent Living (CIL) or another community agency that advocates for the independent living of people with disabilities. These sources of support are important for many individuals with I/DD living independently in self-directed service arrangements.

The four conceptual models discussed, along with health status and medical complexity, age-related changes in needs, and degree of family involvement, shape the way services are delivered to a person with I/DD. However, service delivery is not impacted by these factors alone. Numerous system-level aspects also affect the provision of services to people with I/DD of all ages.

Delivery System Constraints

Service delivery can be complicated by a number of broader social, environmental, and policy-related contingencies. In most cases, such issues cannot be directly controlled by persons receiving care. Rather, these factors require attention and innovation at the system level to improve service delivery to community members. Delivery model success for individuals with I/DD will depend on service availability, accessibility, quality, and system integration.

Availability and Access

Accessing home- and community-based services (HCBS) is a major determinant of delivery model feasibility. When the full spectrum of services is locally unavailable, people with I/DD may not receive all they care they need in community settings. Low service access is largely driven by inadequate networks of HCBS providers, especially in rural areas of the country. This can contribute to the risk of unnecessary institutionalization of people with I/DD. Along with federal and state governments, administrators of Medicaid and Medicare (the primary providers of healthcare insurance to people with disabilities in the USA) are working to decrease state LTSS waitlists and reform payment structures for improved service access and better provider networks (Enquist et al., 2012; Lind & Archibald, 2013). Still, models of delivery are liable to be constricted by the breadth of available services for persons with I/DD in the community. Coordinator-supported models may predominate if service access is limited enough to preclude opportunities for self-direction.

Service Quality

The common conceptualized models of delivery for people with I/DD assume that provision is achieved when a service is accessed. However, a critical obstacle to delivery model effectiveness is the extent to which members of the I/DD population can obtain needed services that are of high quality. Although services may be available, there is no guarantee they provide timely, person-centered care for a person with I/DD. For example, many primary and specialty care providers have little medical training in intellectual and developmental disabilities and choose not to treat patients with I/DD due to low reimbursement rates from payers such as Medicaid and Medicare (Silberman et al., 2009; Williams & Ervin, 2016). Incomplete standard measures of quality for disability-related services add to the extensive variability in care delivered to people with I/DD (Enquist et al., 2012; Iezzoni, Marsella, Lopinsky, Heaphy, & Warsett, 2017). Without change, quality-related issues will continue to hamper effective service delivery to the I/DD population.

Workforce Challenges

Another major issue impacting individuals with I/DD living in the community is the limited number of qualified, professional in-home caregivers. The shortage of direct support workers (DSWs) in the USA has been identified as a concerning industry trend (Brown, Lash, Wright, & Tomisek, 2011). DSWs make independent and community-supported living viable for individuals with disabilities who may require daily support and care at home. However, DSWs are frequently underqualified, undertrained, underpaid, and insufficiently available in rural regions (Brown et al., 2011; Enquist et al., 2012). Oftentimes, family members step in to provide care for people with I/DD if professional direct support services are unavailable. The scarcity of DSWs places additional stress on the families of individuals with I/DD to informally deliver needed support. Overreliance on families to provide coverage for gaps in needed care is not a sustainable path for community-based service delivery (Bowers et al., 2017). Nevertheless, shortages of DSWs and other providers of HCBS make it increasingly difficult for people with I/DD to access and self-direct care services.

System Integration

Many individuals with I/DD are subject to siloed service delivery. Services for people with I/DD are often delivered by different agencies and through separate funding arrangements, commonly leading to misalignment (Alvarez, 2016; Lind & Archibald, 2013). As previously discussed, this creates numerous points of contact and fragments cross-domain coordination. Divisions in delivery structures may engender service underuse, misuse, and the likelihood of unmet needs. Extensive coordination efforts are typically necessary to prevent service disruptions. The complexity of multichannel service provision can be problematic for achieving person-centered delivery and maximizing the potential for self-direction by individuals with I/DD. Integrated systems, on the other hand, bring service domains together to streamline their provision, management, and financing. System integration creates a centralized point of planning and contact for users and decreases the need for cross-coordination efforts. In this way, integrated systems can better support person-centered delivery by reducing possibilities for gaps in needed care related to service access and coordination issues. Self-directed structures may better flourish in simplified, integrated systems that facilitate user-coordination of multiple services.

Conclusion

Service delivery to the I/DD population has evolved over time. The growth in community-based services signaled a fundamental change in service provision to people with I/DD from primarily medicalized models to person-centered approaches to care. Person-centered approaches offer services in context of an individual's needs and preferences and prioritize user involvement in care planning and management. This is important for individuals with I/DD who may receive an array of services over the life course, including medical, direct support, community living, education, employment, social and leisure, and long-term care services. Person-centeredness supports comprehensive provision to meet all an individual's needs.

Delivery framework utilization and the degree of user involvement can be impacted by several contextual factors, including co-occurring health issues and complex medical conditions, changing needs with age, and family involvement. Four conceptual models—uncoordinated, coordinator-supported, residence-based, and self-directed—are identified as common methods of service delivery for persons with I/DD. Each of these models differs in its mechanisms of coordination and potential to support self-determination in service provision. Still, improvement is needed at the system level to expand the availability, accessibility, quality, and integration of services. Without system innovation, such issues will continue to shape opportunities for effective, person-centered provision and user self-direction in service delivery for individuals with I/DD.

References

Alvarez, N. (2016). Large Residential Care Facilities. In *Health care for people with intellectual and developmental disabilities across the lifespan* (pp. 265–275). Cham: Springer.

Bowers, A., Owen, R., & Heller, T. (2017). Care coordination experiences of people with disabilities enrolled in Medicaid managed care. *Disability and Rehabilitation, 39*(21), 2207–2214.

Brown, D., Lash, S., Wright, B., & Tomisek, A. (2011). *Strengthening the direct service workforce in rural areas*. Washington, DC: Centers for Medicare and Medicaid Services, National Direct Service Workforce Resource Center. Retrieved from https://www.medicaid.gov/medicaid/ltss/downloads/workforce/rural-area-issue-brief.pdf

Courtenay, K., Jokinen, N. S., & Strydom, A. (2010). Caregiving and adults with intellectual disabilities affected by dementia. *Journal of Policy and Practice in Intellectual Disabilities, 7*(1), 26–33.

Enquist, G., Johnson, C., & Johnson, W. (2012). *Trends and challenges in publicly-financed care for individuals with intellectual and developmental disabilities*. Chicago, IL: Center for Health Care Strategies. Retrieved from http://www.chcs.org/media/IDD_Service_Delivery_Systems_082812.pdf

Heller, T., Arnold, K., McBride, E., & Factor, A. (2012). Consumer-directed support: Impact of hiring practices on adults with I/DD and families. *International Journal of Integrated Care, 12*, 76–87.

Henman, P., & Foster, M. (2015). Models of disability support governance: a framework for assessing and reforming social policy. *Australian Journal of Social Issues, 50*(3), 233–252.

Iezzoni, L. I., Marsella, S. A., Lopinsky, T., Heaphy, D., & Warsett, K. S. (2017). Do prominent quality measurement surveys capture the concerns of persons with disability? *Disability and Health Journal, 10*(2), 222–230.

Lind, A., & Archibald, N. (2013, February).*Structuring new service delivery models for individuals with intellectual and developmental disabilities*. Chicago, IL: Center for Health Care Strategies. Retrieved from http://www.chcs.org/media/New_Service_Delivery_Models_for_IDD_020413.pdf

Matson, J. L., & Shoemaker, M. (2009). Intellectual disability and its relationship to autism spectrum disorders. *Research in Developmental Disabilities, 30*(6), 1107–1114.

Peacock, G., & Rhodes, C. (2016). Integrating systems of care for individuals with intellectual and developmental disabilities across the lifespan. In *Health care for people with intellectual and developmental disabilities across the lifespan* (pp. 123–134). Cham: Springer.

Silberman, P., Bodfish, J., Brown, A. T., Wainwright, L., Yorkery, B., Lichstein, J., … Holmes, M. (2009). Successful transitions for people with intellectual and other developmental disabilities. *North Carolina Medical Journal, 70*(6), 519–526.

Turygin, N., Matson, J. L., & Adams, H. (2014). Prevalence of co-occurring disorders in a sample of adults with mild and moderate intellectual disabilities who reside in a residential treatment setting. *Research in Developmental Disabilities, 35*(7), 1802–1808.

White, G. W., Lloyd Simpson, J., Gonda, C., Ravesloot, C., & Coble, Z. (2010). Moving from independence to interdependence: A conceptual model for better understanding community participation of centers for independent living consumers. *Journal of Disability Policy Studies, 20*(4), 233–240.

Williams, A., & Ervin, D. A. (2016). Integration of mental and behavioral health in primary care. In *Health care for people with intellectual and developmental disabilities across the lifespan* (pp. 1699–1707). Cham: Springer.

Wood, D., Edwards, L. R., & Hennen, B. (2016). Health care transition. In *Health care for people with intellectual and developmental disabilities across the lifespan* (pp. 219–228). Cham: Springer.

Comorbidity and Intellectual Disability

8

Larry Burd, Michael Burd, Marilyn G. Klug,
Jacob Kerbeshian, and Svetlana Popova

Comorbidity has been defined as the "existence or occurrence of any additional entity during the clinical course of a patient who has the index disease under study" (Feinstein, 1970) Comorbidity is commonly used to refer to the presence of a second or multiple conditions alongside a primary condition of interest. In a technical context, the term refers to the greater than chance likelihood that the presence of one condition will be accompanied by a second or multiple conditions. The presence of the comorbidities would be observed in the population at a higher rate than the product of the base rates would suggest. In this chapter, we concentrate on comorbidity in the context of developmental neuropsychiatric disorders primarily in children.

Comorbidity is often anticipated as a component of many disorders (Burd, Klug, Martsolf, & Kerbeshian, 2003; Feinstein, 1970; Kerbeshian & Burd, 2000). For example, people with intellectual disability (ID) may also have increased rates of attention deficit hyperactivity disorder autism and epilepsy in comparison to people without ID (Burd et al., 2003; Burd, Li, Kerbeshian, Klug, & Freeman, 2009; Kerbeshian & Burd, 2000; Peng, Hatlestad, Klug, Kerbeshian, & Burd, 2009; Weyrauch, Schwartz, Hart, Klug, & Burd, 2017 May). Comorbidities are important from a clinical standpoint as they influence disease activity, prognosis, medication choice, adverse effects, treatment response, patient compliance, and health care costs (Aslam & Khan, 2018). Comorbidities often have a major negative impact on quality of life, increasing functional disability, independent of disease activity. Comorbidities increase the risk for suboptimal care of coexisting disorders and the psychosocial milieu that accompany these comorbidities.

Diagnostic approach is central to understanding comorbidity. Traditionally, diagnosticians could be conceptualized as "lumpers" who *perceive* a broad phenotype encompassing multiple impairments or various behavior disorders as components of the phenotype. Lumpers tend to see conditions as hierarchical in nature, with

L. Burd (✉)
North Dakota Fetal Alcohol Syndrome Center,
University of North Dakota School of Medicine and
Health Sciences, Grand Forks, ND, USA
e-mail: larry.burd@und.edu; larry.burd@med.und.edu

M. Burd
Department of Pediatrics, University of North Dakota
School of Medicine and Health Sciences,
Grand Forks, ND, USA

M. G. Klug
Department of Population Health, University of
North Dakota School of Medicine and Health
Sciences, Grand Forks, ND, USA

J. Kerbeshian
Psychiatrist, Prairie Care Medical Group,
Brooklyn Park, MN, USA

S. Popova
Institute for Mental Health Policy Research,
University of Toronto, Toronto, ON, Canada

J. L. Matson (ed.), *Handbook of Intellectual Disabilities*, Autism and Child Psychopathology
Series, https://doi.org/10.1007/978-3-030-20843-1_8

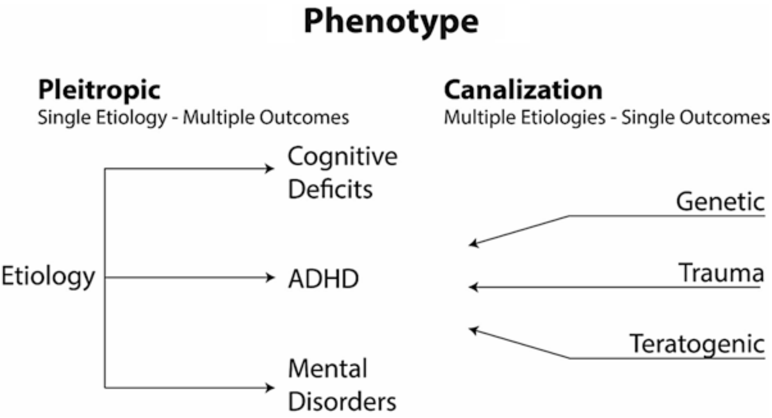


Fig. 8.1 Description of the two most common approaches to diagnoses of neurobehavioral disorders

diagnoses of greater severity, complexity, or impact trumping and incorporating diagnoses of lesser severity, complexity, or impact. Examples of this dynamic can be seen when autism spectrum disorder trumps the specific semantic pragmatic language disorder often present in people with ASD, or in instances where trisomy 21 eclipses problems related to prenatal alcohol exposure. Pleiotropy is roughly the genetic equivalent of the concept of multipotentiality (Fig. 8.1). The term pleiotropy reflects the idea that a common element or a single gene may lead to multiple alternate outcomes or phenotypes, presumably modified by the background of other genes or elements, or epigenetic factors. The concept of equifinality is that of different genes or elements merging into a common outcome or phenotype. The concept of phenocopy also has a similar effect. Canalization refers to a measure of resistance to change of outcome from a single element or a gene, irrespective of the influence of background elements of other genes, epigenetics, or other modifying factors. For example, heavy prenatal use of alcohol may lead to a certain configuration of the eyes and the face, but not to a third eye. Two eyes are heavily canalized, even if there are congenital cataracts, anisophthalmia, etc.

Alternatively, other diagnosticians utilize a broad approach where multiple *different* causes result in a constrained but recognizable pattern or disease phenotype. This approach is *equifinality or convergent homology* canalization (Fig. 8.2). Common examples are genetic causes or environmental causes (prenatal alcohol exposure), intellectual disability, autism spectrum disorders, or cerebral palsy. In practice most clinicians utilize the two approaches together and provide a useful approach to understanding clinically relevant factors that may clarify etiological diagnosis or inform treatment planning (Fig. 8.2). An example of this combined approach can be seen in the diagnosis of fetal alcohol spectrum disorders. Prenatal alcohol exposure is the causal etiology of FASD; however, smoking, maternal depression, prematurity, and drug use are other important elements in the epidemiological causal chain of events, and merit consideration in the diagnosis of fetal alcohol spectrum disorders. These additional elements would be considered as effect modifiers for prenatal alcohol exposure modifying the phenotype both during pregnancy and during postnatal development.

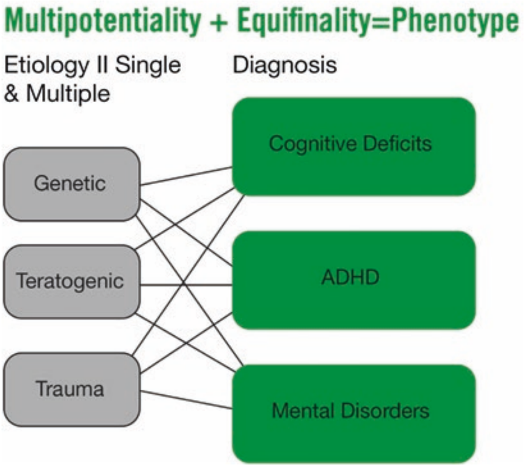


Fig. 8.2 Graphic representation of the components in the construct of multipotentiality

The phenotype of fetal alcohol spectrum disorders is highly variable and is both age and development dependent (Johnson, Moyer, Klug, & Burd, 2017 online first; Weyrauch et al., 2017 May). Clear examples of this variability are the increased risk for development of a substance use disorder *and* contact with the juvenile justice system for individuals with fetal alcohol spectrum disorders. These factors are not apparent in infancy but emerge during childhood and adolescence and may appear as longitudinal comorbidity. In clinical practice it is important to be aware of changes in phenotype in response to age and development. For example, it is important to understand that individuals with FASD have increased rates of contact with juvenile corrections and substance use disorders, and consequently monitor their behavior for signs of issue. These problems are foreseeable risks and should not surprise parents of these children when they reach adolescence. It is important to note that while the risk for these problems is increased in these populations, the problems are not inevitable.

Adverse childhood experiences are frequently seen concurrently with FASD. Adverse childhood events are important effect modifiers, which increases the risk for a variety of problematic health outcomes throughout the life span. In a study comparing FASD participants against non-FASD controls we found large increases in rates of adverse events in patients with FASD compared to controls (in preparation). We examined the role of adverse childhood experiences in people with FASD and the number of comorbid neuropsychiatric disorders compared to the number of adverse childhood experiences and comorbid diagnoses in a group of non-FASD controls. This relationship is summarized in Figs. 8.3 and 8.4.

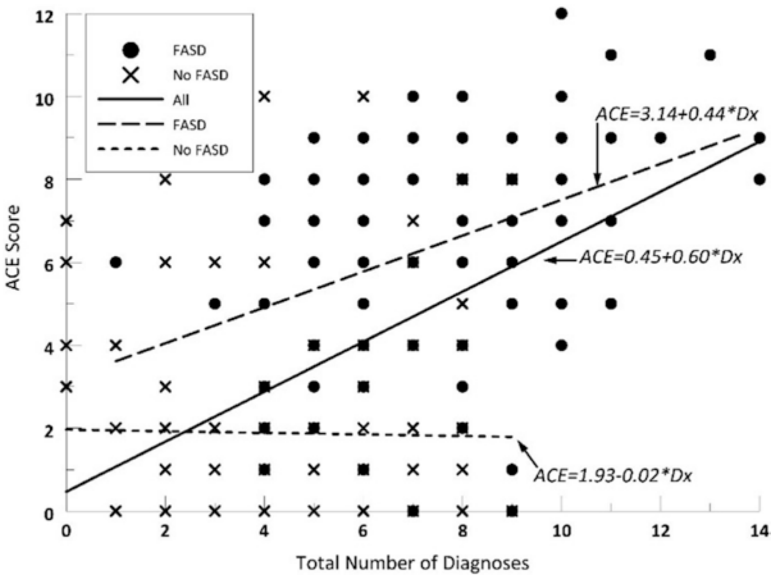
Children with FASD who had multiple diagnoses also had higher ACE scores. A visual depiction of the relationship between ACEs and the number of diagnoses is presented as a nomogram (Fig. 8.3).

The 10 traditional adverse childhood experiences (ACEs) and placement in foster care or residential care programs making a total of 12 possible variables.

The number of adverse childhood experiences appears to be an important effect modifier of risk for comorbid neurodevelopmental disorders in people with FASD but is much less important for non-FASD controls.

In Fig. 8.5 we present a disease specification approach that is also central to the issue of comorbidity. The broad phenome of conditions of interest (childhood developmental disorders) can be examined by the study of multiple phenotypes (phenomics), which comprises a catalogue of phenotypes used to diagnose the different expressions of diseases and disorders.

Fig. 8.3 The relationship between adverse childhood experience and comorbid neurobehavioral diagnoses



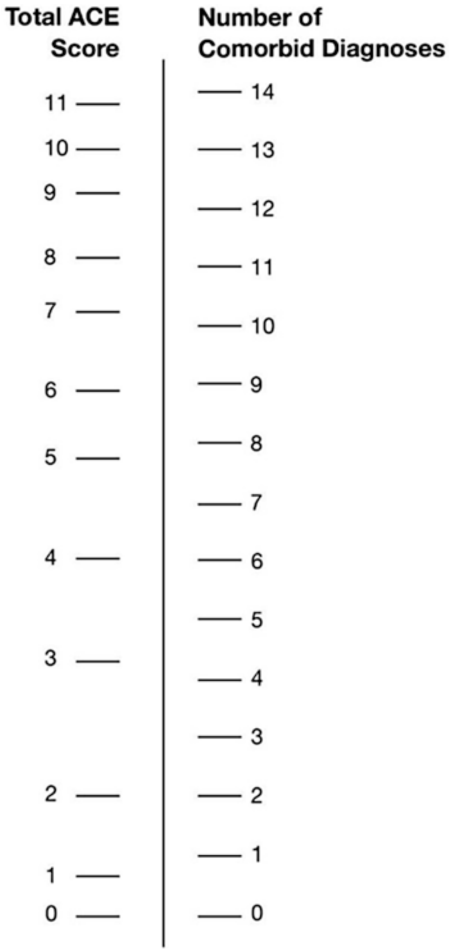


Fig. 8.4 A nomogram depicting the relationship between increasing (ACE) rates of adverse childhood experiences and associated comorbid neurobehavioral disorders

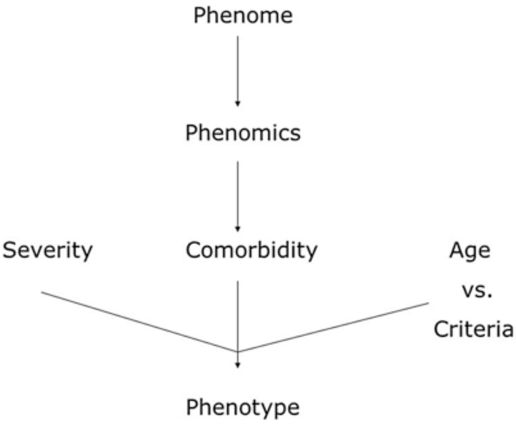


Fig. 8.5 Relationship between all diagnosis phenome and individual disease phenotype in a patient

Most neurodevelopmental disorders have multiple diagnostic features (nosology) that comprise a diagnosis, and these features are typically assessed dimensionally (mild, moderate, severe, or on a symptom diagnostic spectrum such as motor tic disorder, vocal tic disorder, and Tourette syndrome) and on categorical (yes or no) criteria. Figure 8.6 presents the concept of intellectual disability as a comorbid finding in fetal alcohol spectrum disorders and its impact.

Fetal alcohol spectrum disorders and autism spectrum disorders are clear examples of dimensional diagnostic categories which have evolved from more specific categorical diagnoses such as fetal alcohol syndrome, fetal alcohol effects, or alcohol related neurodevelopmental disorders. Autistic disorder, childhood disintegrative disorder, PDDNOS, infantile autism, or childhood onset pervasive developmental disorder also reflects the shift to appreciation of a phenotypic spectrum (Burd, Fisher, & Kerbeshian, 1988; Burd, Fisher, & Kerbeshian, 1989; Fisher, Burd, & Kerbeshian, 1987).

The impact of comorbidity on diagnostic boundaries has profound implications when studying the prevalence, etiology, diagnostic clarity, treatment strategies, and prevention for disorders. A conceptual overview of comorbidity expression suggests that several components merit consideration. Following is a list of the eight components of a comorbidity logic model.

Logic model components of comorbidity

1. Comorbidity increases the complexity of care.
2. Adversity increases the risk for comorbidity.
3. Comorbidity increases the risk for additional comorbidity (the complexity principle).
4. Increasing comorbidity increases diagnostic uncertainty.
5. The prevalence of comorbidity increases with age.
6. Increasing comorbidity increases service utilization and cost of care.
7. Comorbidity decreases access to evidence-based treatments.
8. Comorbidity decreases the likelihood of optimal outcomes.

In clinical practice or research settings, the complexity model may be a function of categorical diagnostic symptom counting (e.g., identification of multiple discreet diagnoses) versus use of

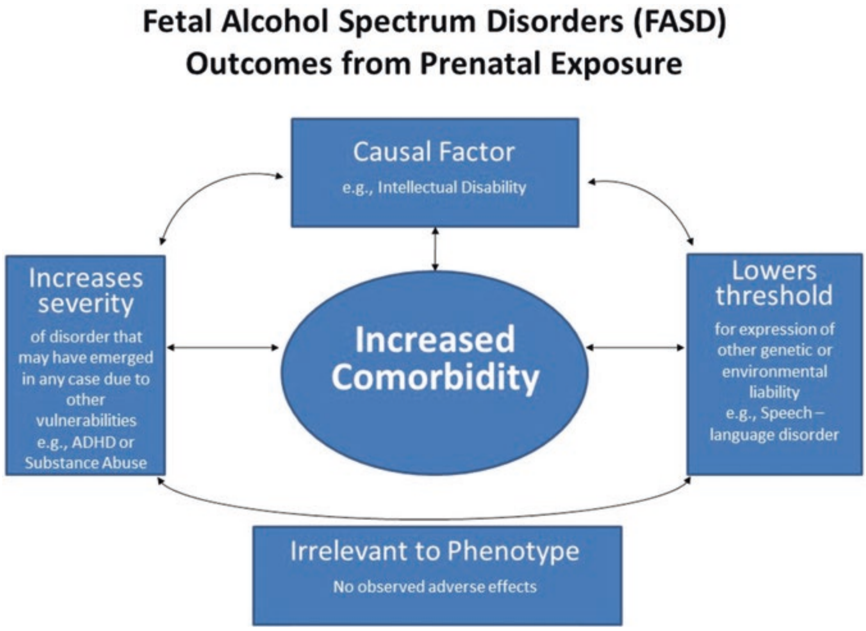


Fig. 8.6 Graphic presentation of factors influencing comorbidity in fetal alcohol spectrum disorders

a broad diagnostic category to subsume the comorbid diagnoses. For example, the overlap of some symptoms of ADHD with symptoms of a manic episode will require fewer additional manic symptom criteria in an ADHD child to make a diagnosis of mania, than in a child without ADHD. Symptoms may be double counted and reduce the threshold for additional comorbidities.

We will provide a brief discussion of each of the logic model components. Where appropriate, we will utilize health claims data and our clinical experience to illustrate each of the comorbidity components. The health claims data and our patient registries have been utilized within other studies of cost and service utilization and phenotype specification (Burd et al., 2001; Burd et al., 2002; Burd, Klug, Coumbe, & Kerbeshian, 2003a; Burd, Klug, Coumbe, & Kerbeshian, 2003b; Peng et al., 2009). For this analysis, we utilized inpatient and outpatient data on 179,873 unique individuals collected over the past two years. The sample's average age was 9.8 years (s.d. 5.4) and ages ranged from 1 year 18 years. This data excluded infants, thus accounting for childbirth expenses, which would skew the results. Just over half of participants (51.1%) were male. The average number of claims per child in the two-year period was 5.1 (s.d. 6.0) and ranged from 1 to 180. We identified 30 individual behavioral diagnoses and the major diagnostic category (MDC) for each claim using the ICD-9 3 digit designations. The average number of behavioral diagnoses per child was 0.14 (s.d. 0.57) and ranged from 0 to 13. After analyzing this data, several conclusions were reached, which are discussed below.

Comorbidity Increases Complexity of Care

It's often difficult for patients or families to decide which diagnoses are priorities for their limited time and funds. Multiple comorbid disorders often require multiple care providers. If a patient sees multiple providers, which one(s) should parents/caregivers prioritize (e.g., home programs for physical therapy, sensory impairments, medications, and/or mental health treatments)? How many appointments at how many different locations are reasonable? Is buying eyeglasses more important than medication for sleep? Patients and caretakers can be overwhelmed by multiple, often conflicting treatment recommendations for patients with comorbidity. This can lead to problems with anger, discouragement, and huge financial burdens for families and health care funders.

In Fig. 8.7 below we present a hypothetical situation depicting increasing complexity of care for a child with increasing comorbidity. The child is born to a mother who was abused and has alcohol use disorder. The father has ADHD, abuses alcohol, and has anger management issues.

This sequence of developmental events increases complexity of care and the cost of care.

Adversity Increases Prevalence and Risk of Comorbidity in Neuropsychiatric Disorders

Prevalence rates of common neurodevelopmental disorders are seen at elevated rates among children with exposure to adverse childhood events (Weyrauch et al., 2017 May). We used claims data for 1506 children with ID to find common comorbid diagnoses. ADHD is by far the most prevalent comorbid condition (42%), and is roughly four times as prevalent as other disorders. Several other disorders were also prevalent among people with ID (Fig. 8.8).

Children with ID are at far greater risk for these comorbidities, when compared to children without ID. However, the risk for a disease doesn't always follow prevalence, meaning that the comorbid disease that children with ID have the most frequently (ADHD) is not the one they are at the most risk for (conduct disorder) (Fig. 8.8). ADHD is very prevalent in children with ID and they are 10 times more likely to have ADHD than other children. However, children with ID are almost 16 times more likely to develop conduct disorder than other children, and 12 times more likely to have an ICD-defined emotional disorder (Fig. 9.9).

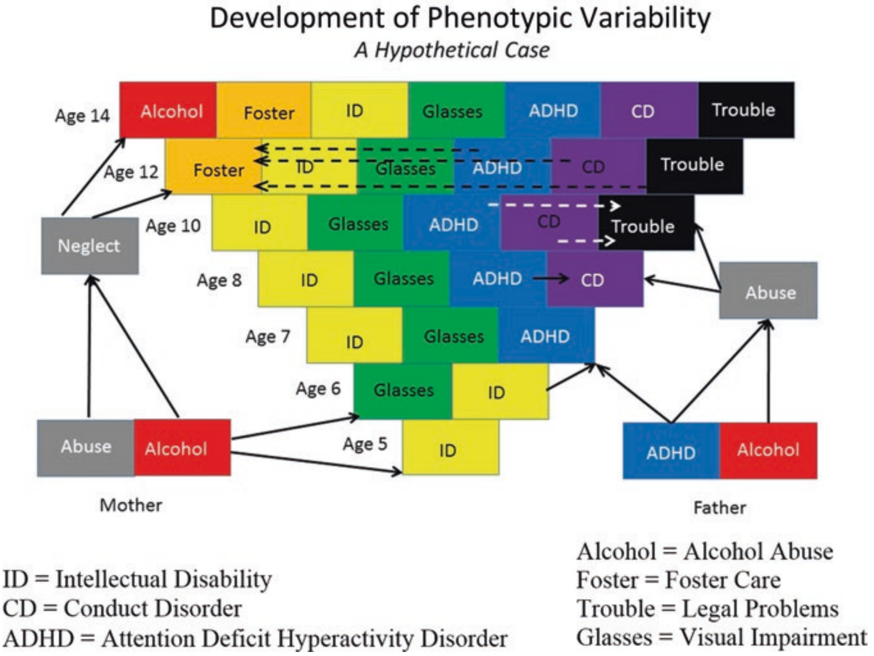


Fig. 8.7 Increasing complexity of ID and the individual phenotype of a person with ID with age. Age 5: The child is diagnosed with an intellectual disability, possibly related to maternal drinking during pregnancy. Age 6: The child needs glasses. Age 7: The child has been diagnosed with ADHD, as is the father. Age 8: The child is abused by the father. Learning and behavior issues emerge at school. Age 10: The child develops conduct disorder. Age 12: Neglect and abuse by the parents, trouble in school, and increasing behavioral impairments lead to foster care placement. Age 14: Substance abuse emerges as a concern. *ID* intellectual disability, *CD* conduct disorder, *ADHD* attention deficit hyperactivity disorder, *Alcohol* alcohol abuse, *Foster* foster care, *Trouble* legal problems, *Glasses* visual impairment

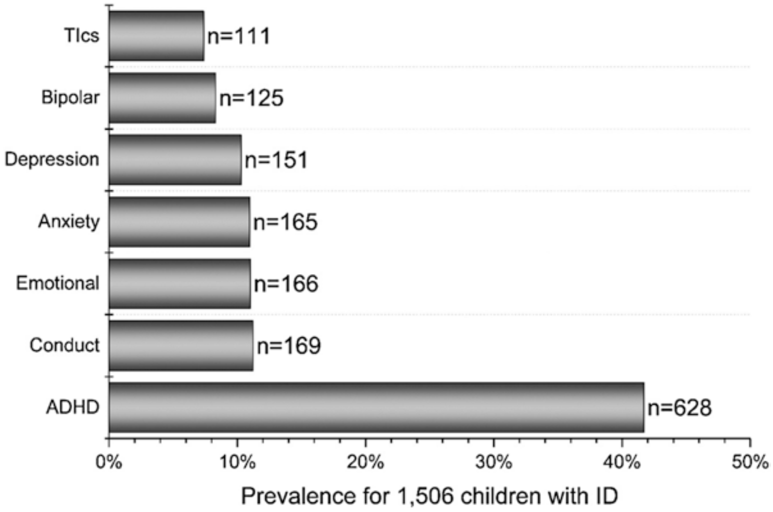


Fig. 8.8 Prevalence of seven most common comorbid behavioral diagnoses for children with ID

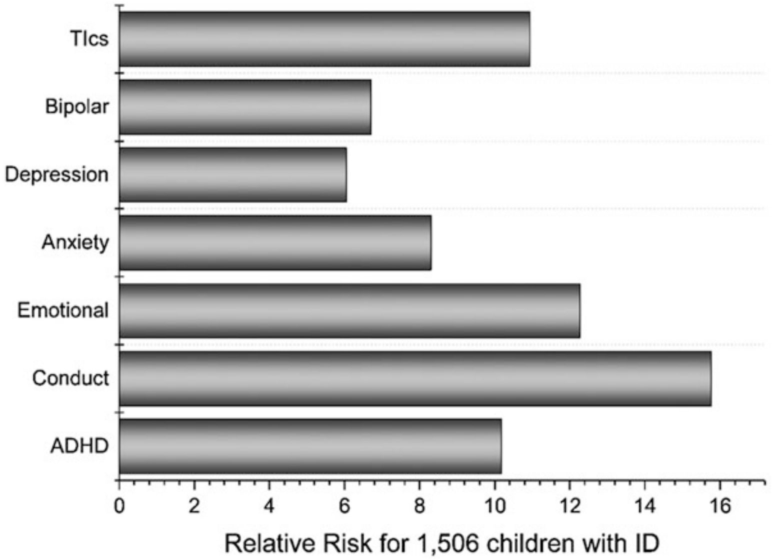


Fig. 8.9 Relative risks of seven most common comorbid behavioral diagnoses for children with ID

Comorbidity Increases Risk for Additional Comorbidities (The Complexity Principle)

Over the past 30 years, we have often observed that the presence of one comorbidity increases the risk for another comorbid disorder. We have described this as the complexity principle. We demonstrate this principle using health claims data for people with ID. As ADHD was the most prevalent comorbid diagnosis seen in children with ID (41.7%) we divided the ID children into those without ADHD (*n* = 878) and those with ADHD (*n* = 628). Figure 8.10 shows the risk of emotional disorder, anxiety, and depression diagnoses for these two groups. ID children with ADHD have a much higher risk of these additional comorbid diagnoses than those without ADHD.

For example, the risk of conduct disorder is 6.6 times more likely for ID children and 28.6 times more likely for ID children with ADHD (compared to children without ID). This means that children with ID and ADHD have over 4 times the risk for conduct disorder than a child

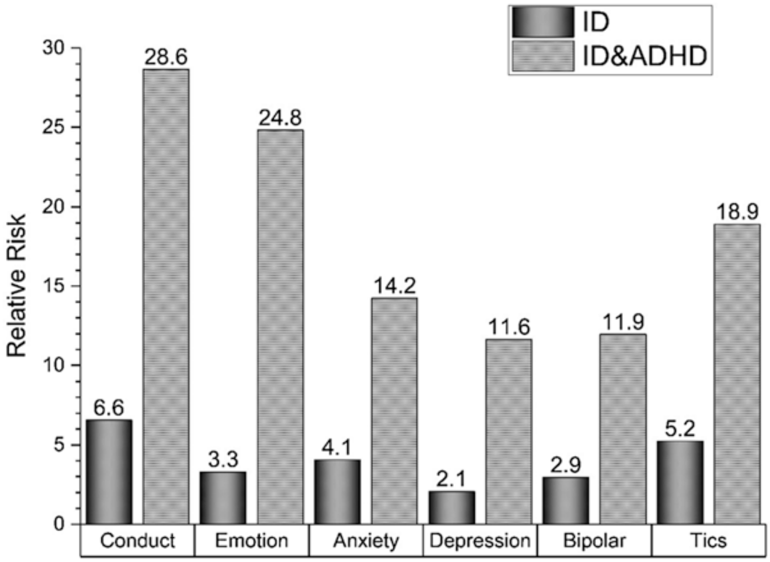


Fig. 8.10 Relative risk of having a second comorbid condition given ADHD previously exists for 1056 children with ID

who just had ID (28.6 compared to 6.6). A child with ID and ADHD is 25 times more likely to have an emotional disorder compared to children without ID, which is 7 times more likely than those with just ID (24.8–3.3). Having ADHD increases the risk of these common conditions by 3.5 (anxiety) to 7.5 times for ICD-defined behavioral and emotional disorders.

This demonstrates that just having one comorbid condition (e.g., ADHD) as well as an existing condition (e.g., ID) greatly increases the likelihood of having another comorbid condition (e.g., conduct or emotional disorder). The likelihood of complexity extends from these examples of having two comorbid conditions to having three or even four comorbid conditions. Figure 8.11 shows 878 children with ID only (left side), and 628 with ID and ADHD (right side). The prevalences of having conduct disorder, anxiety, or emotional disorder diagnoses as second, third, or fourth comorbid conditions are shown in the diagrams inside each box. Though the box on the left (ID only) is larger than the shaded box on the right (ID and ADHD) because it represents more children (878 vs. 628), the figures inside are smaller on the left side indicating only 98 children with ID have a lower prevalence of conduct disorder, anxiety, and emotional disorder as comorbid conditions (compared to 276 on the right, ID and ADHD). As demonstrated in Fig. 8.12, the presence of ADHD alongside ID increases the risk for multiple other neurodevelopmental disorders.

About 5% (3.6% + 0.23% + 0.23% + 0.57%) of the ID children have conduct disorder while 20% (10.2% + 1.9% + 3.8% + 4.5%) of children with ID and ADHD children have conduct disorder (over four times as many). Similarly, 5.4% (4.3% + 0.23% + 0.23% + 0.57%) of children with ID have anxiety and 18.8% (9.6% + 1.9% + 3.8% + 3.5%) of children with ID and ADHD have anxiety. ICD-defined emotional disorders increase from 3% in children with ID only to 22.3% in children with ID and ADHD.

Complexity increases for both groups, when viewing children with two of these comorbid conditions. For the conditions of conduct disorder and anxiety disorder, there are 0.46% (0.23% + 0.23%) of the children with ID and both of those comorbidities, and 5.7% (1.9% + 3.8%) of the children with ID and ADHD have conduct disorder and anxiety, a 12-fold increase. For a combination of anxiety and emotional disorder, 0.8% (0.23% + 0.57%) of children with ID have those two comorbidities, and when ADHD is added as a comorbidity, 7.3% (3.8% + 3.5%) of children have the two additional

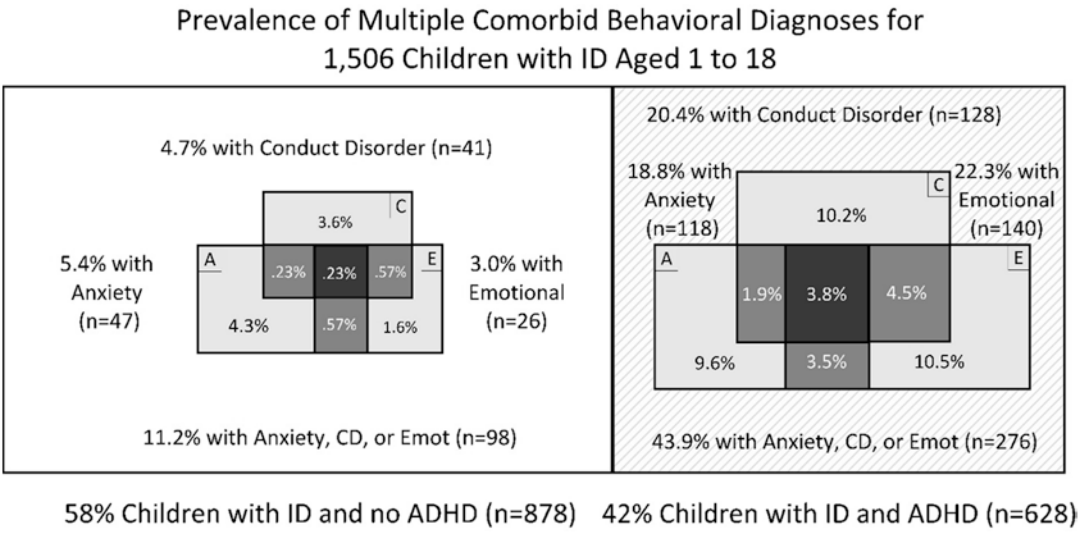
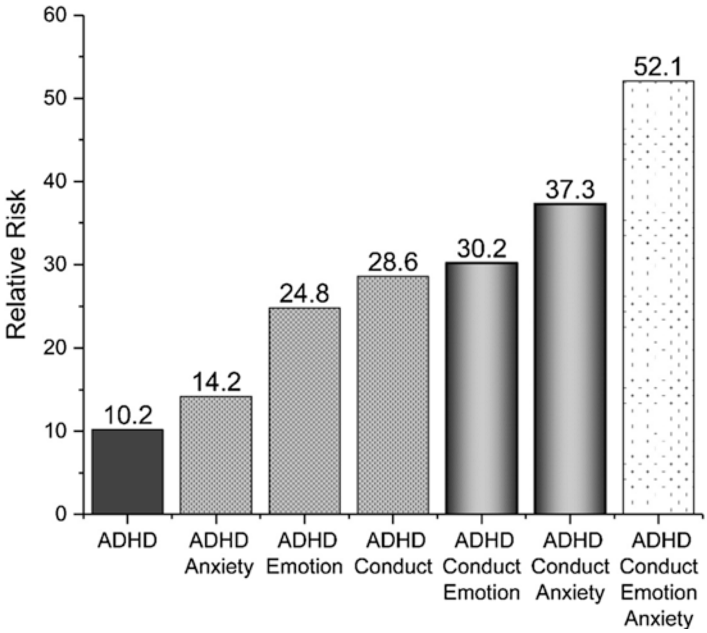


Fig. 8.11 Prevalence of one to three additional comorbid conditions beyond ADHD for children with ID

Fig. 8.12 Relative risk of multiple comorbid conditions in children with ID



comorbidities. Adding more complexity (three of these diseases are comorbid together among both groups of children), we observe that 0.23% of the children with ID have three comorbidities of conduct disorder, emotional disorder, and anxiety disorders (center square). 3.8% or over 16 times that many children with ID and ADHD as a fourth comorbidity have those three diagnoses.

The increase in the likelihood of complexity can also be seen with the relative risks of having one, two, three, or four diagnoses (Fig. 8.12). A child with ID is 10 times more likely to have ADHD than a child without ID (first black bar). The child with ID is 14 to 29 times more likely to have a second comorbid disease, (anxiety, emotional disorder, or conduct disorder, three light gray bars) in addition to ADHD. A child with ID is 30 to 37 times more likely to have two comorbid diseases in addition to ADHD (two vertical striped bars). And finally, a child with ID is over fifty times more likely to have ADHD, anxiety, emotional, and conduct disorder in comparison to a child without ID.

Increasing Comorbidity Increases Diagnostic Uncertainty

As we have demonstrated above, children with ID or other behavioral illnesses are more likely to have comorbid illness, often more than one. Figure 8.13 shows the prevalence of children with two or more MDCs in the two years of claims data gathered. 47% of children with a behavioral diagnosis had two or more MDCs compared to 53% of those without a behavioral diagnosis. This trend quickly changed and children with behavioral diagnoses were more likely to have 3 or more (30 vs. 25%), 4 or more (19 vs. 11%), or even up to 7 or more MDCs. Children with ID (line) were even more likely to have multiple MDCs in the time span with 65% having just two or more MDCs.

There was a high likelihood for children within the sample to have multiple behavioral diagnoses (Fig. 8.14). 71% of children with depression had two or more behavioral diagnoses. This was also true for 59% of children with ID and 39% of children with ADHD. The probability of having three or more comorbidities ranged from 21% to 45% in this sample.

Comorbidity increases diagnostic complexity by presenting multiple disorders simultaneously. Clinicians often diagnose only the most distinctive disorders at the time of the evaluation. It is not uncommon for people to receive multiple different diagnoses as they move from clinician to clinician, and resultantly they often lose an appreciation for the whole child or the child within the context of the family, as they focus on the multiplicity of segmented diagnoses. This is similar to treating parts of the elephant rather than the whole elephant.

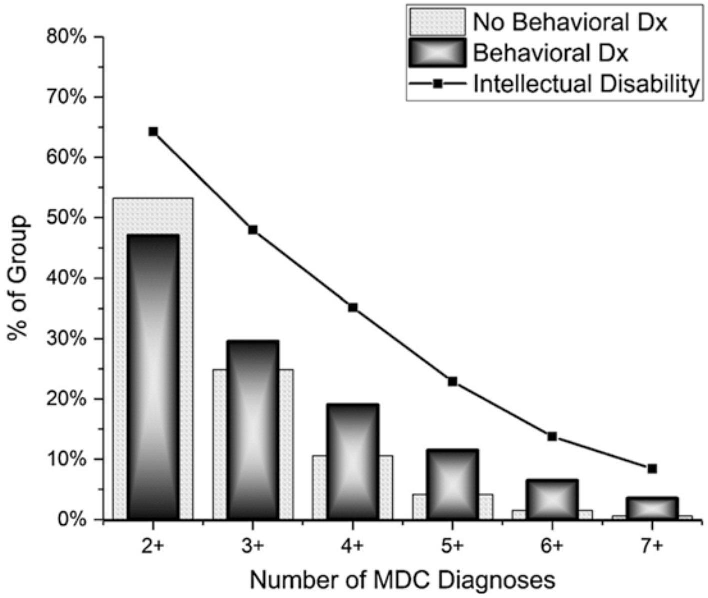
In a study of fetal alcohol spectrum disorder (FASD) we used our FASD patient registry to query the health claims data for information on cost of care and utilization. However, only about 8% of patients on the FASD registry could be identified by diagnosis in the claims data. This is partially because FASD was not specified in the ICD or DSM. The FASD registry contained many patients with 10 or more diagnoses. In the claims data, the diagnosis entered was often the reason for the visit (otitis media, headache). Since FASD was not entered in the registry, our ability to track cost of care, comorbid disorders, or service utilization and to aggregate this data by patient was limited. Even for more severe health related visits in the health claims data (surgical procedures, extensive diagnostic evaluations) FASD was usually not identifiable in the claims data. Children were often noted to have intellectual disability, seizure disorders, attention deficit hyperactivity disorder, or speech and language disorders, but FASD was not a diagnosis included for the visit. We were surprised by this since FASD is likely to be crucial in diagnostic formulation and in treatment planning (*FASD: Diagnosis informed care,* Unpublished manuscript).

The Prevalence of Comorbidity Increases with Age

Recognition of comorbidity is closely related to and dependent on both age and development. Figure 8.15 shows how the average age of children increases as the number of behavioral diagnoses increases. The average age for children with one behavioral diagnosis is 11.4. As the number of diagnoses increases over 8 comorbid conditions, mean age increases to 14.6 years.

Many of the comorbidities we see among patients we care for may have been present earlier but could not be diagnosed until later in

Fig. 8.13 Prevalence of children with multiple MDC diagnoses



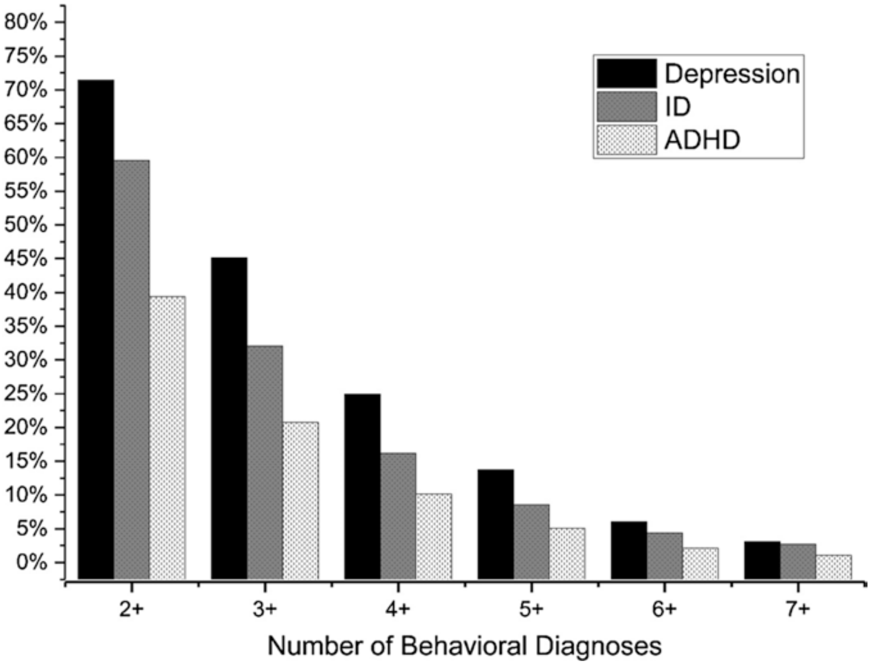


Fig. 8.14 Prevalence of children with multiple behavioral diagnoses by diagnoses

development. Attention deficit hyperactivity disorder, depression, speech and language disorders, and severe mental disorders are relevant examples. The development dependent expression of many of the key criteria for diagnosis is a key to understanding comorbidity (people must achieve a certain developmental level in order for us to reliably diagnose some problems). This duration of effect could be considered as heterotypic continuity, or alternatively longitudinal comorbidity. A classic example of this is seen in the identification of speech or language disorders in children

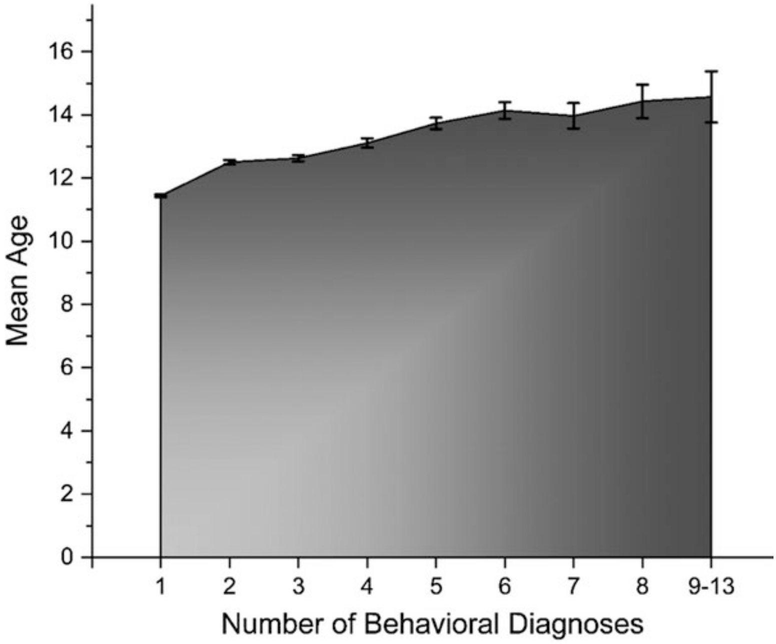


Fig. 8.15 Relationship between mean age and number of behavioral diagnoses for 15,366 children

under 12 months of age or the diagnosis of depression in infancy. A child must have reached a certain age in order to be able to reliably respond to questions about hallucinations or delusions in order for them to receive a diagnosis of some mental disorders. In some cases, children will not be able to express these problems until they reach adolescence. The expression of comorbidity can be limited by developmental impairments. For example, some complex clinical syndromes will be difficult to diagnose in the presence of severe intellectual disability (externalizing disorders or memory impairments).

Increasing Comorbidity Increases Service Utilization and Cost of Care

As is observed in routine health care, people with complicated diagnoses are seen to have higher costs of care. This observation remains true for children with comorbidity. When analyzing the total cost per year (including inpatient and outpatient costs) for 22,994 children with behavioral illness and 1506 with ID, we found the distribution of costs to be highly skewed (Fig. 8.16a). Transforming the data with logarithms showed two distributions of costs, valued under or over approximately 50,000 (Fig. 8.16b). This pattern was similar for the 1506 children with ID, so logarithmic transformations and categories of high and low were used for cost estimates.

Figure 8.17 (four parts) shows the scatter diagrams with regression lines using the number of behavioral diagnoses to predict yearly cost for low spenders (<=$50,000) and high spenders (>$50,000). The top two figures are for lower costs of care, 884 children with ID and 12,535 children with behavioral disorders. Lower costs significantly increase as the number of behavioral diagnoses increases for both groups. The correlation of diagnoses to cost was $r = 0.312$ for ID children and $r = 0.515$ for children with behavioral illness. The bottom two figures are for higher annual costs of care (<$50,000) for both children with ID and behavioral illnesses. The bottom two figures are for higher costs of care, 622 children with ID and 3581 children with behavioral illness. Though the relationships were different, higher costs also significantly increased as the number of behavioral diagnoses increased. Number of behavioral illnesses was significantly correlated with cost for ID children ($r = 0.0841$) and children with behavioral illnesses ($r = 0.123$).

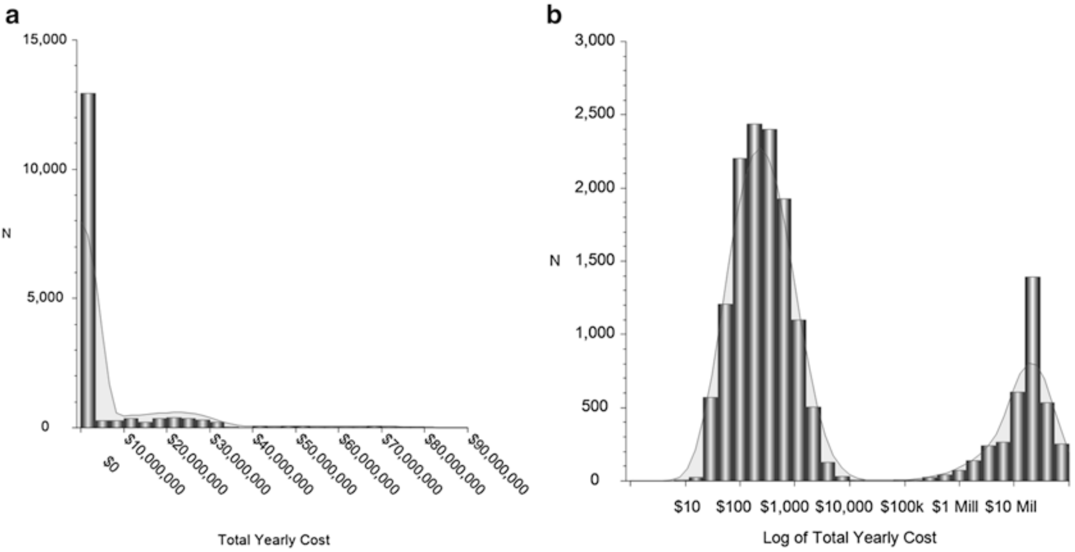


Fig. 8.16 Histograms of number of children in cost categories, original (**a**) and after a logarithmic transformation (**b**)

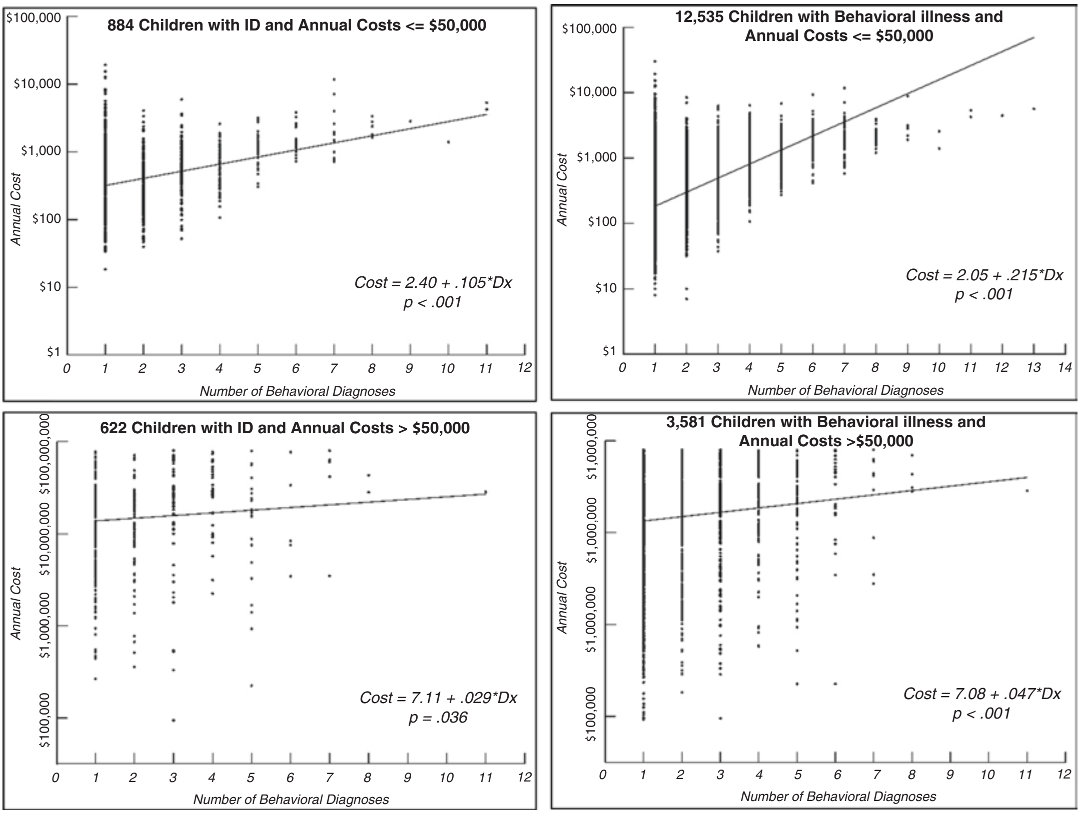


Fig. 8.17 Relationship between number of behavioral diagnoses and increased cost for children with ID or behavioral illnesses

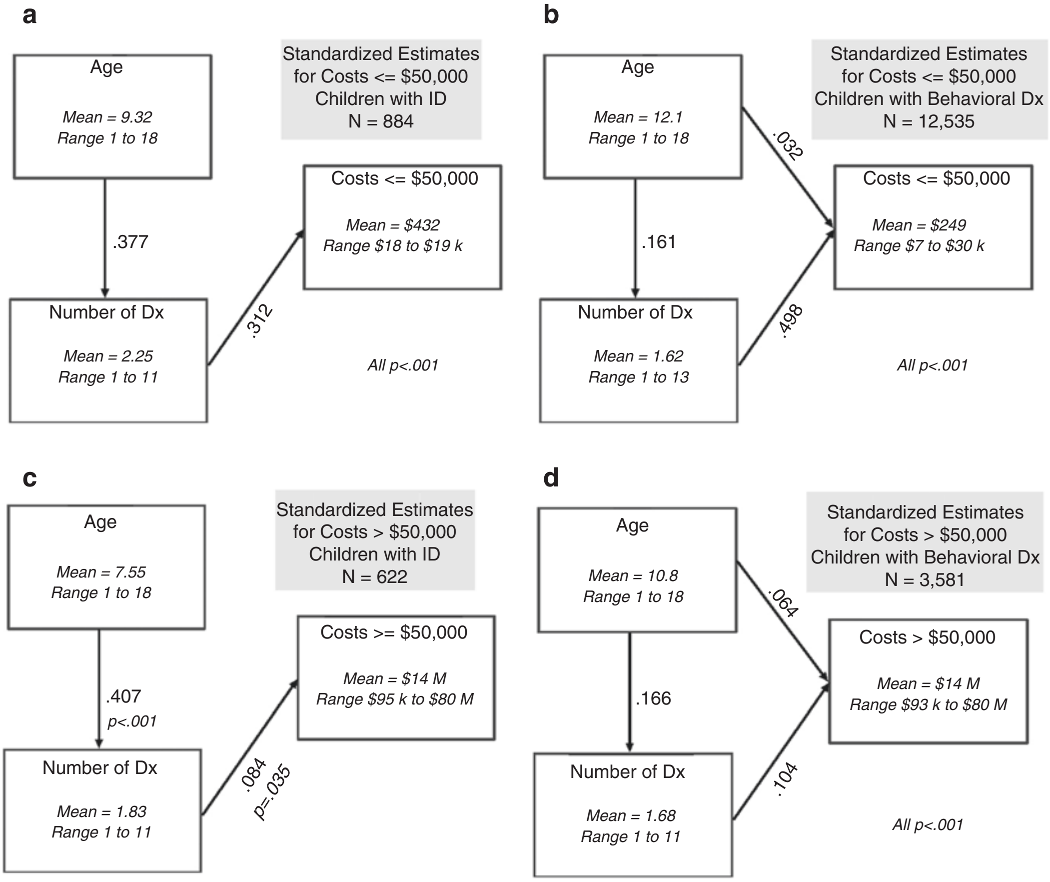


Fig. 8.18 Significant paths relating age and number of diagnoses to high and low costs for children with ID and diagnosed behavior disorder

Age was found to be related to number of behavioral diagnoses (older children had more diagnoses) (Fig. 8.18). Age may also directly or indirectly related to annual cost. Path diagrams can be useful to estimate and understand the full relationships between age, number of diagnoses, and expenses for participants within the high cost and low cost group. The four quadrants of Fig. 8.18 show how age and number of behavioral diagnoses relate to high and low annual costs for children with behavioral diagnoses and ID.

All diagrams show a relationship between age and number of comorbid diagnoses, the relationship is stronger (0.377 and 0.407) for children with ID (a and c). Also, all diagrams show a relationship between number of diagnoses a child has and their annual costs, though this relationship is stronger for children with lower (no expensive hospitalizations) costs (0.312 and 0.498) (a, b, c, d). This creates an indirect relationship between age and annual cost. Age has a direct relationship to annual cost for children with behavioral diagnoses (0.032 and 0.064).

Increasing Comorbidity Decreases Access to Evidence-Based Treatments

In most treatment studies participants are carefully selected to allow for precise examination of the intervention within a minimum sample size. Children with two or more disorders are often excluded from studies and as a result we have limited information on clinical presentation of complex developmental disorders in children and adults.

As a result, we have limited information on the benefits of multiple simultaneous treatments or the risks (adverse effects) from multiple interventions at the same time. This is an area of considerable concern. Do we spend huge amounts of money on interventions which negatively effect (affect) outcomes when delivered together? Are we selecting the optimal mix of treatments that maximize efficiency and benefits when used together? Equally importantly, how do we attribute either harm or benefit when a child is exposed to three, four, or more treatments at the same time? Put another way: Do we have sufficient variance in treatment response to detect either benefit or harm from 3 to 5 treatments utilized simultaneously?

Currently, we seem to operate with the belief that more intervention is better. For example: early intervention services in the home, center-based speech/language therapy, occupational therapy, preschool, and clinical behavior management with medication for sleep and separate medication for attention deficit hyperactivity disorder. This may be a form of therapeutic splitting influenced by both strategies to enhance reimbursement and therapeutic boundaries. Costs are enhanced, but it may be that this is the ideal combination with each intervention enhancing and optimizing the others. It may also be possible that this is just unnecessary duplication (e.g., the structure in preschool, occupational therapy, behavioral consultation, medication, and speech/language interventions overlap). Thus, only 1 or 2 of these interventions may account for the gains and medication or behavior consultation is unnecessary.

Increasing Comorbidity Decreases the Likelihood of Optimal Outcomes

Both the natural history and response to treatment for neurodevelopmental disorders are poorly understood (Burd et al., 2001; Burd et al., 2002; Burd et al., 2003a; Burd et al., 2003b; Kerbeshian & Burd, 2000). The impact of multiple disorders in clinical practice can also result in long-term impacts which can increase the risk for both additional comorbidity or long-term impairment from the combined effects of the disorders (Burd et al., 2003; Burd, 2016). Intellectual disability can have effects on long-term outcomes, and when these effects are combined with additional effects such as comorbid cerebral palsy, attention deficit hyperactivity disorder, or visual impairment, it is clear that more comorbidity nearly always results in additional impairment.

Discussion

Comorbidity increases both cost and utilization of services. The complexity principle is an important concept related to this finding. Increasing comorbidity increases the risk for additional comorbidity and the complexity of care. To illustrate this point, consider a young child who needs speech and language therapy and thus increases the demand on caretakers or parents. This child also needs occupational therapy, physical therapy, early intervention, frequent pediatric visits for multiple medication management, who wears glasses and has a hearing aid represents a huge increase in demand on the child's caretakers. These demands will affect which jobs the parents are able to do, and which promotions that they can accept compared to parents who don't have a child with multiple comorbid conditions.

We conclude with a brief discussion of several key points that have surfaced in this manuscript.

1. We enroll children in developmental therapies using an assumption of endless benefit. We have yet to clearly define benefits or adverse outcomes from multiple simultaneous therapies. We use therapy or multiple therapies as benign interventions and currently associate no downside from using multiple therapies concurrently.
2. Complexity increases both cost and utilization of services. Increasing comorbidity increases complexity which increases both service utilization and cost of care. For example, we have reviewed cases recently where children were sent home after multi-disciplinary assessments with dozens of recommendations.

Parents are forced to choose between what care the child needs and what care can be realistically provided. These choices are difficult as they pose limited affordable treatment options against the numerous treatments that were recommended. The level of care that can be implemented may be determined by travel schedule to different therapies, available time, and financial costs of these therapies. This is especially relevant for families with other children or where more than one child has ongoing health needs.

3. The uncovered or uncompensated cost of these therapies has been largely understudied. This concern applies even if services are funded by health insurance in countries with government-funded care. The implications of parents spending large amounts of money early in the child's life on therapies, travel, and time off from work, where the outcome is highly individualized, often limits funds they have for the care of themselves or other family members and for these children as they become older. Many parents describe this experience as "it's just often too much to hear about." They report feeling overwhelmed by the demands from multiple health care providers and rarely have access to someone to help them prioritize recommendations and to help allocate their resources.
4. Morbidity risk is somewhat predictable for some diagnoses but highly variable in expression. ADHD prevalence is increased in ID. But only some individuals with ID have ADHD. Monitoring for comorbidity should be built into routine care plans with systematic monitoring included within the context of ongoing care. A useful and emerging example is the understanding that increased adverse childhood events increase the risk for other neuropsychiatric diagnoses (submitted). There is an urgent need for strategies which assess whether or not we can prevent or diminish the prevalence and impact these comorbid conditions with early developmental interventions. The optimal developmental timing for these services also merits further examination. Further data regarding the intensity of services and what combinations are most effective or harmful could also be practically utilized.
5. Additionally, we need to optimize strategies and developmental interventions to most effectively treat comorbidity. For example, many children end up on multiple medications to treat multiple comorbid disorders. In some cases, these medications have an evidence-base for individual disorders (attention deficit hyperactivity disorder, depression, or obsessive compulsive disorder) in children. However, large-scale trials of routine clinical populations where children present three, four, five, or six of these other conditions are lacking. Studies of this type will require huge strategic investments in the future. We have very limited understanding of the potential benefits and risks of polypharmacy. Some agencies and parents refuse medications (especially for mental health diagnoses) even when the medications are well studied and offer demonstrable benefits. This seems to be biologically naïve and attributes a huge amount of a child's developmental impairments to disturbed emotions and relationships. This is often viewed by parents as blame. This is especially prominent in foster care where there is a growing mandate to avoid using medications for young children. Under careful examination, it is clear that this mandate applies mainly to mental health application, and is not seen similarly in regard to general medications. Psychological therapies face less critical assessment, despite possessing less evidence of efficacy for developing children.
6. Identification of optimal strategies early in the developmental course to reduce phenotype severity early in life is still limited. Can either early intervention or optimal intervention strategies prevent comorbidity from developing? Could this lead to a reduction in phenotype severity in the future? In populations of children with multiple complex disorders, we need better evidence to guide our practice. These conclusions suggest that a shift in our research practice is needed. It's time to embrace complexity and to begin studies capable of providing evidence to guide our care for these children and their families.

References

Aslam, F., & Khan, N. A. (2018). Tools for the assessment of comorbidity burden in rheumatoid arthritis. *Frontiers in Medicine, 5*, 39. https://doi.org/10.3389/fmed.2018.00039

Burd, L. (2016). FASD and ADHD: Are they related and how? *BMC Psychiatry, 16*, 325. https://doi.org/10.1186/s12888-016-1028-x

Burd, L., Fisher, W., & Kerbeshian, J. (1988). Childhood onset pervasive developmental disorder. *Journal of Child Psychology and Psychiatry and Allied Disciplines, 29*(2), 155–163.

Burd, L., Fisher, W., & Kerbeshian, J. (1989). Pervasive disintegrative disorder: Are rett syndrome and heller dementia infantilis subtypes? *Developmental Medicine and Child Neurology, 31*(5), 609–616.

Burd, L., Kerbeshian, J., Barth, A., Klug, M. G., Avery, P. K., & Benz, B. (2001). Long-term follow-up of an epidemiologically defined cohort of patients with tourette syndrome. *Journal of Child Neurology, 16*(6), 431–437.

Burd, L., Kerbeshian, J., Westerland, A., Labine, J., Barth, A., Klug, M. G., & Wagner, K. (2002). Prospective long-term follow-up of patients with pervasive developmental disorders. *Journal of Child Neurology, 17*(9), 681–688.

Burd, L., Klug, M. G., Coumbe, M. J., & Kerbeshian, J. (2003a). The attention-deficit hyperactivity disorder paradox: 2. Phenotypic variability in prevalence and cost of comorbidity. *Journal of Child Neurology, 18*(9), 653–660.

Burd, L., Klug, M. G., Coumbe, M. J., & Kerbeshian, J. (2003b). Children and adolescents with attention deficit-hyperactivity disorder: 1. Prevalence and cost of care. *Journal of Child Neurology, 18*(8), 555–561.

Burd, L., Klug, M. G., Martsolf, J., & Kerbeshian, J. (2003). Fetal alcohol syndrome: Neuropsychiatric phenomics. *Neurotoxicology and Teratology, 25*(6), 697–705.

Burd, L., Li, Q., Kerbeshian, J., Klug, M. G., & Freeman, R. D. (2009). Tourette syndrome and comorbid pervasive developmental disorders. *Journal of Child Neurology, 24*(2), 170–175. https://doi.org/10.1177/0883073808322666

Feinstein, A. R. (1970). The pre-therapeutic classification of co-morbidity in chronic disease. *Journal of Chronic Diseases, 23*(7), 455–468.

Fisher, W., Burd, L., & Kerbeshian, J. (1987). Comparisons of DSM-III defined pervasive developmental disorders in North Dakota children. *Journal of the American Academy of Child and Adolescent Psychiatry, 26*(5), 704–710.

Johnson, S., Moyer, C. L., Klug, M. G., & Burd, L. (2017 online first). Comparison of alcohol-related neurodevelopmental disorders and neurodevelopmental disorders associated with prenatal alcohol exposure diagnostic criteria. *Jdbp,*

Kerbeshian, J., & Burd, L. (2000). Comorbid Down's syndrome, tourette syndrome and intellectual disability: Registry prevalence and developmental course. *Journal of Intellectual Disability Research, 44*(Pt 1), 60–67.

Peng, C. Z., Hatlestad, P., Klug, M. G., Kerbeshian, J., & Burd, L. (2009). Health care costs and utilization rates for children with pervasive developmental disorders in North Dakota from 1998 to 2004: Impact on medicaid. *Journal of Child Neurology, 24*(2), 140–147. https://doi.org/10.1177/0883073808321059

Weyrauch, D., Schwartz, M., Hart, B., Klug, M. G., & Burd, L. (2017, May). Comorbid mental disorders in fetal alcohol spectrum disorders: A systematic review. *Journal of Developmental and Behavioral Pediatrics, 38*(4), 283–291. https://doi.org/10.1097/DPB.0000000000000440

Part III

Parent/Professional Issues

Parent and Professional Organizations

9

Claire O. Burns, Georgia Shaheen,
and Shelley R. Upton

Introduction

Many resources exist to support individuals with intellectual and developmental disabilities (IDDs) in their daily life as well as promote inclusion in their communities. Family members, professionals such as clinicians and researchers, and community members play an important role in helping individuals with IDDs access the types of supports and services. To this end, several organizations have been developed to facilitate involvement. These include parent organizations for caretakers and family members as well as professional organizations for those whose vocations involve assisting with services for individuals with IDDs. Still others include community organizations, whose aim is to provide an inclusive and accepting society. This chapter reviews several of these organizations, their goals, and their functions.

History of Diagnosis

The most recent version of the Diagnostic and Statistical Manual, Fifth Edition (DSM-5; American Psychiatric Association, 2013) uses the diagnostic category "intellectual disability" rather than "mental retardation." This change in terminology is reinforced by a federal statute known as "Rosa's Law" (Public Law 111–256) which requires that first person language and the term intellectual disability be used in all federal laws. The law was named after an 8-year-old female with Down syndrome whose state representative and senator initiated legislation to enact this change in terminology. The reasons for this shift are (1) to avoid derogatory and pejorative language and (2) to emphasize the neurodevelopmental nature of the disability and the importance of early intervention (Harris, 2013). Intellectual disability has become commonly used in fields relevant to this population, such as medical and educational fields as well as advocacy groups and lay persons (American Psychiatric Association, 2013). Therefore, to be consistent with the current research and diagnostic practices, the term intellectual disability (ID) will be used throughout this chapter, even in reference to resources and articles published before this change that may have used the term mental retardation.

The current criteria for ID include impairments in intellectual abilities and adaptive skills, both of which are present during the developmental period. Severity levels are classified as mild, moderate, severe, and profound. Severity is determined by the level of impairment across conceptual, social, and practical domains. Prevalence rates of ID have been estimated

C. O. Burns (✉) · G. Shaheen · S. R. Upton
Department of Psychology, Louisiana State University, Baton Rouge, LA, USA

J. L. Matson (ed.), *Handbook of Intellectual Disabilities*, Autism and Child Psychopathology Series, https://doi.org/10.1007/978-3-030-20843-1_9

to be approximately 1%; however, prevalence rates vary across age range and severity level (American Psychiatric Association, 2013).

Although ID is a heterogeneous disorder that sometimes has no identifiable etiological cause, there are often prenatal, perinatal, and postnatal factors that are causally related to ID. These include genetic syndromes such as chromosomal disorders or sequence or copy number variations of a variety of genes. There are also high rates of co-occurring psychological and neurodevelopmental disorders in individuals with ID, including attention-deficit/hyperactivity disorder, mood disorders (i.e., depressive, anxiety, bipolar disorders), and autism spectrum disorder (American Psychiatric Association, 2013).

History of Services

A major shift in the field of developmental disabilities was marked by the administration of President John F. Kennedy. Prior to this time, services and supports for individuals with IDDs were minimal. The Kennedy family took an interest in the field of intellectual disabilities and founded the Joseph P. Kennedy Jr. Foundation to advance the field. President Kennedy's sister, Eunice Kennedy Shriver, in particular, was a major proponent of the advancement of advocacy for individuals with IDDs, and founded or laid the groundwork for many of the organizations reviewed in this chapter, such as the Special Olympics. Her son, Anthony Shriver followed in her legacy to found the Best Buddies organization (John F. Kennedy Presidential Library and Museum, n.d.).

In 1961, President John F. Kennedy created a Panel on Mental Retardation, which recommended improvements in service programs as well as research efforts. A meeting was held with representatives serving as coordinators of mental retardation programs from 38 states, with a stated goal that "the new organization would provide a vehicle for effective communication between the federal agencies and the various states concerned with research, training, programs and finance" (NASDDDS, 2018). One of the primary focuses of the report from the panel was the need for additional interdisciplinary research, with the recommendation that research institutes be created in academic settings such as universities. An additional goal was increased access to education for individuals with IDDs. Lastly, the third focus was on a "continuum of care" that included educational, social, and medical services in order to decrease impairments throughout the life span (Berkowitz, 1980). This panel resulted in the creation of the Maternal and Child Health and Mental Retardation Planning Amendment to the Social Security Act in 1963, which was the first legislation to increase research on intellectual disabilities and services for this population. This legislation resulted in funding for research projects focusing on etiology and prevention, as well as facilities that transcended the types of institutions that were commonplace at that time and served to provide access to treatment (John F. Kennedy Presidential Library and Museum, n.d.).

Parent Organizations

The Arc

The Arc of the United States is a community-based organization comprised of and for people with intellectual and developmental disabilities (IDD) that provides a multitude of services and support for individuals and their families across the life span (i.e., from birth, to school, to young adulthood, through their journey to employment, and even into the twilight year; Arc, 2011). The organization is dedicated to promoting and protecting the human rights of people with IDD and works to ensure that people with IDD have the services and supports they need to thrive in communities across the nation.

Prior to the 1950s, little was known about intellectual disability or developmental disabilities, and subsequently programs to support families and assist in the development and care for these individuals did not exist. During this time it was a common practice for doctors to recommend that these individuals be institutionalized

(Arc, 2018a). Displeased with this course of treatment and the lack of services, The Arc was established in 1950 through a grassroots movement led by parents and concerned community members across the country that fought for the rights of individuals with IDD to live as productive members of society rather than living their lives institutionalized (Arc, 2018d).

The organization initially aimed to alter perceptions surrounding children with IDD, educating parents and the public on the capabilities and potential these individuals possessed and could contribute to society (Arc, 2018a). The organization went on to allow access to services for those who were previously refused access to education and the right to work. The Arc is credited as the first known organization to finance research on intellectual and developmental disabilities (Arc, 2018c). As a result, The Arc was involved in better understanding the association between lead poisoning and brain damage in infants and children, advocated for legislation, and created a network of chapters including voluntary groups and professional organizations (Arc, 2018c).

In the United States, The Arc is one of the largest providers of supports and services to individuals with IDD through approximately 700 local and state chapters across the nation. Local chapters provide individual advocacy, programs, services, and supports for individuals with IDD and their families. State chapters lead state-level public policy advocacy, provide local chapter support, and some may also offer direct services (e.g., case coordination, special needs trust planning, public awareness, education). Services also include health care and free and appropriate public education, in addition to early intervention and familial supports. Vocational services aimed to allow access to employment and independent living skills are also primary goals for adults (Arc, 2011).

The Arc also implements several national programs that are funded by grants from government, private foundations, or other dedicated sources. These national programs include Autism NOW: The National Autism Resource and Information Center, Center for Future Planning™, Chapter Excellence Group, Down Syndrome Grant Program, FASD Prevention Project, Family Support Research and Training Center, HealthMeet®, Inclusive Volunteering, National Center on Criminal Justice Disability®, National Council of Self Advocates, National Sibling Council, Self-Advocates with FASD in Action (The SAFA Network), Arc@School, The Arc@Work™, Technology Programs, and Wings for Autism® (The Arc, 2018b).

The Arc is also one of the primary advocates protecting individuals with IDD and their families at the personal, local, state, and national levels. According to The Arc website, “Locally, individual advocacy is available through every local chapter of The Arc. At the state level, advocacy occurs with the executive and legislative branches of government, administrative agencies, school districts and other providers. At the national level, advocacy opportunities exist with regard to influencing federal agencies, policies and funding for disability programs and services” (Arc, 2011).

Easter Seals

Easterseals serves people and families throughout the United States who have physical emotional, intellectual, social, and educational needs (Easterseals, 2018). Easterseals works to provide individuals with disabilities an opportunity to live meaningful and productive lives by removing physical, cultural, attitudinal, and legal obstacles while also providing services and opportunities (Easterseals, 2018). In the United States, services are provided through a network of 73 local Easterseals and four international partners (Australia, Mexico, Puerto Rice, and Canada).

Following the death of his son, Ohio-businessman, Edgar Allen built a hospital in his hometown of Elyria, Ohio, out of his concern for the lack of adequate medical services available (Easterseals, 2018). Through this hospital, he discovered that it was a common practice to hide children with disabilities from public view. Following this discovery, he founded the National Society for Crippled Children in 1919 to make a difference in the way children with disabilities were typically treated. The seal was created during the spring of 1934 when the organization launched its first Easter "seals" campaign to raise money for services. A seal was created and donors showed support by placing the seal on envelopes and letters. In 1945, the organization expanded to include adults and returning WWII veterans. By 1967 the Easter "seals" campaign was so well recognized that the organization formally adopted the name "Easter Seals" (Easterseals, 2018).

Easterseals offers numerous home and community-based services and supports to individuals and families living with a disability. These resources are categorized into five distinct support areas: live (Programs: autism services, day care, in-home care, therapy, mental health), learn (Programs: accessibility, safety tips, transportation, guide for caregivers), work (Programs: employment and training, veteran reintegration, senior career employment), play (Programs: camping & recreation, respite services, brain games) and act (Programs: ways to give, advocacy, personal fundraising, Walk With Me, volunteer; Easterseals, 2018).

Condition-Specific Parent Organizations

As previously discussed, many individuals with IDDs have specific genetic or medical conditions that are related to delayed development. Although not an exhaustive list, some select organizations for parents and families of individuals with such conditions are reviewed below.

Families Affected by FASD (FAFASD)

FAFASD is an organization created by parents of individuals with FASD. This organization is designed to increase education surrounding FASD in communities and professional circles. FAFASD provides training on how to accommodate individuals with FASD, entitled "FASD and the Brain-Based Approach." These trainings are geared toward families of individuals with FASD, as well as professionals and caregivers who may have contact with individuals with FASD. The trainings are available both in person and online and are delivered by trained parents of children with FASD. Additionally, this organization aims to increase support for parents and families of individuals by providing resources and online social networking. Families within their social media (i.e., Facebook) group provide a network where families can seek advice. They also work to increase awareness surrounding FASD—in both intervention for people already diagnosed with FASD and prevention of FASDs by educating women of childbearing age (Families Affected by Fetal Alcohol Spectrum Disorder, 2018).

National Organization on Fetal Alcohol Syndrome (NOFAS)

NOFAS is a national organization established in 1990 dedicated to both prevention of FASDs and interventions for individuals already living with FASDs. Additionally, NOFAS provides support for women during pregnancy, as well as women who are birth mothers of children diagnosed with FASD. This support network operates within NOFAS and it is entitled Circle of Hope. The Circle of Hope aims to provide mentoring to women who have consumed alcohol during pregnancy which resulted in their child being diagnosed with an FASD. This network also works toward reducing the stigma surrounding mothers of children with FASDs and women who are struggling with alcohol addiction (National Organization on Fetal Alcohol Syndrome, 2018).

The NOFAS website provides an extensive database detailing resources available to families and individuals with FASD. The resources are sorted by state, as well as divided by whether the resource is for parents/caregivers or adults living with FASD. The resources available include diagnostic clinics, advocates, families, and doctors with experience treating FASDs. The website also provides extensive education surrounding FASDs and alcohol addiction, with various fact sheets about diagnosing, treating, and living with FASD. NOFAS also offers training and webinars geared toward families and medical professionals who work with individuals with FASD (National Organization on Fetal Alcohol Syndrome, 2018).

United Cerebral Palsy

United Cerebral Palsy (UCP) is a national nonprofit organization that was founded in 1949 by two families of children with cerebral palsy with the goal of improving their overall quality of life. Their website lists extensive resources geared toward helping individuals with cerebral palsy. These resources include helping families navigate health care, find affordable housing, and access education. UCP also connects families to international resources outside of the United States. Their website includes etiquette guidelines for individuals who interact with individuals with disabilities—including how to speak to people who may have difficulty hearing you and how to best interact with someone who uses a wheelchair (United Cerebral Palsy, 2018).

UCP connects individuals with disabilities and their families to the supports they need, whether that may be intervention services, therapy, assistive technology, finding a job, or accessing transportation. They also connect individuals with local affiliates who can work more closely and specifically to that individual's geographic area. These affiliates are located in the United States, Canada, and Australia. UCP also works in public policy, often advocating for legislation that increases funding and resources to improve quality of life in people with a disability. Their website offers links to contact congressional representatives directly as well (United Cerebral Palsy, 2018).

National Association for Down Syndrome

The National Association for Down Syndrome (NADS) is a nation organization founded in 1960 by the mother of a child born with Down syndrome. One of their earliest initiatives was developing their Parent Support Program. This program is designed to help parents of newborn babies with Down syndrome receive support and accurate information regarding their child's syndrome. NADS also provides education and seminars designed for health care workers and caregivers of individuals with Down syndrome. NADS works with governmental bodies to improve legislation for individuals with disability. They work on various initiatives that increase awareness and knowledge of Down syndrome and also offer resources for Spanish-speaking individuals. Although a nation organization, many of their services (e.g., support groups, welcome baskets) are only available in the greater Chicago, IL area (National Association for Down Syndrome, 2018).

National Down Syndrome Society

The National Down Syndrome Society (NDSS) is a national nonprofit organization that was founded in 1979 by parents of a child born with Down syndrome. They began with various initiatives to increase awareness and acceptance of Down syndrome, including designating October as Down Syndrome Awareness month. In 1995, NDSS organized the first Buddy Walk, which has grown to over 250 walks organized worldwide. The Buddy Walk aims to increase acceptance and inclusion of individuals with Down syndrome. In 2017, the Buddy Walk raised over $14 million that will go toward public policy and advocating initiatives for people with Down syndrome. NDSS characterizes itself as a human rights organization that works toward improving the lives of those born with Down syndrome (National Down Syndrome Society, 2018).

Much of their work is devoted to improving employment opportunities for individuals with Down syndrome. For example, their program, Even Strength, pairs individuals with job opportunities at major sporting events where they receive training. NDSS also works in public policy, advocating for legislation that helps individuals with Down syndrome more easily find employment. They have affiliates located across the country that help families contact the various supports they may need. These supports include parent support groups, trainings, interventions, and various recreational and social activities. Their website also includes numerous resources and fact sheets aimed to increase education surrounding Down syndrome (National Down Syndrome Society, 2018).

The 22q Family Foundation

The 22q Family Foundation is an organization geared toward families that aims to increase awareness of 22q11.2 deletion syndrome and provide support for families of individuals with the syndrome. This foundation provides retreats for caregivers of children with 22q deletion syndrome. Additionally, they organize various charity events to raise funding for research. Their website has an extensive database that lists various clinics, support groups, and other resources for individuals with 22q deletion syndrome and their families. Additionally, their website has resources to increase education about the syndrome. 22q Family Foundation works in various ways to connect families to resources or supports that a child may need throughout their life. Many members of their board work directly with education, providing consulting on Individualized Education Plans (IEPs) and awarding scholarships (22q Family Foundation, 2018).

The Angelman Syndrome Foundation

The Angelman Syndrome Foundation (ASF) is a national organization founded in 1992 dedicated to increasing support and funding for Angelman syndrome. They are one of the largest funders of research that examines therapeutic treatments and possible cures. ASF organizes numerous fundraising events every year to support their funding of research, including walks and marathons. They also host a yearly conference where scientists gather to present and discuss the latest research regarding Angelman syndrome. ASF's goals include improving the quality of life and the quality of care for individuals with Angelman syndrome. Their website also includes various resources that connects families with local and national resources and supports. ASF works to provide families and individuals with Angelman syndrome with education, housing, therapy services, medical services, and various recreational services. ASF has also partnered with numerous hospitals across the country to provide comprehensive care for children with Angelman syndrome (Angelman Syndrome Foundation, 2018).

Professional Organizations

American Association on Intellectual and Developmental Disabilities (AAIDD)

One of the most prominent professional organizations is AAIDD. AAIDD was founded in 1876 and was originally named the American Association on Mental Retardation (AAMR). AAIDD includes member both in the United States as well as 55 other countries and is an interdisciplinary organization that focuses on professional training, research, and supports. AAIDD publishes several research journals, specifically *American Journal on Intellectual and Developmental Disabilities*, *Intellectual and Developmental Disabilities*, and *Inclusion*, as well as measures such as the *Support Intensity Scale (SIS)* and the *Diagnostic Adaptive Behavior Scale (DABS)*. The organization also holds educational events and annual conferences, as well as provides educational resources (AAIDD, 2018).

The mission of this organization is to promote policies, research, and practice that pertain to and improve the quality of life of individuals with IDDs. Their three primary goals are to increase the abilities of professionals who work with individuals with IDD, promote societal inclusion and acceptance, and to maintain an organization that is effective at achieving these goals. AAIDD strives to improve the lives of individuals with IDD by promoting leadership and increasing skills of those working the field and promote education of students who are interested in careers in the field. Another goal is the protection of human rights (e.g., equality, dignity, respect, and choice) and support access to accommodations to increase participation in society and opportunities for choice and autonomy, as well as access to services such as education, vocational opportunities, and health care.

AAIDD further endeavors to promote inclusive attitudes and public awareness to promote inclusion on a societal level, as well develop partnerships with other organizations with the aim to further these goals (AAIDD, 2018).

American Network of Community Options and Resources (ANCOR)

ANCOR is a 501(c)3 organization and a nonprofit trade association for providers for individuals with disabilities. This organization focuses on private providers and their goals are to positively influence public policy, provide resources and development to providers, and to strengthen the community of providers to better provide integrated and long-term services. The mission of the ANCOR foundation is to "advance the ability of our members in supporting people with intellectual and developmental disabilities to fully participate in their communities" (ANCOR, 2018a). This organization also hosts annual conferences, provides trainings such as online courses and webinars, distributes publications, and provides resources for contacting ANCOR members. Resources targeting advocacy such as public policy principles and an advocacy toolkit are also included on their website (ANCOR, 2018b).

Association of University Centers on Disabilities (AUCD)

AUCD is a membership organization whose purpose is to promote interdisciplinary programs in universities and serves as a resource for local, state, national, and international organizations. Members of this organization include University Center for Excellence in Developmental Disabilities (UCEDD), Leadership Education in Neurodevelopmental Disabilities (LEND), and Intellectual and Developmental Disability Research Centers (IDDRC; AUCD, 2018). The aims of AUCD are to provide leadership, advocacy, and networking to connect these organizations on a national level. It also strives to promote communication both within this network as well as externally with other organizations (AUCD, 2018).

Intellectual and Developmental Disability Research Centers (IDDRC)

IDDRCs were founded in 1963 and were formerly referred to a Mental Retardation Developmental Disabilities Research Centers (MRDDRCs). IDDRC is a network of 14 sites funded by the Eunice Kennedy Shriver National Institute of Child Health and Human Development that serve as "center for excellence" for research on IDDs (AUCD, 2018). The sites included in the IDDRC are the Albert Einstein College of Medicine, Baylor College of Medicine, Children's Hospital of Boston, Children's Hospital of Philadelphia, Children's Research Institute/Children's National Medical Center, Hugo W. Moser Research Institute at Kennedy Krieger, University of California, Davis, University of California, Los Angeles, University of Kansas, Lawrence, University of North Carolina, Chapel Hill, University of Washington, University of Wisconsin, Madison, Vanderbilt University Medical Center, and Washington University (NIH, 2018).

The research aims of these centers include to increase understanding of factors and conditions related to IDD and to develop and implement evidence-based practices. Although centers are distinct, each supports many research projects (typically between 40 and 100) and principal investigators (20–70) per year. In addition to wide-scale research projects, sites also offer educational experiences such a pre- and post-doctoral fellowships as well as career development and training awards (AUCD, 2018).

National Alliance for Direct Support Professionals (NADSP)

NADSP is a national nonprofit organization. Their aim is to serve direct support professionals and advance practice standards and skills as well as encourage system reform. This organization emphasizes person-centered supports and stresses the importance of education and support of direct support professionals to achieve these supports. NADSP hosts annual conferences and provides access to a variety of training services, such as Front Line Supervisor (FLS) and Direct Support Professionals (DSP) certification (NADSP, 2018).

National Association of State Directions of Developmental Disabilities Services (NASDDDS)

NASDDDS represents US agencies that offers services to individuals with IDDs and their families. This organization works to provide agencies with the tools to develop support delivery systems. Principles of the organization include that individuals with DDs deserve to be treated with respect and dignity, make independent choices, participate in different areas such as family, community, and work, opportunities to achieve their full potential, and receive appropriate supports and services (NASDDDS, 2018).

The organization currently represents all 50 states in providing services to individuals with IDDs. It also sponsors conferences, projects such as National Core Indicators, State Employment Leadership Network, Community of Practice, and Evidence-Based Policy, and publications. The NASDDDS website also provides a resource library that includes resources on topics that include, but are not limited to: ADA/Olmstead, Behavioral Challenges, Case Management/Support Coordination, Closing Institutions, Employment, Housing, Managed Care, Medicaid HCBS Authorities, Preadmission Screening and Resident Review (PASRR), Person-Centered Practices, Quality, Self-Advocacy, Self-Determination/Self-Directed Services, Supporting Families, and General Information on Administering State Programs (NASDDDS, 2018).

TASH

TASH is a 501(c)(3) nonprofit organization that focuses on advocacy for individuals with disabilities. TASH was founded in 1975 under the name The American Association for the Education of the Severely and Profoundly Handicapped. Five years later in 1980 the name was changed to The Association for the Severely Handicapped, and then again in 1983 to the Association for Persons with Severe Handicaps (TASH). In 1995, it was decided by the Board of Directors that the acronym TASH would continue to be used but that the full name would be withdrawn. The primary goal is to promote participation of children and adults with disabilities and increase inclusion, in addition to decreasing inequality (TASH, 2018) (Table 9.1).

Community Resources

Special Olympics

The Special Olympics is an organization that promotes sporting events that are inclusive of children and adults with intellectual disabilities. The goals of this organization are to raise awareness about the abilities of those with IDDs, give an opportunity for the development of self-confidence, improve health, and “shift focus to what our athletes CAN do, not what they can’t” (Special Olympics, 2018). Sports included in the Special Olympics include badminton, basketball, Bocce ball, bowling, cricket, cross country skiing, cycling, equestrian, figure skating, floorball, floor hockey, football, golf, gymnastics, handball, Judo, kayaking, netball, open water swimming, powerlifting, roller skating, sailing, short track speed skating, snowboarding, snowshoeing, softball, swimming, table tennis, tennis, triathlon, and volleyball, among others (Special Olympics, 2018). Special Olympics participants include family members, professionals, and community members who participate as coaches, officials, and volunteers.

Like individuals without developmental disabilities, athletes with ID participate in sports not just for exercise but for social interaction and enjoyment (Harada & Siperstein, 2009). This is emphasized by researchers who have shown that, beyond cardiovascular health benefits (Wright &

Table 9.1 Summary of parent and professional resources

Organization	Summary of function	Link
Parent resources		
The ARC	A community-based organization that provides services for individuals with IDDs	https://www.thearc.org/
Easter Seals	A nonprofit 501(c)(3) organization that serves as a resource for individuals with disabilities and their families	http://www.easterseals.com/
Condition-specific resources		
Families affected by FASD (FAFASD)	An organization that aims to increase awareness of FASD	http://fafasd.org/
National Organization on Fetal Alcohol Syndrome (NOFAS)	An organization dedicated to the prevention of FASDs as well as effective treatment options	https://www.nofas.org/
United Cerebral Palsy	A nonprofit organization for families of those with CP to aid in improving quality of life	http://ucp.org/
National Association for Down Syndrome	An organization whose goal is to provide support and education for parents of children with Down syndrome	http://www.nads.org/
National Down Syndrome Society	A nonprofit organization that aims to increase inclusion and acceptance of individuals with Down syndrome	https://www.ndss.org/
The 22q Family Foundation	An organization that aims to increase awareness of 22q and support families	https://22qfamilyfoundation.org/
The Angelman Syndrome Foundation	AN organization whose goal is to increase funding and support for individuals with Angelman syndrome	https://www.angelman.org/
Professional resources		
American Association on Intellectual and Developmental Disabilities (AAIDD)	Organization that promotes policies, research, and practice that benefits individuals with IDDs	https://aaidd.org/
American Network of Community Options and Resources (ANCOR)	A nonprofit trade association that aids private community providers in supporting individuals with IDDs	http://www.ancor.org/
Association of University Centers on Disabilities (AUCD)	Organization that supports members that comprise a network of university programs	https://www.aucd.org/template/index.cfm
Intellectual and Developmental Disability Research Centers (IDDRC)	A network of sites that serve as "centers for excellence" for research to promote understanding of IDDs	https://www.nichd.nih.gov/about/org/der/branches/iddb
National Alliance for Direct Support Professionals (NADSP)	A national nonprofit organization that aids direct support professionals in developing skills and services	https://nadsp.org/
National Association of State Directions of Developmental Disabilities Services (NASDDDS)	Organization that provides agencies with resources to develop support delivery systems	https://www.nasddds.org/
TASH	A 501(c)(3) organization that aims to promote advocacy and inclusion of individuals with IDDs	https://tash.org/
Community resources		
Special Olympics	A sports organization for individuals with IDDs	https://www.specialolympics.org/
Best Buddies	A nonprofit 501(c)(3) organization dedicated to promoting inclusion of individuals with IDDs	https://www.bestbuddies.org/
VSA	An international organization promoting access to the arts	http://education.kennedy-center.org/education/vsa/

Cowden, 1986), the length of time individuals were involved in Special Olympics is related to improved social competence and self-concept (Dykens & Cohen, 1996; Gibbons & Byshakra, 1989; J. Wright & Cowden, 1986), as well as self-worth and physical competence (Weiss, Diamond, Demark, & Lovald, 2003).

Best Buddies

Best Buddies is a nonprofit 501(c)(3) organization dedicated to promoting inclusion of individuals with IDDs through friendship, integrated employment, and leadership development. The program was started by Anthony Shriver in 1989 at Georgetown University and currently has programs in all 50 states of the USA as well as internationally in a total of 48 countries. In 2010, the Eunice Kennedy Shriver Act was passed and authorized the use of federal funds for the program. At the time of this writing, Best Buddies has a total of 2151 chapters with 109,033 participants (Best Buddies International, 2018).

Throughout its history, Best Buddies had included eight programs: Best Buddies Citizens, Best Buddies Jobs, Best Buddies High Schools, Best Buddies Middle Schools, Best Buddies Colleges, e-Buddies, Best Buddies Ambassadors, and Best Buddies Promoters. These eight programs are divided between the three main program pillars: One-to-One friendships (Middle Schools, High Schools, Colleges, Citizens, e-Buddies), integrated employment (Jobs), and leadership development (Ambassadors and Promoters; Best Buddies International, 2018).

A study by Hardman and Clark (2006), which included participants in the college program, found that university students as well as individuals with IDDs enjoyed the experience and engaged in mutually beneficial friendships. Individuals with IDDs also reported improvements in quality of life (Hardman & Clark, 2006). These findings strengthen the anecdotal accounts of individuals and family members that the Best Buddies program increases social inclusion and participation of those with IDDs.

Very Special Arts (VSA)

VSA, the international organization on arts and disability, was founded in 1974 by Jean Kennedy Smith and was originally named the National Committee Arts for the Handicapped (VSA, 2018). It was renamed Very Special Arts in 1985 and then simply VSA in 2010. The goal of this organization is to provide opportunities to participate in arts education for individuals with IDD. The aims of the VSA are to ensure that all individuals with disabilities have access to arts education as well as facilities and activities, to prepare educators and artists to teach students with developmental disabilities, and to promote opportunities for individuals to develop skills to enable them to pursue careers in the arts. Programs encompassed in this organization include arts education, professional development, cultural access, and community engagement. Some examples of specific programs include the VSA International Art Program for Children with Disabilities, VSA Emerging Young Artists Program, VSA Playwright Discovery Award Program, and the VSA International Young Soloists Award Program (VSA, 2018).

Conclusion

The last several decades have marked substantial advancement in the field of IDD and the collaboration between this field and the general community to promote inclusion of individuals with intellectual and developmental disabilities. Researchers recommend a family-centered support approach for families of children with intellectual disabilities (Dempsey, Keen, Pennell, O'Reilly, & Neilands, 2009), and so access to these types of supports is important for family members. Parent and professional organizations have been developed to serve as intermediaries to assist individuals with IDDs in accessing appropriate supports and services that may improve their quality of life. Community organizations such as the Special Olympics and Best Buddies also target goals such as social inclusion within society. The emotional wellness of children and adults with IDDs is even now an often overlooked component of support systems, and organizations whose goals emphasize the importance of societal inclusion and social and emotional support are crucial for optimizing the potential of these individuals. The organizations reviewed in this chapter, while not meant to represent an exhaustive list of all available resources, strive to improve the lives of those with IDDs and may serve as helpful and relevant resources for those people who seek to support them.

References

AAIDD. (2018). Mission. Retrieved April 21, 2018, from http://aaidd.org/about-aaidd/mission#.WtuZB1Mvw6g

American Psychiatric Association. (2013). *Diagnostic and statistical manual of mental disorders* (5th ed.). Washington, DC: American Psychiatric Association.

ANCOR. (2018a). About ANCOR. Retrieved April 22, 2018, from http://www.ancor.org/about-ancor

ANCOR. (2018b). Advocacy. Retrieved April 22, 2018 from http://www.ancor.org/advocacy

Angelman Syndrome Foundation (2018). Retrieved from https://www.angelman.org/

AUCD. (2018). About Us. Retrieved April 22, 2018, from https://www.aucd.org/template/page.cfm?id=1

Berkowitz, E. D. (1980). The politics of mental retardation during the Kennedy administration. *Social Science Quarterly, 61*(1), 128–143.

Best Buddies International. (2018). Best Buddies International. Retrieved May 19, 2018, from https://www.bestbuddies.org/

Dempsey, I., Keen, D., Pennell, D., O'Reilly, J., & Neilands, J. (2009). Parent stress, parenting competence and family-centered support to young children with an intellectual or developmental disability. *Research in Developmental Disabilities, 30*(3), 558–566. https://doi.org/10.1016/j.ridd.2008.08.005

Dykens, E. M., & Cohen, D. J. (1996). Effects of Special Olympics International on Social Competence in Persons with Mental Retardation. *Journal of the American Academy of Child & Adolescent Psychiatry, 35*(2), 223–229. https://doi.org/10.1097/00004583-199602000-00016

Easterseals. (2018). Who We Are: The Story of the Easterseals. Retrieved from http://www.easterseals.com/who-we-are/history/

Families Affected by Fetal Alcohol Spectrum Disorder. (2018). Retrieved from http://fafasd.org/

22q Family Foundation. (2018). Retrieved from https://22qfamilyfoundation.org/

Gibbons, S. L., & Byshakra, F. B. (1989). Effects of special olympics participation on the perceived competence and social acceptance of mentally retarded children. *Adapted Physical Activity Quarterly, 6*(1), 40–51. https://doi.org/10.1123/apaq.6.1.40

Harada, C. M., & Siperstein, G. N. (2009). The sport experience of athletes with intellectual disabilities: A national survey of special olympics athletes and their families. *Adapted Physical Activity Quarterly, 26*(1), 68–85. https://doi.org/10.1123/apaq.26.1.68

Hardman, M. L., & Clark, C. (2006). Promoting friendship through best buddies: A National Survey of College Program Participants. *Mental Retardation, 44*(1), 56–63.

Harris, J. C. (2013). New terminology for mental retardation in DSM-5 and ICD-11. *Current Opinion in Psychiatry, 26*(3), 260. https://doi.org/10.1097/YCO.0b013e32835fd6fb

John F. Kennedy Presidential Library and Museum (n.d.) John F. Kennedy and People with Intellectual Disabilities. Retrieved from https://www.jfklibrary.org/JFK/JFK-in-History/JFK-and-People-with-Intellectual-Disabilities.aspx

NASDDDS. (2018). About - NASDDDS. Retrieved April 22, 2018, from http://www.nasddds.org/about-nasddds/

National Alliance for Direct Support Professionals. (2018). What we are; Who we are; Our guiding principles; Our services; Other services we offer. Retrieved from https://www.nadsp.org/

National Association for Down Syndrome. (2018). Retrieved from http://www.nads.org/

National Down Syndrome Society. (2018). Retrieved from https://www.ndss.org/

National Organization on Fetal Alcohol Syndrome. (2018). Retrieved from https://www.nofas.org/

NIH. (2018). Eunice Kennedy Shriver Intellectual & Developmental Disabilities Research Centers (EKS-IDDRCs). Retrieved from https://www.nichd.nih.gov/research/supported/eksiddrc

Special Olympics. (2018). What We Do. Retrieved April 21, 2018, from https://www.specialolympics.org/Sections/What_We_Do/What_We_Do.aspx?src=navwhat

TASH. (2018). About TASH. Retrieved from https://tash.org/about/

The Arc. (2011). Resources: Get to Know the Arc. Retrieved from https://www.thearc.org/what-we-do/resources/fact-sheets/get-to-know-the-arc

The Arc. (2018a). History of the Arc. Retrieved from https://www.thearc.org/who-we-are/history

The Arc. (2018b). Programs & Services. Retrieved from https://www.thearc.org/what-we-do/programs-and-services

The Arc. (2018c). Six Decades of Advocacy. Retrieved from https://www.thearc.org/who-we-are/history/advocacy

The Arc. (2018d). Who we are. Retrieved from https://www.thearc.org/who-we-are

United Cerebral Palsy. (2018). Retrieved from http://ucp.org/

VSA. (2018). About VSA. Retrieved from http://education.kennedy-center.org/education/vsa/

Weiss, J., Diamond, T., Demark, J., & Lovald, B. (2003). Involvement in Special Olympics and its relations to self-concept and actual competency in participants with developmental disabilities. *Research in Developmental Disabilities, 24*, 281–305.

Wright, J., & Cowden, J. E. (1986). Changes in self-concept and cardiovascular endurance of mentally retarded youths in a special olympics swim training program. *Adapted Physical Activity Quarterly, 3*(2), 177–183. https://doi.org/10.1123/apaq.3.2.177

Staff Training and Supervision

10

John Rose and Abigail Gallivan

Introduction

High-quality training and supervision for staff working with clients with challenging behavior is considered to be a vital requirement in services for people with intellectual disabilities. Training can include a range of longer-term professional courses that lead to specific qualifications in a range of professions including medicine, psychology, social work, nursing, occupational therapy, and speech and language therapy. However, this chapter will focus on the brief training that is provided mainly for direct care staff who are working in residential and community services for people with intellectual disabilities. In particular, we will focus on training relating to reducing the challenging behaviors expressed by people with intellectual disability.

Challenging behavior has been defined as "culturally abnormal behaviors of such an intensity, frequency of duration that the safety of the person or others is likely to be placed in serious jeopardy, *or* behavior which is seriously likely to limit use of, or result in the person being denied access to, ordinary community facilities" (Emerson, 1995), estimates of the percentage of people in intellectual disability services who have challenging behavior varies considerably however, Emerson et al. (2001) suggest 10–15%. Challenging behavior may be the result of mental health issues or a wide variety of other causes in people with intellectual disabilities. It is therefore important for staff to have a good knowledge of managing both challenging behavior and mental health issues in this population.

Providing effective services for those with complex needs including challenging behavior is especially demanding for care staff (Campbell, 2011) and difficulties in doing so are a contributing factor to work place stress, a major problem in services for people with intellectual disabilities in the UK (e.g., Jenkins, Rose, & Lovell, 1997; Rose, Mills, Silva, & Thompson, 2013). Staff stress can lead to poorer quality interactions with clients (Rose, Jones, & Fletcher, 1998a, 1998b), lowered job satisfaction, burnout, and higher sickness rates (Rose, 1995). Failures to provide staff with the relevant skills, knowledge and motivation in dealing with challenging behavior is costly for the wellbeing of clients and staff alike (McKenzie, Paxton, Patrick, Matheson, & Murray, 2000), yet a gap has been noted between what is known to work in reducing challenging behavior and what staff do in practice (Campbell, 2011). This indicates the need for a

J. Rose (✉)
Centre for Applied Psychology, School of Psychology, The University of Birmingham, Edgbaston, Birmingham, UK
e-mail: j.l.rose@bham.ac.uk

A. Gallivan
Forward Thinking Birmingham, Finch Road Primary Care Centre, Lozells, Birmingham, UK

J. L. Matson (ed.), *Handbook of Intellectual Disabilities*, Autism and Child Psychopathology Series, https://doi.org/10.1007/978-3-030-20843-1_10

comprehensive training approach to ensure that all staff are updated with the approaches and techniques they require to work effectively and safely.

The implementation of knowledge acquired through training requires appropriate continued support as an important element to maintain effective services. This includes a range of staff supports such as ensuring staff have the right values base, knowledge and risk management skills and a space to reflect on their work through supervision (e.g., Rose & Burns, 2011). The leadership of staff is also important to ensure that these systems are maintained and practice leadership will also be considered (e.g., Deveau & McGill, 2014).

Short Training Courses for Direct Care Staff

There is a widespread belief in the benefits of staff training for improving staff performance (Campbell, 2007), and as a result staff training has been used to educate and support staff who work with people with intellectual disabilities across a broad range of areas. For example, the literature has highlighted the benefits of training for staff in working with clients who have experienced sexual abuse (Hames, 1996), in increasing self determination (Wong & Wong, 2008), enhancing interactions with clients (Finn & Sturmey, 2009; van der Meer et al., 2017), communicating about death and dying (Tuffrey-Wijne, Rose, Grant, & Wijne, 2017), working with people who have dysphagia (Chadwick et al., 2014), working with older people (Webber, Bowers, & Bigby, 2016), and raising awareness of mental health problems among staff (Costello, Bouras, & Davis, 2007; Rose, Rose, & Kent, 2012).

Over recent years, there have been significant developments in approaches to training staff who are working with clients with intellectual disabilities who exhibit challenging behavior. Previously, training focused on the management of challenging behavior (Grey, Hastings, & McClean, 2007), particularly on behavioral and physical interventions. One criticism of this approach was that it could increase the use of aversive methods with a group of people who were unable to consent, and that techniques could be implemented without a proper understanding of the function of a person's challenging behavior (Berryman, Evans, & Kalbag, 1994). Ethical and legal concerns have also been expressed about pain-compliance methods which have been used (Allen & Tynan, 2000). Alongside these concerns it has become evident that simply attending to the process of skill acquisition of clients is often unlikely to be sufficient to change the general approach and performance of staff in the workplace (Wong & Wong, 2008).

Over recent years there has been a much stronger emphasis in the development of training approaches on working within a broader organizational and interpersonal system to support people with Intellectual Disabilities with Challenging Behavior and Mental Health issues. These are organized within a number of frameworks; in a recent systematic review Cox, Dube, and Temple (2015) characterized them as: Positive Behavioral Support, Active Support, crisis prevention and response training (with a focus on Physical interventions), and communication training. However, the variety of possible frameworks can be extended by a variety of short-term training sessions that focus particularly on developing values and attitudes.

Positive behavior support is a set of research-based strategies used to increase quality of life and decrease challenging behavior by teaching new skills and making changes in a person's environment which combines values, theory and evidence-base and process (Gore et al., 2013). Active Support embraces Positive Behavioral Support with an emphasis on a range of approaches that enable people with intellectual disabilities to take part in a broad range of activities that have meaning to them so that ultimately they can exercise more control over their lives and become more independent and live as valued members of the community. As a result Active support has a focus developing staff skills in promoting engagement and developing service capacity to provide opportunities for people with intellectual disabilities and tends to take a whole systems approach with training integrated on a regular basis for staff (Koritsas, Iacono, Hamilton, & Leighton, 2008).

Ensuring that staff have an awareness of a broader context such as advocated in the Active Support approach has implications for training staff and more recent staff training has necessarily included a more extensive range of components. These include training in established behavioral methods but also training with a stronger emphasis on values and perceptions (e.g., Rose, Gallivan, Wright, & Blake, 2014). Other staff training has developed a focus on attributions, particularly the understanding of the cognitions, the emotional understanding of staff and how these are integral to the setting conditions for staff responses to challenging behavior (Grey et al., 2007). It is useful to consider how effective a more diverse approach to training has been, especially given the breadth of training now available as a result of a broader organizational focus. Due to the increased range of factors considered to influence challenging behavior many training studies focus on a broad range of outcomes so may be referred to on a number of occasions. As a result of these complexities we have chosen a different way to consider evaluations of training as: Crisis prevention and intervention; Increasing knowledge; Changing attributions; Emotional Intelligence; Communication and values based approaches and Environmental approaches. Throughout the chapter we will select examples of illustrative studies and the impact they have had in order to orient the reader to some of the main issues.

Crisis Prevention and Response Training

A number of studies have examined the effects of training on the quality of staff physical interventions in response to challenging behavior (e.g., Allen & Tynan, 2000; Baker and Bissmire (2000); Van Oorsouw, Embregts, Bosman, & Jahoda, 2010). The argument for focusing on developing staff ability to undertake physical interventions is to reduce the likelihood of injury for clients and staff because if staff do not feel safe, they will be unlikely to implement other behavioral or communicative strategies effectively (Allen, 1999).

One example of this approach to training is provided by Van Oorsouw et al. (2010) who adopted a quasi-experimental control group design consisting of two experimental groups and two control groups to investigate the impact of a 5-day training program for staff working with clients who exhibit challenging behavior. The training took place 1 day a week for 5 weeks and consisted of 25½ h of teaching on the causes of challenging behavior, early signs of escalation, and caring for colleagues involved in incidents and 7½ h teaching on physical intervention skills. An exclusion criterion was used to ensure that none of the staff had participated in any comparable training for at least 2 years and groups were matched in terms of their professional role, the severity of challenging behavior of the people they worked with and gender. Staff in the control group received the training once the study was completed. Following the training, a significant improvement was found in the levels of knowledge of staff and in the quality of their physical interventions.

Demonstrations of physical intervention techniques were videoed and analyzed using a standardized observation manual developed by the authors. In addition, knowledge and physical intervention scores remained significantly higher at 5-month follow-up in the intervention group than pre-training scores. No significant differences were found in the control group between pre- and post-training. However, staff knowledge and physical intervention scores at follow-up in the intervention group were significantly lower than immediately after training. This suggests that maintenance of high levels of knowledge and physical interventions may require additional input and regular refresher training.

McDonnell (1997) evaluated a 3-day course which aimed to develop a number of skills including skills in defusing situations and restraint, increasing understanding of challenging behavior, and increasing the confidence of care staff. At the end of the course all participants also demonstrated competence in physical restraint skills, they also showed a significant increase in self-confidence (measured by the Managing Challenging Behavior Confidence Scale), but no significant difference in knowledge (measured by the Violence Incident Knowledge Test). These results are of interest but as with all of the evaluations in this area they need to be treated with some caution as the design of the research could have been more robust, for example, in this case, as in others, participants were not randomly allocated to conditions and the numbers involved were small (22 participants). While it is important to ensure that staff are confident and have the ability to physically manage challenging behavior, it is perhaps a more essential skill to focus on preventing challenging behavior.

Increasing Knowledge

Most of the recent approaches to reducing challenging behavior include a preventative element and an element of developing knowledge of challenging behavior. While Allen and Tynan (2000) included physical skills in their training they also had a preventative and knowledge acquisition focus. They utilized a mixed design to compare a training program in which there were two groups of staff, one who had previously received the training ($n = 51$) and another who had not ($n = 58$). Training was carried out using the "Management of Aggression Training Program." The main emphasis of this program was the development of preventative approaches to challenging behavior through antecedent or ecological change (altering aspects of the environment that may trigger challenging behavior). It consisted of an introductory theory day followed by 1–2 days of physical intervention practice according to need. Staff who had previously undertaken training were identified from attendance lists at previous training sessions and compared to an "untrained" staff group with no previous record of training. Following training, a significant improvement was found in the levels of knowledge and confidence in the previously "untrained group" (who were now trained). However, staff with experience of training in the previously "trained group" also achieved significantly higher scores on knowledge and confidence compared to the "untrained" group scores post-training. It is possible that preexisting differences found between the groups (including length of service and nature of challenging behavior experienced by the staff) may have accounted for this variation. Matching participants in the two groups in this study would have addressed this issue and further increased the validity of the findings.

McKenzie, Sharp, Paxton, and Murray (2002), McKenzie et al. (2000), Kalsy, Heath, Adams, and Oliver (2007), and Gentry, Iceton, and Milne (2001) examined the effects of brief training on enhancing staff knowledge. The basic content of training included sections on the causes of challenging behavior, signs of escalation, and preventative strategies. All of these studies assessed 1-day training apart from Gentry et al. (2001) who evaluated a 3-day program.

McKenzie et al. (2000) investigated the impact of training on the knowledge of staff relating to challenging behavior. The study consisted of a training group and a control group who did not receive the training. Knowledge was measured with a questionnaire concerning the criteria for a learning disability, defining and managing challenging behavior and duty of care. Following the training, significant increases were found in the knowledge of the trained group in relation to defining a learning disability, duty of care and defining challenging behavior but not the management of challenging behavior. The authors suggest that this may be because staff who participated in the training felt the production of behavioral guidelines and functional analysis were outside the remit of their work. Follow-up data demonstrated that their overall knowledge scores remained significantly higher at 6 and 12 months after the intervention. The control group showed no similar increase in knowledge. This study was later replicated with 36 staff with similar results (McKenzie et al., 2002).

Gentry et al. (2001) investigated a 3 day "Interactive Staff Training" (IST) on levels of staff knowledge. The IST approach was developed by Corrigan and McCracken (1997) originally for use in psychiatric settings; the training also included elements directed at awareness of organizational and motivational barriers to the implementation of new knowledge and skills. Key features of the IST approach were to train staff as a whole group (including managers) who worked together, obtaining administrative support for changes, assessing staff needs prior to training, and forming a committee responsible for decision-making regarding the organization of the training. In addition to the topics typically covered, the training also included sessions on the organizational barriers to implementing strategies and practical implications of management guidelines. Significant improvements in staff knowledge were found following training, although no follow-up was conducted in this study.

Kalsy et al. (2007) examined if a 4-h workshop could improve the knowledge of staff who worked with clients with Down syndrome, dementia and challenging behavior. The intervention consisted of teaching on the disease course of dementia, health problems, behavioral descriptors, assessment, and intervention options. Significant increases in knowledge were found following the training, though the measure related to aging and intellectual disabilities not specifically to challenging behavior. Overall, results suggest that short programs can increase knowledge; however, there are some inconsistencies as to how much and which elements of knowledge are retained.

Changing Attributions

There is an increasing recognition that addressing the way that staff understand challenging behavior can have a significant effect on how staff behave towards clients (Ager & O'May, 2001; Rose, 2011). Weiner's (1980, 1993) work provides a basis for understanding the impact of attributions, suggesting that if care staff attributed greater personal control over their actions to a client they were less sympathetic and less likely to help them than if they attributed the causes of the challenging behavior to be due to factors which were outside of the client's personal control (Rose, 2011). Research has supported the role of controllability attributions in mediating helping behavior (Dagnan, Trower, & Smith, 1998; Hill & Dagnan, 2002), suggesting that staff who hold negative perceptions are more likely to confront clients (Jahoda & Wanless, 2005), which can be unhelpful and that controllability attributions can be altered.

Williams, Dagnan, Rodgers, and McDowell (2012) reviewed the evidence for changes in carers attributions towards the behavior of people with intellectual disabilities as a consequence of training in challenging behavior. Eleven papers were reviewed that generally used a behavioral framework for staff training. While none explicitly set out to change attributions 8 of the 11 papers they reviewed reported changes in attributions. This implies that changes in attributions can occur even though these are not identified as a focus for training.

Studies have used a variety of means to assess whether staff appraisal of challenging behavior can be altered by training interventions. Berryman et al. (1994) was one of the first studies to address staff attributions of challenging behavior and move beyond a traditional operant behavioral approach. They evaluated the effects of two types of 1-day training. One group received training in traditional behavior management while a second group received training in understanding behavior in relation to a person's past experiences and social context, including teaching on functional alternatives in communication and improving quality of life. The results indicated that staff in the latter group reported a significant increase in attributions for the cause of the challenging behavior as due to external reasons such as escape-avoidance processes and tangible reinforcement and a significant reduction in the selection of categories of internal attributions such as the clients' emotions and low self-esteem as causes of challenging behavior (measured by the Causal Attributions for Challenging Behavior Scale). This change was significantly different from the group who were provided with the traditional behavioral management intervention, who tended to attribute clients' challenging behavior as a direct result of internal processes within the client such as emotions. The differences between groups were maintained at 9-month follow-up suggesting that staff who received the training focusing on client experiences continued to consider more external reasons for the cause of challenging behavior. This result suggests that staff who received the training focusing on past experiences of clients would be more likely to help and support their clients through their challenging behavior (Rose, 2011).

The study also assessed how the training influenced the development of behavioral plans by staff. Significant differences were found between the two groups, with no significant changes over time found in the intervention plans of staff in the traditional behavior management intervention whereas the group who focused on past experiences demonstrated a greater emphasis on helping clients to achieve new skills and a greater emphasis on functional analysis in their plans. These changes are likely to have a beneficial impact on client outcomes.

Significant changes in attributions after training were also found by Kalsy et al. (2007) who also investigated changes in attributions in addition to knowledge following a 4-h training session. The Controllability Beliefs Scale (Dagnan, Grant, & McDonnell, 2004) was used to assess how much control care staff believed clients with Down syndrome and dementia had over their challenging behavior. Following the training, staff reported significantly lower controllability attribution scores for their clients, suggesting that significantly less control was attributed to clients over their challenging behavior. Again, this result would suggest that staff are more likely to try and understand the reasons for the challenging behavior they are managing with and to use more constructive approaches to management.

Studies of attributional change have not always found consistent effects. Tierney, Quilan, and Hastings (2007) assessed changes in attributions, emotional reactions, and feelings of self-efficacy of staff after attending a 3-day training on understanding challenging behavior. The training included teaching on behavioral and functional assessment, using a "Positive Behavioral Support Plan," coping with stress and provided techniques from the "Non Violent Crisis Intervention Training Programme." The program centered on crisis development and appropriate interventions during and following challenging behavior. Attributions were measured by the Challenging Behavior Attributions Scale (Hastings, 1997), emotional reactions by the Emotional Reactions to Challenging Behavior Scale (Mitchell & Hastings, 1998), and self-efficacy by a 5-item Likert scale. Following training, there were no significant changes in either attributions or emotional reaction scores but a significant increase was found in staff ratings of self-efficacy. Tierney et al. (2007) had wanted to establish whether a "typical staff training approach" (i.e., fairly standard material being covered, e.g., causes of challenging behavior, functional analysis, importance of communication, precipitating factors to challenging behavior) could lead to cognitive and emotional changes in staff. Their findings suggest that a "typical" training is likely to be sufficient to improve staff feelings of self-efficacy but is insufficient to alter their cognitions or negative emotional reactions to challenging behavior and that or more targeted approach may be needed which specifically includes information on values and the specific causes of challenging behavior.

Dowey, Toogood, Hastings, and Nash (2007) investigated whether a 1-day workshop could alter staff causal explanations of challenging behavior. The workshop was presented as a pre-training for later skills based training. Lectures, vignettes, and role-play exercises were used to teach staff about the causes of challenging behavior, including the role of the environment in shaping behaviors and aspects of Applied Behavioral Analysis. The training also included a lecture on quality of life issues such as choice, respect, community presence, and participation. Changes in attributions were measured using a modified subscale of the Self-Injury Behavioral Understanding Questionnaire (Oliver, Hall, Hales, & Head, 1996). This required participants to read 11 scenarios and select from four possible causal explanations for the challenging behavior that reflected behaviorally correct, behaviorally incorrect, internal emotional or internal organic explanations. Following training, there was a significant increase in the use of behavioral explanations compared to explanations relating to the emotional or organic state of the client. However, the increase in behavioral explanations consisted of a significant increase in both behaviorally correct and behaviorally incorrect explanations. This study implies that training can change causal thinking about challenging behavior and that staff may have gained a general understanding that challenging behavior can be related to environmental and situational reasons but 1 day may not have allowed enough time for staff to develop their thinking to answer the questions correctly.

McKenzie et al. (2002) also measured attributions in addition to knowledge, but only in a small subgroup of participants ($n = 14$). Attributions were measured using two methods. The first was a bipolar scale based on four attributional dimensions suggested by Munton, Silvester, Stratton, and Hanks (1999) which were internal-external, controllable-uncontrollable, stable-unstable, and global-specific. The second was open ended questions about the causes of challenging behavior, which were scored using Bromley and Emerson's (1995) categories which included a wide range of possible causes such as internal psychological state, environment, stimulation, communication, medical, mental illness, or escape (McKenzie et al., 2002). Practice was assessed in a 4-h assessment of a series of tasks set in relation to a selected client in accordance with the Periodic Service Review (PSR, La Vigna, Willis, Shaull, Abedi, & Sweitzer, 1994). Examples of PSR tasks included the accurate recording of a client's behavior, reactive strategies and treatments selected, and the appropriate use of reinforcement. This was done immediately after the training and at 8 weeks follow-up; however, no significant changes were found in staff ratings on attributional dimensions but a significant decrease was found in the selection of the category of "communication deficit" at follow-up compared to pre-training. This suggests that some attributions may have been changing. Significant changes were also found in staff practice following training. The authors concluded that attributional change does not play a key role in changing staff practice however, the numbers of staff involved were small and the design weak as there was no comparison group.

Rose et al. (2014) provided a 1-day training which focused on developing a positive attitude and understanding challenging behavior as an alternative means of communication. This was embedded within a PBS approach where staff attributions and attitudes were measured at four time points, 1 week prior to training, immediately before and after training, and after 2 months follow-up. Following training, significant changes in staff attributions and attitudes were recorded, after the training, staff judged challenging behavior to be less under clients' personal control than they had been prior to the training. The changes in attributions were consistent with findings by Berryman et al. (1994), Kalsy et al. (2007), and Dowey et al. (2007). In accordance with Weiner's model (Weiner, 1980, 1993), these findings should lead to a corresponding increase in the helping behavior of staff towards the clients they work with. Following training, staff also demonstrated significantly more positive attitudes towards working with people with intellectual disabilities and challenging behavior and staff retained a more positive attitude at 2 months follow-up.

This training was designed as a base for educating front-line staff on the principles of PBS and resulted in a positive impact on staff attributions and attitudes.

Enabling staff to develop the right attitude in working with clients with an intellectual disability and challenging behavior was considered fundamental to this training, and as a result there was an emphasis on attitude change.

Emotional Intelligence

Emotional Intelligence has been defined as "an array of emotional, personal and social abilities and skills that influence an individual's ability to cope effectively with environmental demands and pressures" (Bar-On & Parker, 2000, p. 1108). This has led to a number of training interventions that aim to develop emotional intelligence along with improvements in interactions between staff and people with intellectual disabilities that are designed to reduce challenging behavior (e.g., Embregts, Zijlmans, Gerits, & Bosman, 2017;

Zijlmans, Embregts, Gerits, Bosman, & Derksen, 2011; Zijlmans, Embregts, Gerits, Bosman, & Derksen, 2015).

Emotional Intelligence has been suggested as a focus for training after studies have found a link between staff emotions and the challenging behavior of people they care for. Wanless and Jahoda (2002) compared different methods of examining emotional and cognitive responses of staff who frequently worked with aggressive clients. They examined responses to descriptive vignettes and real incidents of aggression previously experienced by staff and found that staff experienced more negative emotions when recalling real incidents of aggression when compared to vignettes. Staff perceptions of their aggressive clients were often linked to their cognitive and emotional responses to the aggression they had experienced previously. Mills and Rose (2011) also found that negative emotions mediate the relationship between challenging behavior and burnout in staff. It has also been suggested that adaptive success by staff in working with people who have challenging behavior is determined by their emotional intelligence (Gerits, Derksen, & Verbruggen, 2004).

Zijlmans et al. (2015) focused on the effectiveness of staff training aimed at staff emotional intelligence and staff interactions with clients. The effects of the training on emotional intelligence, coping style, and emotions of support staff were investigated. Two-hundred and fourteen support staff working in residential settings for individuals with ID and challenging behavior were split into an experimental group of 76 staff and 138 staff in two control groups. Questionnaires addressing emotional intelligence, coping, and emotions assessed effectiveness. The emotional intelligence of the experimental group changed significantly more than the two control groups. The results with regard to task oriented coping and emotions were mixed. Follow-up data suggested that the improvements within the experimental group were present 4 months after the training concluded, suggesting the intervention was effective in improving emotional intelligence of support staff.

Embregts et al. (2017) extended this work on training to evaluate the effects on emotional intelligence and support staffs' awareness of the impact on their behavior based on interactional patterns. The support provided for staff focused on the needs for autonomy, relatedness, and competence. This intervention was a pre-post test control group design ($N = 29$), with 17 support staff in the experimental group. The training program initially focused on the concept of emotional intelligence and its significance for caring and teamwork over the first 2 days. Support staff also received feedback on their scores on the Bar-On Emotional Quotient-inventory (EQ-i) (Derksen, Jeuken, & Klein Herenbrink, 1998). This approach was used to formulate goals that were translated into individual developmental plans such as aiming to work in a more structured way or to improve an ability to understand the emotional needs of specific clients. Support staff were asked to work on plans individually with feedback and support from the trainers. Support staff were asked to make video recordings of themselves interacting with clients. These videos were reviewed by support staff and trainers together and linked to the training protocols which aimed to increase desirable behaviors related to emotional intelligence and their chosen specific goals. Trainers followed specific protocols for the description of responses such as contingent praise, and corrective comments to ensure consistency across sessions. On completion of training support staff received feedback on their new EI profiles, based on a newly administered EQ-i. Staff were also asked to make new video recordings of them interacting with their clients for a further evaluation. For both groups of staff video recordings of interactions with residents were analyzed. Interactional patterns in the videos suggested that the training had had a positive impact on the support that staff gave to residents in developing their autonomy, relatedness, and competence. These results suggest that training with a focus on emotional intelligence can improve staff interactions with residents and results are similar to more general stress management training with staff which has been shown to both reduce stress in staff and increase the amount of assistance staff provide to residents (Gardner, Rose, Tyler, & Cushway, 2005; Rose et al., 1998a). However, whether these results are achieved by increasing emotional intelligence or through some other means needs further scrutiny as other research suggests that emotional intelligence is not clearly linked to staff outcomes such as burnout and stress (Shead, Scott, & Rose, 2016).

Communication and Values Based Approaches

Some programs have targeted different elements of staff behavior with a view to change attributions of staff. Smidt, Balandin, Reed, and Sigafoos (2007) found that after the implementation of four, two and a half hour training sessions using a "MOSIAC" package (Model of Interaction for the Analysis of Interaction and Communication), there were some initial increases in the use of augmentative communication skills by staff (n = 18) and some small changes in attributions (measured by the CHABA). However, this was only maintained at follow-up (6 month and 12 month) by one organization out of three who participated. There was also little impact on challenging behavior recorded by staff which was ascertained through an audit of clients' incident forms.

The brief training described by Rose et al. (2014) which had a focus on attributional change also had an emphasis on encouraging staff to think about how they themselves would feel and respond in a situation where they were unable to communicate with a view to helping staff understand why challenging behavior might occur. This approach may have contributed to a change in attitudes and attributions by increasing staffs' understanding and empathy towards people with intellectual disabilities and challenging behavior.

Environmental Approaches

An alternative focus for staff training is provided by nidotherapy which attempts to reduce challenging behavior by changing the living environment to create a better fit between the person, their environment and society and in so doing reduce the frequency of challenging behaviors expressed by clients. Nidotherapy attempts to treat the problems of challenging behavior not by treating the behavior directly, but by changing the environment to create a better fit between the person and their environment (Tyrer, Sensky, & Mitchard, 2003). Nidotherapy has been applied to different chronic mental health problems (Tyrer, Kramo, Milošeska, & Seivewright, 2007), often when other interventions have failed. In a recent study (Tyrer et al., 2017), nidotherapy was introduced to staff by providing a series of structured staff training sessions to all staff involved in the direct care of clients over a period of 6 months. The four components of training include: (1) nidotherapy—person-environment understanding; (2) environmental analysis; (3) creation of a new environmental pathway (nidopathway); and (4) monitoring of the pathway. These training sessions were delivered to staff over a 6-month period. These training sessions are delivered with discussion in relation to actual episodes of challenging behavior that occur within the environment. In a randomized control trial staff in twenty homes either received training in nidotherapy or the enhanced care program approach (ECPA), the latter approach involved structured assessment of the person with challenging behavior, the development of individual goals, and monitoring of those goals. The duration of training provided was equivalent within each arm of the trial. A number of measures of challenging behavior were used to record incidents over the course of the trial. A total of 200 residents entered the trial, 115 allocated to the ECPA arm and 85 to the nidotherapy. No statistically significant reductions in challenging behavior were demonstrated over the trial in either group, but in the last 7 months, those allocated to nidotherapy had a 33% reduction in Modified Overt Aggression Scale scores and a 43% reduction in Problem Behavior Check List scores compared with much smaller reductions for the ECPA group. The changes on the MOAS were close to statistical significance over the last 7 months of the trial. This suggests that a nidotherapy approach may be worth exploring further as the delay in possible effect seems consistent with an intervention provided through staff in relation to environmental change where a delay in implementation and effect would seem likely.

Methodological Issues

The Tyrer et al. (2017) study of nidotherapy was one of the most methodologically robust of those reported as it was a cluster randomized control trial with blinding between the two intervention groups; however, it was the exception and there were a range of methodological weaknesses identified with many of the other studies reported here. In general, there was a lack of clear reporting with regard to the populations from which the samples were recruited, the numbers of staff prepared to participate compared to the numbers of staff invited to training or the numbers of participants lost to follow-up. In many studies there was also an absence of blinding procedures, some studies did not collect follow-up data and there was no consideration of factors that could confound the results, e.g., whether staff had recently attended other relevant behavior training. Another internal validity issue frequently identified was the accuracy of the main outcome measures used. For example, the measure used in Gentry et al.'s (2001) study that detected highly significant increases in staff knowledge scores was an "ad hoc" measure with no detail about its structure, development, reliability, or validity. Psychometric properties are also not reported for the 20-item knowledge quiz used in the study by Kalsy et al. (2007). McKenzie et al. (2002) employed a self-assessment visual analogue scale on which staff were required to rate how much they believed their knowledge of challenging behavior had changed following training. Therefore, subjective perceptions about improvements in knowledge, not actual changes, were obtained.

While there appeared to be a greater use of standardized tools for the measurement of attributions, there are still some issues. Tierney et al. (2007) found no changes in staff attributions following a 3-day workshop. They used the CHABA (Hastings, 1997) to measure attributions and the authors themselves highlight the low levels of internal consistency of the CHABA on several sub scales which may lead to these results being questionned. Another issue was that post-training scores were only gathered after 3 months and so their conclusion that training did not significantly change staff attributions may be inaccurate, as a better design incorporating both immediate and follow-up data collection may have shown that changes in attribution scores occurred but were not maintained at follow-up. McDonnell (1997) found that a large number of staff attributed their increase in confidence to the role-play exercises but there are also concerns about the generalizability of changes in staff attributions translating to a positive impact on working directly with clients.

The Impact of Short Staff Training

Despite concerns about the methodology of many of the studies, the evidence suggests that brief training has a role to play in increasing staff knowledge, for example, knowledge on intellectual disabilities, challenging behavior, and proactive and reactive strategies. The effectiveness of training of staff is consistent, at least with regard to immediate impact. The evidence also suggests that training can change staff attributions relating to challenging behavior in that following training, staff generally give more consideration to reasons external to the person (i.e., environmental or situational) as causes of challenging behavior with less focus on internal state. The literature also implies that training can improve staff confidence, emotional intelligence, the quality of physical intervention techniques and some other aspects of staff practice such as the development and implementation of behavioral guidelines. Increasing staff knowledge may alter the way staff approach clients, which could reduce the chances of incidents occurring (Van Oorsouw et al., 2010). It is recognized that in care services there are often poor levels of knowledge but all staff require the necessary knowledge to work with people with challenging behavior (Ball, Bush, & Emerson, 2004). As a result it seems important that all staff should have access to at least basic training to help them gain an appreciation of challenging behavior.

There is some indication from the studies discussed here that a "typical training approach" of a single day is sufficient to change levels of knowledge but may be insufficient to change staff attributions or emotional reactions, even where training is 3 days long rather than a single day. A more targeted approach which addresses values and beliefs of staff may be required for changes in these variables. The evidence for changing staff behavior is even weaker; however, some studies are starting to encompass measures of behavior change indicating some positive changes are possible (e.g., Embregts et al., 2017).

The Maintenance of the Impact of Training

The evidence suggests that there is a role for staff training and that it is important to train staff so that they can support individuals with intellectual disability and challenging behavior effectively. This leads to the implication that services need to ensure that all staff can become trained, understand what they are told and are able to apply what they have learnt consistently and over time. At present, much training is not mandatory particularly in relation to challenging behavior and there is a need for services to address training needs.

It would be beneficial for any organizational strategy to include a system which ensured that staff are regularly updated and those who require further training are identified. In the study by Berryman et al. (1994) changes in attributions were maintained at 9-month follow-up and the authors describe the use of biweekly supervision as a methodology for reinforcing the training intervention. This also implies that the role of supervision as part of an overall strategy to maintain improved staff performance may be important. It has been recognized that there is variation in what staff and employers deem to be the remit and responsibilities of their jobs (Campbell, 2011). For a multifaceted strategy to work, value must be placed on staff development by all involved. Recognizing the impact of organizational barriers on new learning, Gentry et al. (2001) and Dowey et al. (2007) used approaches that addressed aspects of organizational culture. Individualizing training for a team may improve the likelihood of training having an impact, by increasing the contextual fit between the taught ideas and their acceptability (Grey et al., 2007).

Rose and Burns (2011) and Rose, Harris, and Burns (2010) suggested a framework for staff support which provides a range of potential areas to support staff and prevent staff burnout. This framework includes staff support interventions that can be introduced proactively such as supervision and risk assessment but also reactive strategies such as appropriate physical skills training and responsive elements such as incident analysis and behavioral skills training. A modified version of this framework is provided in Fig. 10.1 which reframes many elements which were elements of a support framework in terms of training interventions and ongoing management functions that are likely to ensure that the benefits of training are maintained and applied effectively on a regular basis. This revised framework suggests a range of discrete areas of training that can be provided to staff over time dependent upon their needs and a range of organizational methodologies that can be used to maintain these skills. Some of these areas could be seen as universally applied to all staff such as ensuring that they are able to demonstrate appropriate attitudes and values. For example, all staff are likely to be involved with incidents of challenging behavior as a result they need to be able to record incidents accurately and in a way that can be understood by others. However, staff may not be required to formulate the reason for the challenging behavior as other staff such as psychologists or behavioral specialists may be available to support them to understand what is going on and develop collaborative formulations. Supervision and regular monitoring of performance by management would be an essential part of the development and monitoring

Proactive Training	**Reactive Training**	**Responsive Training**
• Person centered values and attitudes training • Increasing knowledge • Training on the communicative aspects of challenging behavior • Training on attributions of challenging behavior • Training on emotional intelligence • Training on Risk assessment/risk management culture	• Appropriate physical skills training	• Nidotherapy • Behavioral skills training. • Incident analysis • Formulation
Proactive Support	**Reactive Support**	**Responsive Support**
• Reflective practice groups • Supervision • Stress management processes	• Post incident support	• Individual support (formal and informal) • Psychological support

Fig. 10.1 A training and support framework (Adapted from Rose & Burns, 2011)

of staff performance to ensure that staff are interpreting and implementing their training effectively.

Models exist for the implementation of training focused framework described here, particularly, Positive Behavioral Support and Active Support; however, by reviewing the individual elements of training it may be possible to consider which elements are required in any particular environment and as a result target training interventions more precisely to the needs of the people with intellectual disability, thus focusing effort and resources more effectively.

The development of focus in training interventions has synergy with the idea of practice leadership in intellectual disability settings. In a qualitative study, interviewing managers of staffed group homes, Deveau and McGill (2014) identified monitoring staff performance, supporting new ways of working, shaping staff performance, influence of external agencies, and importance of participants' personal values and experiences as a set of themes for defining practice leadership. These themes resonate with the suggestions for implementing staff training interventions and further work by Deveau and McGill (2016) and Deveau (2016) suggests that effective practice leadership is associated with greater job satisfaction and positive experiences for staff.

Conclusions

Training for direct care staff is one important element of providing effective support to people with intellectual disabilities with challenging behavior. The evidence base suggests that short training courses can be effective; however, there is considerable variation in the results of evaluations and that a number of factors need to be considered to ensure training success and particularly in relation to any gains made by training. The evidence for any change in the behavior of staff and people with intellectual disabilities as a result of training also remains relatively poor with few studies examining these outcomes. However, it is clear that to provide effective and appropriate training to staff in direct care roles who work with people with challenging behavior requires considerable thought and organization which needs to be maintained in relation to the changing needs of the people with intellectual disabilities that they serve.

References

Ager, A., & O'May, F. (2001). Issues in the definition and implementation of 'best practice' for staff delivery of interventions for challenging behavior. *Journal of Intellectual & Developmental Disability, 26*, 243–256.

Allen, D. (1999). Success and failure in community placements for people with intellectual disabilities and challenging behaviour: An analysis of key variables. *Journal of Mental Health, 8*, 307–320.

Allen, D., & Tynan, H. (2000). Responding to aggressive behavior: Impact of training on staff members' knowledge and confidence. *Mental Retardation., 38*, 97–104.

Baker, P. A., & Bissmire, D. (2000). A pilot study of the use of physical intervention in the crisis management of people with intellectual disabilities who present challenging behaviour. *Journal of Applied Research in Intellectual Disabilities, 13*(1), 38–45.

Ball, T., Bush, A., & Emerson, E. (2004). *Psychological interventions for severely challenging behaviors shown by people with learning disabilities: Clinical Practice Guidelines*. Leicester: British Psychological Society.

Bar-On, R. E., & Parker, J. D. (2000). *The handbook of emotional intelligence: Theory, development, assessment, and application at home, school, and in the workplace*. San Francisco, CA: Jossey-Bass.

Berryman, J., Evans, I. M., & Kalbag, A. (1994). The effects of training in non-aversive behavior management on the attitudes and understanding of direct care staff. *Journal of Behavior Therapy and Experimental Psychiatry, 25*, 241–250.

Bromley, J., & Emerson, E. (1995). Beliefs and emotional reactions of care staff working with people with challenging behavior. *Journal of Intellectual Disability Research, 39*, 341–352.

Campbell, M. (2007). Staff training and challenging behavior – Who needs it? *Journal of Intellectual Disabilities, 11*, 143–156.

Campbell, M. (2011). Workforce development and challenging behavior: Training staff to treat, to manage or cope. *Journal of Intellectual Disabilities, 14*, 185–196.

Chadwick, D. D., Stubbs, J., Fovargue, S., Anderson, D., Stacey, G., & Tye, S. (2014). Training support staff to modify fluids to appropriate safe consistencies for adults with intellectual disabilities and dysphagia: An efficacy study. *Journal of Intellectual Disability Research, 58*(1), 84–98.

Corrigan, P. W., & McCracken, S. G. (1997). *Interactive staff training*. New York: Plenum Press.

Costello, H., Bouras, N., & Davis, H. (2007). The role of training in improving community care staff awareness of mental health problems in people with intellectual disabilities. *Journal of Applied Research in Intellectual Disabilities, 20*, 228–235.

Cox, A. D., Dube, C., & Temple, B. (2015). The influence of staff training on challenging behavior in individuals with intellectual disability: A review. *Journal of Intellectual Disabilities, 19*(1), 69–82.

Dagnan, D., Grant, F., & McDonnell, A. (2004). Understanding challenging behaviour in older people; the development of the controllability beliefs scale. *Behavioural and Cognitive Psychotherapy, 32*(4), 501–506.

Dagnan, D., Trower, P., & Smith, R. (1998). Care staff responses to people with a learning disability and challenging behavior: A cognitive emotional analysis. *British Journal of Clinical Psychology, 37*, 59–68.

Derksen, J. J. L., Jeuken, J., & Klein Herenbrink, A. J. M. (1998). *Bar-On Emotioneel Quotiënt Vra-genlijst, Nederlandse vertaling en bewerking*. Nimwegen: PEN Tests Publisher.

Deveau, R. (2016). Commentary on "can active support improve job satisfaction?". *Tizard Learning Disability Review, 21*(2), 61–64.

Deveau, R., & McGill, P. (2014). Leadership at the front line: Impact of practice leadership management style on staff experience in services for people with intellectual disability and challenging behavior. *Journal of Intellectual and Developmental Disability, 39*(1), 65–72.

Deveau, R., & McGill, P. (2016). Practice leadership at the front line in supporting people with intellectual disabilities and challenging behaviour: A qualitative study of registered managers of community-based, staffed group homes. *Journal of Applied Research in Intellectual Disabilities, 29*(3), 266–277.

Dowey, A., Toogood, S., Hastings, R. S., & Nash, S. (2007). Can brief workshop interventions change care staff understanding of challenging behavior. *Journal of Applied Research in Intellectual Disabilities, 20*, 52–57.

Embregts, P. J., Zijlmans, L. J., Gerits, L., & Bosman, A. M. (2017). Evaluating a staff training program on the interaction between staff and people with intellectual disability and challenging behavior: An observational study. *Journal of Intellectual & Developmental Disability, 44*(2), 131–138.

Emerson, E. (1995). *Challenging behavior: Analysis and intervention with people with learning difficulties*. Cambridge: Cambridge University Press.

Emerson, E., Kiernan, C., Alborz, A., Reeves, D., Mason, H., Swarbrick, R., … Hatton, C. (2001). The prevalence of challenging behavior: A total population study. *Research in Developmental Disabilities, 22*, 77–93.

Finn, L. L., & Sturmey, P. (2009). The effect of peer-to-peer training on staff interactions with adults with dual diagnoses. *Research in Developmental Disabilities, 30*, 96–106.

Gardner, B., Rose, J. M., Tyler, P., & Cushway, D. (2005). Cognition and coping in work-related stress: An intervention study. *Work and Stress, 19*(2), 137–152.

Gentry, M., Iceton, J., & Milne, D. (2001). Managing challenging behavior in the community: Methods and results of interactive staff training. *Health and Social Care in the Community, 9*, 143–150.

Gerits, L., Derksen, J. J., & Verbruggen, A. B. (2004). Emotional intelligence and adaptive success of nurses caring for people with mental retardation and severe behavior problems. *Mental Retardation, 42*(2), 106–121.

Gore, N. J., McGill, P., Toogood, S., Allen, D., Hughes, J. C., Baker, P., … Denne, L. D. (2013). Definition and scope for positive behavioural support. *International Journal of Positive Behavioural Support, 3*(2), 14–23.

Grey, I. M., Hastings, R. S., & McClean, B. (2007). Editorial-staff training and challenging behavior. *Journal of Applied Research in Intellectual Disabilities, 20*, 1–5.

Hames, A. (1996). The effects of experience and sexual abuse training on the attitudes of learning disability staff. *Journal of Intellectual Disability Research, 40*, 544–549.

Hastings, R. P. (1997). Measuring staff perceptions of challenging behavior: The challenging behavior attributions scale. *Journal of Intellectual Disability Research, 41*, 495–501.

Hill, C., & Dagnan, D. (2002). Helping, attributions, emotions and coping style in response to people with learning disabilities and challenging behavior. *Journal of Learning Disabilities, 6*, 363–372.

Jahoda, A., & Wanless, L. K. (2005). Knowing you: The interpersonal perceptions of staff towards aggressive individuals with mild to moderate intellectual disabilities in situations of conflict. *Journal of Intellectual Disability Research, 49*, 544–551.

Jenkins, R., Rose, J., & Lovell, C. (1997). Psychological well-being of staff working with people who have challenging behaviour. *Journal of Intellectual Disability Research, 41*(6), 502–511.

Kalsy, S., Heath, R., Adams, D., & Oliver, C. (2007). Effects of training on controllability attributions of behavioral excesses and deficits shown by adults with Down syndrome and dementia. *Journal of Applied Research in Intellectual Disabilities, 20*, 64–68.

Koritsas, S., Iacono, T., Hamilton, D., & Leighton, D. (2008). The effect of active support training on engagement, opportunities for choice, challenging behaviour and support needs. *Journal of Intellectual and Developmental Disability, 33*(3), 247–256.

La Vigna, G. W., Willis, T. J., Shaull, J. F., Abedi, M., & Sweitzer, M. (1994). *The periodic service review: A total quality assurance system for human services and education*. Baltimore, MD: Brookes.

McDonnell, A. (1997). Training care staff to manage challenging behavior: An evaluation of a three day training course. *The British Journal of Developmental Disabilities, 43*, 156–162.

McKenzie, K., Paxton, D., Patrick, S., Matheson, E., & Murray, G. C. (2000). An evaluation of the impact of a one-day challenging behaviour course on the knowledge of health and social care staff working in learning disability services. *Journal of Learning Disabilities, 4*(2), 153–165.

McKenzie, K., Sharp, K., Paxton, D., & Murray, G. C. (2002). The impact of training and staff attributions of staff practice in learning disability service. A pilot study. *Journal of Intellectual Disabilities, 6*, 239–251.

Mills, S., & Rose, J. (2011). The relationship between challenging behavior, burnout and cognitive variables in staff working with people who have intellectual disabilities. *Journal of Intellectual Disability Research, 55*(9), 844–858.

Mitchell, G., & Hastings, R. (1998). Learning disability care staff's emotional reactions to aggressive behaviors: Development of a measurement tool. *British Journal of Clinical Psychology, 37*, 441–449.

Munton, A. G., Silvester, J., Stratton, P., & Hanks, H. (1999). *Attributions in action: A practical approach to coding qualitative data*. Chichester: Wiley.

Oliver, C., Hall, S., Hales, J., & Head, D. (1996). Self-injurious behavior and people with intellectual disabilities: Assessing the behavioral knowledge and causal explanations of care staff. *Journal of Applied Research in Intellectual Disabilities, 9*, 229–239.

Rose, J. (1995). Stress and staff in residential settings: Towards an integration of existing research. *Mental Handicap Research, 8*, 220–236.

Rose, J. (2011). How do staff psychological factors influence outcomes for people with developmental and intellectual disabilities in residential services? *Current Opinion in Psychiatry, 24*(5), 403–407.

Rose, J. And Burns, M. (2011) Supporting yourself and your team. *In Challenging behavior: A reader.* Brighton *Pavillion*. Stephen Hardy and Theresa Joyce.

Rose, J., Gallivan, A., Wright, D., & Blake, J. (2014). Staff training using positive behavioral support: The effects of a one-day training on the attributions and attitudes of care staff who work with people with an intellectual disability and challenging behavior. *International Journal of Developmental Disabilities, 60*(1), 35–42.

Rose, J., Harris, P., & Burns, M. (2010). Supporting staff. In S. Hardy & T. Joyce (Eds.), *Challenging behaviour: A training pack*. Brighton: *Pavillion*.

Rose, J., Jones, F., & Fletcher, B. (1998a). Investigating the relationship between stress and worker behavior. *Journal of Intellectual Disability Research, 42*, 163–172.

Rose, J., Jones, F., & Fletcher, B. C. (1998b). The impact of a stress management programme on staff well-being and performance at work. *Work & Stress, 12*(2), 112–124.

Rose, J., Mills, S., Silva, D., & Thompson, L. (2013). Testing a model to understand the link between client characteristics, organisational variables, staff cognitions and burnout in care staff. *Research in Developmental Disabilities, 34*(3), 940–947.

Rose, N., Rose, J., & Kent, S. (2012). Staff training in intellectual disability services: A review of the literature and implications for mental health services provided to individuals with intellectual disability. *International Journal of Developmental Disabilities, 58*(1), 24–39.

Shead, J., Scott, H., & Rose, J. (2016). Investigating predictors and moderators of burnout in staff working in services for people with intellectual disabilities: The role of emotional intelligence, exposure to violence, and self-efficacy. *International Journal of Developmental Disabilities, 62*(4), 224–233.

Smidt, A., Balandin, S., Reed, V., & Sigafoos, J. (2007). A communication training programme for residential staff working with adults with challenging behavior: Pilot data on intervention effects. *Journal of Applied Research in Intellectual Disabilities, 20*, 16–29.

Tierney, E., Quilan, D., & Hastings, R. P. (2007). Impact of a 3-day training course on challenging behavior on staff cognitive and emotional responses. *Journal of Applied Research in Intellectual Disabilities, 20*, 58–63.

Tuffrey-Wijne, I., Rose, T., Grant, R., & Wijne, A. (2017). Communicating about death and dying: Developing training for staff working in services for people with intellectual disabilities. *Journal of Applied Research in Intellectual Disabilities, 30*(6), 1099–1110.

Tyrer, P., Kramo, K., Milošeska, K., & Seivewright, H. (2007). The place for nidotherapy in psychiatric practice. *Psychiatric Bulletin, 31*, 1–3.

Tyrer, P., Sensky, T., & Mitchard, S. (2003). Principles of nidotherapy in the treatment of persistent mental and personality disorders. *Psychotherapy and Psychosomatics, 72*(6), 350–356.

Tyrer, P., Tarabi, S. A., Bassett, P., Liedtka, N., Hall, R., Nagar, J., … Tyrer, H. (2017). Nidotherapy compared with enhanced care programme approach training for adults with aggressive challenging behaviour and intellectual disability (NIDABID): Cluster-randomised controlled trial. *Journal of Intellectual Disability Research, 61*(6), 521–531.

van der Meer, L., Waddington, H., Sigafoos, J., Balandin, S., Bravo, A., Ogilvie, E., … Sawchak, A. (2017). Training direct-care staff to implement an iPad®-based communication intervention with adults with developmental disability. *International Journal of Developmental Disabilities, 63*(4), 246–255.

Van Oorsouw, W. M. W. J., Embregts, P. J. C. M., Bosman, A. M. T., & Jahoda, A. (2010). Training staff to manage challenging behavior. *Journal of Applied Research in Intellectual Disabilities, 23*, 192–196.

Wanless, L. K., & Jahoda, A. (2002). Responses of staff towards people with mild to moderate intellectual disability who behave aggressively: A cognitive emotional analysis. *Journal of Intellectual Disability Research, 46*(6), 507–516.

Webber, R., Bowers, B., & Bigby, C. (2016). Confidence of group home staff in supporting the health needs of older residents with intellectual disability. *Journal of Intellectual and Developmental Disability, 41*(2), 107–114.

Weiner, B. (1980). A cognitive (attribution)-emotion-action model of helping behavior: *An analysis of judgement of help giving*. *Journal of Personality and Social Psychology, 39*, 1142–1162.

Weiner, B. (1993). On sin versus sickness: A theory of perceived responsibility and social motivation. *American Psychologist, 48*, 957–965.

Williams, S., Dagnan, D., Rodgers, J., & McDowell, K. (2012). Changes in attributions as a consequence of training for challenging and complex behavior for carers of people with learning disabilities: A systematic review. *Journal of Applied Research in Intellectual Disabilities, 25*, 203–216. https://doi.org/10.1111/j.1468-3148.2011.00654.x

Wong, P. K. S., & Wong, D. F. K. (2008). Enhancing staff attitudes, knowledge and skills in supporting the self-determination of adults with intellectual disability in residential settings in Hong Kong: A pretest-posttest comparison group design. *Journal of Intellectual Disability Research, 52*, 230–243.

Zijlmans, L. J. M., Embregts, P. J. C. M., Gerits, L., Bosman, A. M. T., & Derksen, J. J. L. (2011). Training emotional intelligence related to treatment skills of staff working with clients with intellectual disabilities and challenging behavior. *Journal of Intellectual Disability Research, 55*(2), 219–230.

Zijlmans, L. J. M., Embregts, P. J. C. M., Gerits, L., Bosman, A. M. T., & Derksen, J. J. L. (2015). The effectiveness of staff training focused on increasing emotional intelligence and improving interaction between support staff and clients. *Journal of Intellectual Disability Research, 59*(7), 599–612.

11

Multidisciplinary Teams

Patricia M. Noonan and Amy S. Gaumer Erickson

The complex nature of intellectual disabilities means that multiple professionals must work together to promote quality transition services and successful post-school outcomes. The composition of these teams may vary based on the complexity of a student's disability or disabilities, the student's age, and the context in which services are provided. Schools, community agencies, healthcare providers, and nonprofits can work together to provide quality supports and services for individuals, but there are innate challenges. This chapter explores how to achieve productive collaboration by incorporating specific strategies and developing necessary capacities for promoting and facilitating adult professional teaming.

Overview

Young adults with intellectual disabilities too often experience low post-school outcomes such as: (a) poor graduation rates from high school, (b) low employment rates after high school, and (c) low participation in post-secondary education. These outcomes often lead to lower quality of life and an increasing number of youths receiving Social Security or other welfare program benefits or being incarcerated (Johnson, Thurlow, & Stout, 2007; National Center on Secondary Education and Transition, 2004; Newman, Wagner, Cameto, Knokey, & Shaver, 2010; Taylor, Krane, & Orkis, 2010). One critical strategy for improving these outcomes is increased collaboration and teaming between professionals to facilitate seamless supports and services tailored to each individual.

For over two decades, the field has repeatedly acknowledged that interagency collaboration plays a critical role in positive post-school outcomes (Carter et al., 2009; Kohler & Field, 2003; Morningstar, Kleinhammer-Tramill, & Lattin, 1999; Noonan, Morningstar, & Gaumer Erickson, 2008; Noonan, Morningstar, & Lattin, 2008; Test et al., 2009). These critical linkages are significant factors in promoting higher employment and post-secondary educational outcomes, especially for students with intellectual disabilities (Bullis, Davis, Bull, & Johnson, 1995; Hasazi, Furney, & DeStefano, 1999; Rabren, Dunn, & Chambers, 2002; Repetto, Webb, Garvan, & Washington, 2002).

While collaboration between agencies supporting adults with disabilities is important, it is vital to establish linkages between agencies, the K-12 education system, and other relevant agencies early on, while students are receiving K-12 services and supports. Interagency collaboration

---

P. M. Noonan (✉) · A. S. Gaumer Erickson
Research Collaboration, University of Kansas,
Lawrence, KS, USA
e-mail: pnoonan@ku.edu; agaumer@ku.edu;
http://www.researchcollaboration.org;
http://www.researchcollaboration.org

J. L. Matson (ed.), *Handbook of Intellectual Disabilities*, Autism and Child Psychopathology
Series, https://doi.org/10.1007/978-3-030-20843-1_11

is critical to transition planning and related supports in middle and high school; additionally, it facilitates better linkages to adult agencies and higher outcomes for young adults with disabilities (Hasazi et al., 1999; Kohler & Field, 2003; Rabren et al., 2002; Repetto et al., 2002). In other words, for optimal success, we must start early in planning and providing important transition services, and educators should not be working in isolation from adult agencies who may later be providing critical supports.

In addition to promoting better post-school outcomes, much can be gained from increased collaboration between entities such as K-12 education, vocational rehabilitation, workforce investment, employment agencies, developmental disability organizations, social services, chambers of commerce, nonprofits, and others. Collaboration can lead to less duplication of resources, improved professional development, and a cross-flow of information (Blalock, 1996; Wehman, 1998). As we partner to meet the needs of young adults with disabilities in-school and post-school, we often learn that there are gaps in resources and supports which can be filled if understood and targeted. We also learn about other agency services, challenges, and overall goals, and piece together more meaningful supports specific to each individual.

While there is clearly much to be gained, working with other professionals to coordinate services and supports can be challenging. There are numerous barriers to interagency collaboration, including: (a) poor/inaccurate perceptions of outside agencies by school staff, students, and parents (and vice versa) and (b) nonexistent/ineffective procedures for collaboration between school and agency staff throughout the referral, eligibility, determination, and transition planning process (Agran, Cain, & Cavin, 2002; Benz, Johnson, Mikkelsen, & Lindstrom, 1995; Li, 2004). Additional research (Certo, Pumpian, Fisher, Storey, & Smalley, 1997) noted that there are other limitations inherent to the public school systems (i.e., locus of service delivery and staffing patterns) and the adult service sector (i.e., discrepancies between entitlement vs. eligibility and differing supports and services).

More specifically, barriers to interagency collaboration include: (a) lack of shared information on students across agencies; (b) lack of follow-up data on program recipients that could be used to improve transition services; (c) lack of attention to IEP post-secondary goals; (d) lack of systematic transition planning with adult agencies; (e) ineffectual interagency agreements; and (f) difficulties in projecting post-secondary needs and services (Johnson, Stodden, Emanuel, Luecking, & Mack, 2002; Noyes & Sax, 2004). In one study that examined collaboration among adult service providers, the most challenging barriers to overcome were the difficulty in merging professional culture differences and the loss of professional identities among agencies (Timmons, Cohen, & Fesko, 2004). In a review of multiple projects providing mental health services to youths with emotional disabilities, some of the challenges identified were blame and distrust among child-serving agencies, inflexibility, and fear of change (Hodges, Nesman, & Hernandez, 1999). Furthermore, the report concluded that to be effective, collaboration must occur at multiple administrative levels within an agency and across multiple agencies.

To summarize, early research on collaboration and transition succeeded in identifying the critical nature of building the relationships and linkages, as well as identifying barriers to effective professional collaboration in complex systems. Providing quality transition supports in middle and high schools to support positive post-school outcomes requires involvement and commitment from multiple agencies, the family, and the student, throughout both planning and implementation (Kleinhammer-Tramill, Rosenkoetter, & Tramill, 1994; Noyes & Sax, 2004). The ultimate goal of collaboration is to ensure a seamless transition from one agency to the next, thereby providing needed services with minimum disruptions (Halpern, 1994). To accomplish this, educators and agency professionals must successfully engage in collaboration.

Strategies for Collaboration

Interagency collaboration by nature is dynamic; it develops slowly over time as a group as well as between individual members representing agencies. Agencies, such as businesses, healthcare providers, social services, and educators begin to take specific steps toward productive partnerships (Johnson, Zorn, Tam, Lamontagne, & Johnson, 2003). Each agency participates within its own system, and at its own pace, yet continually develops toward providing better services in collaboration with others (Kleinhammer-Tramill et al., 1994; Timmons et al., 2004).

Collaboration as a concept may seem elusive, but research has lent clarity to the definition and construct of collaboration (including characteristics of collaborative activities). Abramson and Rosenthal (1995) defined collaboration as "a fluid process through which a group of diverse, autonomous actors (organizations or individuals) undertakes a joint initiative, solves shared problems, or otherwise achieves common goals." Research indicates that while collaboration is a complex concept, it can be broken down into a theoretical model that contains five variable dimensions: (a) governance, (b) administration, (c) organizational autonomy, (d) mutuality, and (e) norms (Thomson & Perry, 2006; Thomson, Perry, & Miller, 2007). These five dimensions can be assessed by the group, with targeted areas for improved collaboration through activities such as creating group norms and improved communication systems, adding relevant decision-makers as team members, and ensuring an equal balance of power among voices and participating agencies. Their work illustrates that many factors influence and support collaboration, which is an inherently complex concept.

Unsurprisingly, collaborative relationships take time to develop. Achieving optimal levels of interagency collaboration requires members to determine how to work together and to communicate effectively. Purposeful collaborative activities can be characterized as: (a) establishing mutual relationships and goals, (b) jointly developing structures and sharing responsibility, (c) sharing authority and crediting everyone with success, and (d) sharing resources and rewards (Mattessich, 2003; O'Looney, 1993; Parent Advocacy Coalition for Educational Rights, 1994; Walsh, Brabeck, & Howard, 1999). Specific behaviors and mechanisms have been shown to increase collaboration over time (Mancini, Marek, Byrne, & Huebner, 2004).

Researchers in a variety of fields have argued that collaboration develops in distinct stages of between three and seven steps (deFur, 1997; Dunst & Bruder, 2002; Frey, Lohmeier, Lee, & Tollefson, 2006; Gajda, 2004; Hogue, 1993; Peterson, 1991). Frey et al.'s (2006) model includes five clear stages: (a) networking, (b) cooperation, (c) coordination, (d) coalition, and (e) collaboration. Networking is characterized by awareness of the organization, loosely defined roles, and low levels of communication for the purposes of referral only. Cooperation, the second stage, is characterized by providing information to each other, somewhat defined roles, formal communication, and independent decision-making. Coordination, the third stage, is exemplified by sharing information and resources, defined roles, frequent communication, and some shared decision-making. The fourth stage is coalition, which consists of shared ideas, shared resources, frequent prioritized communication, and shared decision-making. Finally, the highest stage is collaboration, exemplified by members belonging to one system, frequent communication with mutual trust, and consensus reached on all decisions.

In 2003, Johnson et al. identified seven interrelated factors related to successful interagency collaboration, which are: (a) commitment, (b) communication, (c) strong leadership from key decision-makers, (d) understanding the culture of other agencies, (e) engaging in serious preplanning, (f) providing adequate resources for collaboration, and (g) minimizing turf issues. These factors were evident in professional partnerships, as adults felt it was part of their charge to learn other systems, build relationships with other agency staff, and gain support of their bosses (e.g., dedicated time, ability to contribute to joint activities, some level of decision-making) to participate. Professionals who collaborated gained an awareness of the culture of other agencies, such as pressures, organization structures, and priorities. That awareness and understanding led to more joint planning and less competition and hesitancy to partner on activities that supported youth and adults with disabilities.

A significant focus of collaboration research has been identifying what quality collaboration in transition looks like, as well as common characteristics of the educators and communities involved. The research led to the identification and description of the standard or goal for collaboration: when agencies collaborate, they function as one entity and are actively engaged in problem-solving, information sharing, and merging of resources (Cashman, 1995; deFur, 1997). When educators and agency professionals can function as one entity, individual members of the transition team are able to make better decisions, as the collective wisdom of the group provides many alternative solutions and ways to address needs (deFur, 1997). This benefits not only the agencies and the student but also families. These collaborative efforts result in families gaining role clarity and greater understanding of the differences between K-12 and adult services and supports, as well as the strengths and limitations of various service provider offerings.

Additionally, these collaborative efforts cannot happen once, or be time-limited. Successful interagency collaboration needs to be sustained, systematized, and characterized by: (a) key positions jointly funded by education and adult services, (b) monthly interagency planning meetings, (c) cross-agency training opportunities, and (d) the use of a variety of practices for collaboration and team-building (Benz, Lindstrom, & Yovanoff, 2000; Hasazi et al., 1999). Mechanisms such as ongoing teaming, joint training, and merged funding are important elements to keep collaboration efforts going over time to truly meet the vision of operating as one entity with strong collaboration.

Researchers have examined and identified specific strategies for increasing collaboration (Spath, Werrbach, & Pine, 2008). One strategy is the concept of "linking agents" (Crandall, 1977; Hamilton et al., 2002; Havelock & Havelock, 1971). A linking agent is a person who fulfills one or more of the four key roles to facilitate collaboration: (a) catalysts, who empower others to bring about change; (b) solutionists, who build awareness of the new ideas; (c) facilitators, who support the processes and procedures that effect change; and (d) linkers, who link to resources and help others receive the support, information, and expertise they need for long-term, sustainable change. The highly knowledgeable linking agent functions as the "go between" for numerous entities and agencies to bridge the implementation gap and facilitate change (Monahan & Scheirer, 1988).

In 2008, Noonan, Morningstar, and Gaumer Erickson identified eleven key strategies that high-performing districts implemented at the local-level related to interagency collaboration. The strategies are: flexible scheduling and staffing, follow-up after transition, administrative support, variety of funding sources, state-supported technical assistance, ability to build relationships, agency meetings with students and families, training students and families, joint training of school and agency staff, meetings with agency staff and transition councils, and dissemination of information to a broad audience. Research results indicated the clear need for a systematic approach to local community readiness and commitment of key stakeholders (i.e., special educators, transition coordinators, administrators, families, and agency staff). Many of the identified strategies required joint efforts of school administrators and staff, community agencies, and families—meaning that the task of increasing collaboration could not fall solely on one entity.

In 2014, Dr. Noonan and the Council for Exceptional Children published the book *Transition Teaming: 26 Strategies for Interagency Collaboration* (Noonan, 2014). The book provides secondary special educators and their adult agency counterparts with concrete strategies for building collaborative relationships. Part of the book focuses on strategies that individual educators can use immediately, such as reflection, increased awareness of partners, relationship-building activities, and outreach. Through these daily mechanisms, individuals expand their personal efforts toward collaboration. In the book, each personal strategy is identified and then described with specific activities and examples in detail. Strategies and activities include:

Strategy 1: Gain an understanding of how coworkers' jobs are related to transition.

- Interview school personnel to better understand charge.

Strategy 2: Increase awareness and knowledge of adult agency services.

- Develop or expand a list of community resources.
- Join community organizations and committees.

Strategy 3: Gain administrator buy-in.

- Present at a school board meeting.
- Update administration regularly via email or meetings.

Strategy 4: Communicate information about transition to coworkers.

- Present to coworkers.
- Provide information in a communal space.

Strategy 5: Communicate information about transition to local community.

- Develop an "elevator speech."
- Write an article for the local newspaper.
- Present to community organizations.

Strategy 6: Communicate information about transition to families.

- Disseminate information to families.
- Facilitate meetings with agencies, students, and families.
- Tour local agencies.

Strategy 7: Coordinate with coworkers to provide transition services.

- Assess your school's college and career readiness practices.
- Identify school assessment practices that support transition.

Strategy 8: Coordinate with community agencies to provide transition services.

- Connect your students to community agencies.
- Plan a collaborative project.

Strategy 9: Participate in professional development related to transition.

- Attend regional or state transition conferences.
- Join national organizations and attend national conferences.

Strategy 10: Participate in professional development sponsored by community agencies.

- Attend trainings provided by community entities.

In later chapters, *Transition Teaming: 26 Strategies for Interagency Collaboration* details a step-by-step process to form a functioning, action-oriented transition team. In the early 1990s, community transition teams (CTTs) emerged as a model of facilitating interagency collaboration, with the key function of creating linkages between education and adult services through teaming (Blalock & Benz, 1999). Community transition teams (also referred to as transition councils) are typified as being composed of local, community-level representatives of schools; disability-related agencies and community organizations; families and students; and other stakeholders who join together to improve local transition services for youths with disabilities. The theory behind this model is that community transition teams will be able to help students and families secure resources to accomplish their transition plan by improving the capacity of schools and communities to deliver better services (Benz, Lindstrom, & Halpern, 1995). Because community transition teams focus on services at the local level, they are better able to: (a) share resources, (b) hold informational fairs, and (c) influence local policies and procedures (deFur, 1999). Team membership often includes representatives of the local schools, students and/or former students, family members, One-Stop centers, vocational rehabilitation, developmental disability organizations, independent living centers, post-secondary training organizations, and local businesses (Halpern, Benz, & Lindstrom, 1992). Community transition teams work to create community cohesiveness and increase community capacity by avoiding service duplication and targeting emerging community transition needs (Clark & McDonnell, 1994). As Benz, Lindstrom, and Halpern (1995) reflected, systems change depends on the critical components of: (a) active participation of diverse stakeholders, (b) viewing change as a process and not as an event, and (c) local community partnerships that are supported by a larger structure that sustains and validates efforts.

Community transition teams work to identify needs, plan and implement new programs, and evaluate team efforts in order to modify and improve services for youths with disabilities. Structured teaming with the ongoing use of data to drive decision-making sustains transition teaming (Noonan, 2014). Through strategies 11–26, individuals expand their interpersonal efforts toward collaboration. In the book, each teaming strategy is identified and described with specific activities and examples in detail. Strategies and activities include:

Strategy 11: Develop a community transition team.

- Discuss your community's and students' needs.
- Generate a list of potential team members.
- Plan the first meeting.

Strategy 12: Identify a shared vision.

- Identify individual visions.
- Identify a team vision.

Strategy 13: Conduct community transition team meetings.

- Establish meeting norms.
- Use agendas and minutes.

Strategy 14: Organize your community transition team.

Strategy 15: Conduct community resource mapping.

Strategy 16: Host a community conversation.

Strategy 17: Determine team structure—critical representation, team organization.

Strategy 18: Engage in action-oriented teaming—annual planning, action planning, short-term projects.

- Create a detailed action plan spanning a specific period of time.
- Brainstorm specific activities that will lead to a commonly desired outcome.
- Provide sufficient, relevant details and include follow-up steps.

Strategy 19: Come to consensus.

- Adopt techniques for quick voting, weighted voting.

Strategy 20: Assess shared leadership.

Strategy 21: Use Data as a community transition team.

Strategy 22: Use data that is already available to your community transition team.

- Review post-school outcome and academic data.
- Discuss community data, such as employment data.

Strategy 23: Engage in data collection and information gathering.

- Utilize surveys to improve transition curriculum.
- Research to expand opportunities or services.
- Create your own survey.

Strategy 24: Plan for sustainability.

- Identify sustainability strengths and barriers.
- Launch a sustainability survey and develop sustainability goals.
- Apply for funding.
- Create brochures, articles, and products that preserve the history.

Strategy 25: Develop bylaws.

Strategy 26: Develop formal Interagency Agreements.

- Create Memorandums of Understanding (MOUs).

To further facilitate community transition teaming, Noonan and Gaumer Erickson (2013) developed a fidelity tool, where teams discussed several indicators and determined if each was in place. If it was not in place, teams brainstormed action items to promote better teaming. Indicators include:

1. Community transition team is established and includes critical representation based on community needs (e.g., school personnel, Centers for Independent Living, Department of Mental Health and Vocational Rehabilitation counselors, family members, employers).
2. Community transition team is representative of community and reflects community needs.
3. Community transition team has established a team name and used the Mini-Maps process to develop a shared vision with the expanded team.
4. Community transition team meets monthly at a minimum.
5. Community transition team has identified community and transition areas of need and prioritized five major goals. Entire team is knowledgeable of these goals.
6. Based on team goals, community transition team jointly developed an action plan that outlines activities, persons responsible, and timeline for reaching goals. Action plan is reviewed and updated at every meeting.
7. Team norms or ground rules have been established and agreed upon.
8. Meeting structure (i.e., agenda and timing) has been established and agreed upon.
9. An organizational system for tracking meeting notes, materials, and data has been established and is maintained.
10. A system of assigned or rotated roles (e.g., facilitator, note-keeper, time-keeper) is defined to assure high quality and effective meeting time.
11. Community transition team members have equal voice when planning team activities.
12. Process for reaching a team decision (i.e., consensus or majority vote) has been defined and adopted.
13. Building administration—that is, principal or district level administrator—attends community transition team meeting at least three times a year (e.g., fall, winter, spring) and receives agenda and minutes for every meeting.
14. Community transition team collaboratively reflects on areas of local need identified through data (e.g., Indicator 13 compliance data, drop-out data, graduation rates, outcomes, etc.).
15. Community transition team systematically uses data to drive decision-making.
16. Community transition team systematically shares information with appropriate district-wide staff and administrators.
17. Community transition team systematically shares transition information with community and families.
18. Community transition team systematically communicates with surrounding districts about team initiatives.
19. Transition-related professional development events are included on CTT meeting agendas for dissemination and discussion.
20. At least annually, community transition team revisits and updates an action plan that addresses the prioritized needs identified through data analysis.
21. District leadership is familiar with the contents of the action plan.
22. Community transition team membership is reviewed at least annually and new members are recruited.
23. A process is in place to welcome new members to team.
24. Bylaws have been developed by the community transition team.
25. Sustainability plan has been developed by CTT and is revisited at least annually.

On an ongoing basis, community transition teams can discuss the list of indicators in order to identify strengths as well as areas for improvement. The higher the degree to which teams address the indicators, the more likely they are to sustain membership and maintain a high level of functioning.

Competencies to Support Collaboration

Although specific strategies for interagency collaboration have been identified, professionals from both education and support agencies are not always provided foundational training on the behaviors necessary to develop collaborative working relationships or productive teams. Knott and Asselin (1999) surveyed secondary special education regarding the most important aspects of their jobs, and found that while teachers rated interagency collaboration as important, they did not know how to accomplish this task. Teachers also indicated that pre-service and in-service training needed to focus less on philosophical, historical, and legal foundations of transition and more on communication and interagency collaboration. Some felt that training should move beyond, "What is transition planning?" to "How do I accomplish planning that will lead to successful transitions?" (Knott & Asselin, 1999, p.3). Li (2004) surveyed 1000 secondary special educators and transition coordinators and found that educators as a whole recognized the importance of interagency collaboration (4.35 on 5.0 scale). This study also found that if the respondents were adequately trained in and committed to transition, they had higher perceptions of interagency collaboration and more transition involvement. Therefore, two major barriers to interagency collaboration are a lack of training in transition and a lack of commitment to transition activities (Li, 2004).

Collaboration must begin with adults using clear, purposeful behaviors to build relationships with each other. In its essence, "collaboration is a way of thinking and relating, a philosophy, a paradigm shift, an attitude change. It requires a set of behaviors, beliefs, attitudes, and values. The result is a sense of shared ownership, shared responsibility, shared success" (Bishop, Wolf, & Arango, 1993).

When educators and adult agency staff agree that building partnerships with others is part of their personal charge, they expand their efforts to identify and implement strategies to increase collaboration. These concepts and strategies can be embedded to improve transition outcomes for students. To increase collaboration between entities, professionals should focus efforts at the individual level on a daily basis. As we work to collaborate, the benefits are immense, as students with intellectual disabilities will have better access to higher quality services. Professionals will benefit from improved staff relationships and the cross-fertilization of ideas. This shared responsibility may bring expenses at times, but it also comes with decreased duplication of efforts. Despite the potential gains, many educators find themselves unable to implement collaboration strategies due to limitations in necessary inter- and intrapersonal competencies. To address this need, professionals can work to develop these competencies by learning the basics, practicing, taking risks, and working to increase self-awareness throughout the process. Six approaches can support educators and disability professionals in a self-improvement process, including:

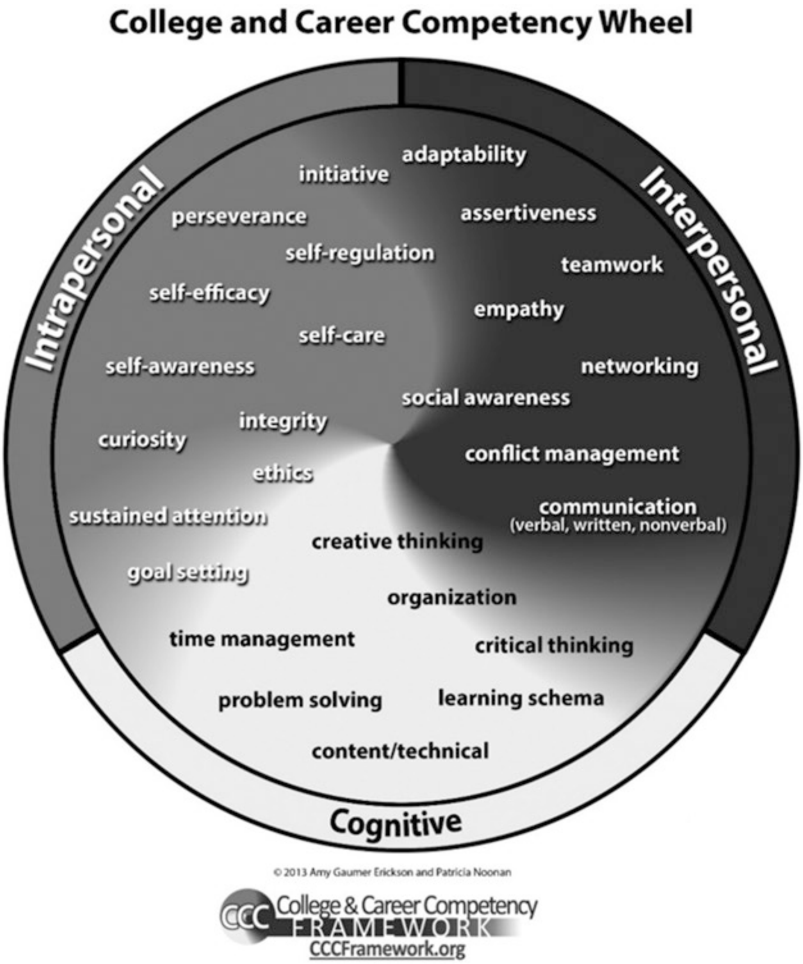
1. Speak a common language.
2. Learn services and agencies outside your own.
3. Partner with others to accommodate and meet their charge.
4. Employ action-oriented behaviors.
5. Work through past and current issues.
6. Believe that your collaboration abilities will improve.

Speak a Common Language

To speak a common language among professionals, we must begin describing students' intra- and interpersonal strengths in a consistent, easy-to-understand way to allow better communication between educators, adult agency staff, students, parents, nonprofits, and employers. As Noonan and Gaumer Erickson note in their 2018 book *The Skills that Matter: Teaching Interpersonal and Intrapersonal Competencies in Any Classroom,* college and career competencies can provide us a language to communicate with each other more meaningfully. Their work includes the *College and Career Competency Wheel*, which identifies 26 competencies critical to in-school and post-school success, categorized into three domains: intrapersonal (focusing on students' internal, reflective capacities), interpersonal (focusing on capacities related to cooperation and interaction with others), and cognitive (focusing on effectively processing and using information).

The 26 competencies can be considered social/emotional competencies, as well as critical employability skills for success in career paths and post-school education and training. Each of the 26 competencies has a foundation of evidence that they can be taught to adolescents, have demonstrable behaviors associated with them, and if developed, impact in-school and post-school achievement (see http://CCCFramework.org for Teacher Guides that summarize the research and provide details on instructional practices and assessments).

The *College and Career Competency Wheel* can be used to describe critical competencies for students' post-school success, but also to articulate an overall vision or purpose of a group. The competency wheel promotes a common language with students, teachers, employers, community members, law enforcement, etc. We can embed these competencies into our instruction and this will positively impact students with disabilities, gifted students, those at risk, and those on track, regardless of the student's career path. Additionally, professionals from adult services, nonprofits, and even families can understand and support the development of the competencies. As professionals from numerous disciplines examine and embrace the vision of better-prepared students, they are better able to work together, share resources, and advocate for policy changes to create more socially and emotionally engaged, career-equipped, lifelong learners. This common charge promotes collaborative behaviors from all sides.



Learn Services and Agencies Outside Your Own

In order to learn and work with services and agencies outside of your own, it is important to build relationships with other invested partnering agencies. To first learn and then truly partner with other entities, consider focusing on your networking abilities.

Networking includes these essential components:

- Create ties among individuals.
- Utilize ties for support to overcome barriers and achieve goals.
- Support others to overcome barriers and achieve goals (Gaumer Erickson & Noonan, 2013).

Consider the degree to which you currently network with agencies such as vocational rehabilitation, parent organizations, workforce centers, and nonprofits. You may be aware of a person working within an entity, and you may even have exchanged contact information or discussed a student. These are good steps in beginning to network and build relationships, but genuine networking requires more. To fulfill the components of networking, we not only create the connections or ties, but also use them for support to overcome barriers and achieve goals, such as coordinating services for a student in need. And more importantly, we use our connections to support other professionals to overcome barriers and achieve goals, even when it might not directly benefit our role or our students. In other words, we use our ties to make connections between others so that they build mutually beneficial relationships with others. In this way, we are not simply capturing contact information for later use, but instead are building and participating in a true professional network.

Partner with Others to Accommodate and Meet Their Charge Also

So often when we strive to collaborate with others, we make the mistake of primarily focusing on identifying and communicating our needs. While asking for help is important, we want to do so in a way that strives to build relationships with others based on authentic mutual understanding. Focusing on understanding and improving our assertiveness and empathy can help us build quality professional relationships for effective collaboration.

Assertiveness supports both making connections with others and keeping these relationships strong, and includes these essential components:

- Even when it's difficult, express my wants, needs, and thoughts.
- Even when it's difficult, respect what others want, need, and think (Gaumer Erickson & Noonan, 2013).

Before we can effectively express our wants, needs, and thoughts, we must first be aware of and understand them. To be truly assertive, in addition to articulating our points, we must respect others' points of view. Too often, professionals can operate in ways that are too passive or too aggressive, such as not genuinely engaging in discussion or dominating conversations and agendas. Assertiveness can benefit both adults who are too passive and those who are too aggressive by enhancing their ability to communicate more effectively. In groups, we have all experienced a dominating group member who directs decision-making without adequately listening to diverse perspectives. It is likely that these groups or teams are not functioning at high levels, and may not be effective in achieving goals or completing activities. As we self-assess our assertiveness, we may find that this is an area for growth through techniques like active listening, creating and using assertive statements, and ongoing reflection.

In addition to assertiveness, empathy supports the building and maintaining of relationships. To practice empathy as adults, we:

- Make efforts to understand others: their contexts, feelings, and behaviors.
- Communicate our understanding of someone's personal situation (Gaumer Erickson & Noonan, 2013).

While this can take time and require structures such as one-on-one time, private meeting space, and active listening techniques, striving to understand others is critical to working together effectively. Adult agencies are supported by different funding streams which often have supporting legislation, regulation, procedures, and processes. Different work cultures, pressures, timelines, and priorities influence the work of peer professionals. In working to build understanding and communicate that understanding, you will increase awareness of opportunities for mutual benefit and critical connections.

Employ Action-Oriented Behaviors

When working with others, it is often necessary to identify clear tasks or goals that you will work on and divide the work across the team. In emerging relationships, these early experiences accomplishing action-oriented tasks are critical ways to build trust and understanding. At times, adults and team members fail to accomplish important goals or long-term tasks. Often, the reason behind the failure is not a lack of desire to do the work, but a breakdown somewhere in the process. While a clear overall goal may be identified, adults at times lack the self-regulation necessary to accomplish each individual activity or task involved. This might include difficulty getting back on track when things do not go as planned. Self-regulation and teamwork can support our efforts to produce quality work in a timely fashion.

Self-regulation consists of four essential components:

- Plan for and articulate what you want to accomplish
- Immediately monitor progress and interference regarding your goal.
- Control change by implementing specific strategies when things are not going as planned.
- Reflect on what worked and what you can do better next time (Gaumer Erickson & Noonan, 2013).

Many of us have experienced working to accomplish a task, but then failing due to unforeseen events. Adults can practice self-regulation by identifying a clear goal or task, creating a detailed plan, and monitoring the plan for not only progress, but also challenges that might be preventing progress. If something is preventing progress, a critical piece of self-regulation (and one that many people have difficulty with) is taking action to address the barrier by identifying and using specific strategies to continue moving forward. Finally, it is necessary to reflect throughout the process. When we use self-regulation in this way, we are better able to meet deadlines as intended and achieve our overarching goals.

Basic teamwork components can support the success of individual team members as well as the overall functioning of the team. Teamwork consists of these essential components:

- Work effectively with others to achieve a common goal.
- Do your fair share of any team assignments.
- Share your ideas or express your opinions while being open to others' ideas, opinions, and perspectives.
- Respect your fellow team members even when you do not agree with them (Gaumer Erickson & Noonan, 2013).

Have you been on a team where members failed to address one or more of these components? Many of us have, and it is often a difficult teaming environment. Consider your strengths and areas for improvement for working on a team. As each team member works on personal behaviors and components, the team as a whole accomplishes more work of better quality, and membership is sustained.

Work Through Past and Current Issues

Many adult relationships suffer from the inability to address conflict, with people predominantly choosing avoidance as a response. While avoidance can be a good response in some conflict situations, more sophisticated responses of cooperation and collaboration yield better results and stronger relationships.

To manage conflict, consider these three essential components:

- Understand your natural response to conflict.
- Understand the context of the conflict, including the perspectives of all involved.
- Apply a conflict management approach that is appropriate to the situation (Gaumer Erickson & Noonan, 2013).

Understanding how we typically respond to conflict and learning new strategies for constructively addressing conflict supports our ability to work effectively with others. Conflict is a normal occurrence that happens often, but our natural response may include less desirable approaches, such as competition and avoidance. Building our self-awareness and employing strategies to better understand the context and perspectives involved can help us in trying alternate, more effective approaches to conflict.

Believe that Your Collaboration Abilities Will Improve

Sometimes we struggle to understand that just because we are not good at something at first, it does not mean that we cannot learn it or improve with effort. Self-efficacy refers to perceptions an individual has about his or her capabilities to perform at an expected level and achieve goals or milestones.

The main components of self-efficacy are:

- Self-efficacy increases with the belief that ability can grow with effort.
- Believe in your ability to meet specific goals and/or expectations (Gaumer Erickson & Noonan, 2013).

As we embrace collaborative opportunities, our outcomes may not initially be as successful as hoped. Mistakes are easily made in new partnerships as we struggle to understand context and perspectives of others. We must realize that our abilities to work with other professionals grow as we continue our efforts. With each small success in challenging tasks, our self-efficacy as collaborators grows.

Summary

While the task of increasing collaboration on a personal level through teaming strategies and/or personal competency development may seem overwhelming, focusing on small parts is a good approach. Even minor behaviors could add meaning to your work with students and young adults—positively impacting students' post-school achievement. Throughout this work, it is important to take risks. As we leave our comfort zone and expand how we conceive our work and how we support students, we will sometimes fail, but we will know that although we have not mastered that strategy yet, our abilities are growing as we take risks. Finally, as educators, we are often so thoroughly focused on providing supports to students that we rarely spend any time acknowledging our personal or team successes. Make time to acknowledge your own achievements, as well as those of your partners. This both sustains and expands the effort and allows us to be a part of a larger community all working toward improved post-school outcomes.

References

Abramson, J. S., & Rosenthal, B. B. (1995). Interdisciplinary and interorganizational collaboration. In *Encyclopedia of social work* (19th ed.). Washington, DC: NASW.

Agran, M., Cain, H. M., & Cavin, M. D. (2002). Enhancing the involvement of rehabilitation counselors in the transition process. *Career Development for Exceptional Individuals, 25*(2), 141–155.

Benz, M. R., Johnson, D. K., Mikkelsen, K. S., & Lindstrom, L. E. (1995). Improving collaboration between school and vocational rehabilitation: Stakeholder identified barriers and strategies. *Career Development for Exceptional Individuals, 18*(2), 133–144.

Benz, M. R., Lindstrom, L. E., & Halpern, A. S. (1995). Mobilizing local communities to improve transition services. *Career Development for Exceptional Individuals, 18*(1), 21–32.

Benz, M. R., Lindstrom, L. E., & Yovanoff, P. (2000). Improving graduation and employment outcomes of students with disabilities: Predictive factors and student perspectives. *Exceptional Children, 66*(4), 509–529.

Bishop, K. K., Wolf, J., & Arango, P. (1993). *Family/professional collaboration for children with special health care needs and their families* (pp. 11–12). Burlington, VT: Department of Social Work, University of Vermont.

Blalock, G. (1996). Community transition teams as the foundation for transition services for youth with learning disabilities. *Journal of Learning Disabilities, 29*(2), 148–159.

Blalock, G., & Benz, M. R. (1999). *Using community transition teams to improve transition services*. Austin, TX: PROe-ED.

Bullis, M., Davis, C., Bull, B., & Johnson, B. (1995). Transition achievement among young adults with deafness: What variables relate to success? *Rehabilitation Counseling Bulletin, 39*, 130–150.

Carter, E. W., Trainor, A. A., Cakiroglu, O., Cole, O., Swedeen, B., Ditchman, N., & Owens, L. (2009). Exploring school-employer partnerships to expand career development and early work experiences for youth with disabilities. *Career Development for Exceptional Individuals, 32*(3), 145–159.

Cashman, J. (1995). Collaboration and reform: The role of interagency linkages in developing a coherent strategy for transition. *Journal for Vocational Special Needs Education, 17*(3), 103–107.

Certo, N. J., Pumpian, I., Fisher, D., Storey, K., & Smalley, K. (1997). Focusing on the point of transition: A service integration model. *Education and Treatment of Children, 20*(1), 68–84.

Clark, G. M., & McDonnell, J. (1994). The role of local transition councils in rural communities. *Rural Special Education Quarterly, 13*(1), 3–8.

Crandall, D. P. (1977). Training and supporting linking agents. In N. Nash & J. Culbertson (Eds.), *Linking processes in educational improvement: Concepts and applications* (pp. 189–274). Columbus, OH: University Council for Educational Administration.

deFur, S. H. (1997). Collaboration as a prevention tool for youth with disabilities. *Preventing School Failure: Alternative Education for Children and Youth, 41*(4), 173–178.

deFur, S. H. (1999). Transition, special education, and school-based services: Are they meant for each other. In S. H. deFur & J. Patton (Eds.), *Transition and school-based service: Perspectives for enhancing the transition process* (pp. 15–50). Austin, TX: Pro-Ed.

Dunst, C. J., & Bruder, M. B. (2002). Valued outcomes of service coordination, early intervention, and natural environments. *Exceptional Children, 68*(3), 361–375.

Frey, B. B., Lohmeier, J. H., Lee, S. W., & Tollefson, N. (2006). Measuring collaboration among grant partners. *American Journal of Evaluation, 27*(3), 383–392.

Gajda, R. (2004). Utilizing collaboration theory to evaluate strategic alliances. *American Journal of Evaluation, 25*(1), 65–77.

Gaumer Erickson, A. S., & Noonan, P. M. (2013). *College and career competency framework: Networking Teacher Guide*. Retrieved from http://researchcollaboration.org/uploads/TeacherGuide-Networking.pdf.

Halpern, A. S. (1994). The transition of youth with disabilities to adult life: A position statement of the Division on Career Development and Transition, the Council for Exceptional Children. *Career Development for Exceptional Individuals, 17*(2), 115–124.

Halpern, A. S., Benz, M. R., & Lindstrom, L. E. (1992). A systems change approach to improving secondary special education and transition programs at the community level. *Career Development for Exceptional Individuals, 15*(1), 109–120.

Hamilton, J., Dailey, D., Mesmer, E., Ritter, S., Shami, M., Nishi, L., … Bauman, W. (2002). *Elementary and middle school technical assistance center (EMSTAC): Final report*. Washington, DC: American Institutes for Research.

Hasazi, S. B., Furney, K. S., & DeStefano, L. (1999). Implementing the IDEA transition mandates. *Exceptional Children, 65*(4), 555–566.

Havelock, R. G., & Havelock, M. C. (1971). *Training for change agents: A guide to the design of training programs in education and other fields*. Ann Arbor, MI: Institute for Social Research.

Hodges, S., Nesman, T., & Hernandez, M. (1999). *Promising practices: Building collaboration in systems of care. Systems of care: Promising practices in children's mental health*. Rockville, MD: Substance Abuse and Mental Health Services Administration [SAMHSA].

Hogue, T. (1993). *Community-based collaboration: Community wellness multiplied*. Bend, OR: Chandler Center for Community Leadership. Retrieved from http://crs.uvm.edu/nnco/collab/wellness.html

Johnson, D. R., Stodden, R. A., Emanuel, E. J., Luecking, R., & Mack, M. (2002). Current challenges facing secondary education and transition services: What research tells us. *Exceptional Children, 68*(4), 519–532.

Johnson, D. R., Thurlow, M. L., & Stout, K. E. (2007). *Revisiting graduation requirements and diploma options for youth with disabilities: A national study*. (Technical Report 49). Minneapolis, MN: University of Minnesota, National Center on Educational Outcomes.

Johnson, L. J., Zorn, D., Tam, B. K., Lamontagne, M., & Johnson, S. A. (2003). Stakeholders' views of factors that impact successful interagency collaboration. *Exceptional Children, 69*(2), 195–209.

Kleinhammer-Tramill, P. J., Rosenkoetter, S. E., & Tramill, J. L. (1994). Early interventions and secondary/transition services: Harbingers of change in education. *Focus on Exceptional Children, 27*(2), 1–14.

Knott, L., & Asselin, S. B. (1999). Transition competencies: Perceptions of secondary special education teachers. *Teacher Education and Special Education, 22*(1), 55–65.

Kohler, P. D., & Field, S. (2003). Transition-focused education: Foundation for the future. *The Journal of Special Education, 37*(3), 174–183.

Li, J. (2004). *Interagency collaboration in transition practice: An initial model* (Published Doctor of Education dissertation). Greeley, CO: University of Northern Colorado.

Mancini, J. A., Marek, L. I., Byrne, R. A., & Huebner, A. J. (2004). Community-based program research: Context, program readiness, and evaluation usefulness. *Journal of Community Practice, 12*(1–2), 7–21.

Mattessich, P. (2003). *Can this collaboration be saved? Twenty factors that can make or break any group effort*. Montclair, NJ: National Housing Institute. Retrieved from https://shelterforce.org/2003/05/01/can-this-collaboration-be-saved/

Monahan, J. L., & Scheirer, M. A. (1988). The role of linking agents in the diffusion of health promotion programs. *Health Education Behavior, 15*(4), 417–433.

Morningstar, M. E., Kleinhammer-Tramill, P. J., & Lattin, D. L. (1999). Using successful models of student-centered transition planning and services for adolescents with disabilities. *Focus on Exceptional Children, 31*(9), 2–20.

National Center for Secondary Education and Transition [NCSET]. (2004). *Findings & outcomes: State priorities and needs for assistance.* (Report from the NCSET National Leadership Summit on Improving Results for Youth). Minneapolis, MN: University of Minnesota, Institute on Community Integration. Retrieved from http://www.ncset.org/summit03/findings.htm

Newman, L., Wagner, M., Cameto, R., Knokey, A. M., & Shaver, D. (2010). *Comparisons across time of the outcomes of youth with disabilities up to 4 years after* *high school: A report of findings from the National Longitudinal Transition Study (NLTS) and the National Longitudinal Transition Study-2 (NLTS2)*. Menlo Park, CA: National Center for Special Education Research.

Noonan, P. M. (2014). *Transition Teaming: 26 Strategies for Interagency Collaboration*. Arlington, VA: Council for Exceptional Children.

Noonan, P. M., & Gaumer Erickson, A. S. (2013). *Team process checklist*. Lawrence, KS: University of Kansas, Research Collaboration.

Noonan, P. M., & Gaumer Erickson, A. S. (2018). *The skills that matter: Teaching interpersonal and intrapersonal competencies in any classroom*. Thousand Oaks, CA: Corwin.

Noonan, P. M., Morningstar, M. E., & Gaumer Erickson, A. (2008). Improving interagency collaboration: Effective strategies used by high-performing local districts and communities. *Career Development for Exceptional Individuals, 31*(3), 132–143.

Noonan, P. M., Morningstar, M. E., & Lattin, D. L. (2008). *Arizona community transition team manual: Developing comprehensive community-wide transition systems to improve results*. Lawrence, KS: University of Kansas, Transition Coalition.

Noyes, D. A., & Sax, C. L. (2004). Changing systems for transition: Students, families, and professionals working together. *Education and Training in Developmental Disabilities, 39*(1), 35–44.

O'Looney, J. (1993). Beyond privatization and service integration: Organizational models for service delivery. *Social Service Review, 67*(4), 501–534.

Parent Advocacy Coalition for Educational Rights [PACER]. (1994). *Interagency collaboration and transition*. Salt Lake City, UT: Center for Expertise. Retrieved from http://www.pacer.org

Peterson, N. L. (1991). Interagency collaboration under Part H: The key to comprehensive, multidisciplinary, coordinated infant/toddler intervention services. *Journal of Early Intervention, 15*, 89–105.

Rabren, K., Dunn, C., & Chambers, D. (2002). Predictors of post-high school employment among young adults with disabilities. *Career Development for Exceptional Individuals, 25*(1), 25–40.

Repetto, J. B., Webb, K. W., Garvan, C. W., & Washington, T. (2002). Connecting student outcomes with transition practices in Florida. *Career Development for Exceptional Individuals, 25*(2), 123–139.

Spath, R., Werrbach, G. B., & Pine, B. A. (2008). Sharing the baton, not passing it: collaboration between public and private child welfare agencies to reunify families. *Journal of Community Practice, 16*(4), 481–507.

Taylor, H., Krane, D., & Orkis, K. (2010). *The ADA, 20 years later*. New York: Harris Interactive. Retrieved October 19, 2010, from http://www.2010disabilitysurveys.org/pdfs/surveyresults.pdf

Test, D. W., Fowler, C. H., Richter, S. M., White, J., Mazzotti, V., Walker, A. R., … Kortering, L. (2009). Evidence-based practices in secondary transition. *Career Development for Exceptional Individuals, 32*(2), 115–128.

Thomson, A. M., & Perry, J. L. (2006). Collaboration processes: Inside the black box. *Public Administration Review, 66*(s1), 20–32.

Thomson, A. M., Perry, J. L., & Miller, T. K. (2007). Conceptualizing and measuring collaboration. *Journal of Public Administration Research and Theory, 19*(1), 23–56.

Timmons, J. C., Cohen, A., & Fesko, S. L. (2004). Merging cultural differences and professional identities: Strategies for maximizing collaborative efforts during the implementation of the Workforce Investment Act. *Journal of Rehabilitation, 70*(1), 19–27.

Walsh, M. E., Brabeck, M. M., & Howard, K. A. (1999). Interprofessional collaboration in children's services: Toward a theoretical framework. *Children's Services: Social Policy, Research, and Practice, 2*(4), 183–208.

Wehman, P. (1998). *Editorial. Journal of Vocational Rehabilitation, 10*(1), 1–2.

12 Reports and How to Write Them

Jerrica Guidry, Peter Castagna,
and Thompson E. Davis III

Introduction

Psychological testing has been a cornerstone of psychological practice for decades, spanning back to the initial development of intelligence. During the early 1900s, Sante De Sanctis and Alfred Binet developed the first mental tests to determine the "educability" of children with abnormal delays (as cited in Cicciola, Foschi, & Lombardo, 2014). From there, Binet revised his 1905 intelligence test a number of times, resulting in what he claimed was an objective, scientific way to measure the complex mental phenomenon known as intelligence (as cited in Cicciola, 2008). Intelligence tests are designed to measure a variety of mental functions, including reasoning, comprehension, and judgment. Since there are no universal biomarkers associated with intellectual disability diagnoses rely heavily on evaluation practices (Tasse, 2016).

It is estimated that approximately three million Americans have intellectual disability (Larson et al., 2001). Given the implications of an intellectual disability diagnosis, a thorough understanding of the diagnostic criteria is crucial. Significant shifts in diagnostic criteria, terminology, and expectations and inclusiveness over time have advanced the protection and quality of life of people with intellectual disabilities; however, changes have also inevitably caused confusion among some evaluators. For instance, what is now known as "intellectual disability," was previously termed "mental retardation." Increasingly, there have also been growing movements to include those with intellectual disability in areas such as education and employment (Fesko, Hall, Quinlan, & Jockell, 2012).

The American Association on Intellectual and Developmental Disabilities (AAIDD) defined "intellectual disability" in an individual in 2002 as someone with significant limitations in both intellectual functioning and adaptive behavior including social, practical, and conceptual skills, which begins before the age of 18. Although U.S. federal law (Developmental Disabilities Act of 2000; PL 106-402) defines the end of the developmental period for developmental disabilities at age 22, the end of the developmental period for intellectual disabilities has historically been set at the age of 18 (Schalock et al., 2010). AAIDD continues their definition of intellectual disability (ID) with intellectual quotient (IQ) score cutoffs at a standard score of 70, which is 2 standard deviations below the mean of 100 for most IQ tests.

The *Diagnostic and Statistics Manual of Mental Disorders* (*DSM-5*) agrees on the need to evaluate both intellectual functioning and adaptive

J. Guidry · P. Castagna · T. E. Davis III (✉)
Department of Psychology, Louisiana State University, Baton Rouge, LA, USA
e-mail: ted@lsu.edu

J. L. Matson (ed.), *Handbook of Intellectual Disabilities*, Autism and Child Psychopathology Series, https://doi.org/10.1007/978-3-030-20843-1_12

behavior; however, following new American Psychiatric Association guidelines, there is no set age where the developmental period ends. The *DSM-5* states that clinical judgment should be used to determine if the onset of intellectual disabilities happens during the developmental period even after the age of 18 (American Psychiatric Association, 2013). Clinical judgment should not just be the opinion of the assessor but should be based on the standardized tools in combination with relevant clinical evaluations (Schalock & Luckasson, 2013). As a result, evaluations for ID typically generate a great deal of data, background information, developmental history, medical history, etc. and frequently challenge practitioners to summarize this information in a succinct, practical format.

Begin with Consent

Informed consent is an ongoing process; the client is provided information about their rights and possible risks/discomforts that may be associated with the evaluation. This allows the clinician to uphold the rights to confidentiality in regard to the evaluation and report. The legal and mental capacity of the person on which the evaluation is being conducted will determine whether they are able to provide consent on their own behalf or if a legal guardian will be in charge of decisions. Legal capacity is the right to make decisions for oneself and have these decisions upheld (Devi, 2013; Quinn, 2011). Mental capacity refers to the individual's ability to make decisions cognitively; this differs from legal capacity, which looks more at the person's ability to express will (McSherry, 2015). There are multiple components of mental capacity including understanding, appreciation, reasoning, and choice. Figure 12.1 (adapted from Grisso & Appelbaum, 1998) explains what components to consider when determining a person's mental capacity. Individuals with ID often have parts, if not all, of their legal capacity revoked and are appointed guardianship with consideration of the severity of intellectual disability (Kohn, Blumenthal, & Campbell, 2013).

When conducting an evaluation, client's mental capacity and legal guardianship should be assessed before continuing. If legal guardianship is appointed to someone other than the client, the guardian should be well informed on the procedures continuously throughout the evaluation. Even if an adult with ID does not have the legal or mental capacity to provide informed consent, informed assent should be included so that all parties involved agree and understand their rights—a developmentally appropriate description of the procedures and testing to be conducted should be provided. Informed assent should be sought for those with a legal guardian. Children being assessed for ID should also receive informed assent regardless of mental abilities due to their vulnerable nature and their inability to make legal decisions. Furthermore, informed assent is a vital aspect of the rights to those with ID. Previous research has shown that individuals diagnosed with ID are more likely to respond affirmatively to questions despite not understanding what is being asked (Sigelman, Budd, Spanhel, & Schoenrock, 1981). Due to the contradicting nature of responses, the informed assent process should expand further than just yes or no questions and be phrased in a way relative to the client's mental capacity (Ramisch & Franklin, 2008). By providing informed consent and assent, the client and any participating third party (e.g., parents, legal guardian) understand the process as well as the risks the evaluation and report may entail.

Although state law may require legal guardians with decision-making authority to provide consent for clients with ID, psychologists should uphold a higher ethical standard by seeking assent from the client as well to continue services (Ramisch & Franklin, 2008). In a recent study by Horner-Johnson and Bailey (2013), most people diagnosed with ID were found to have the ability to consent or assent to psychological research processes in at least low-risk studies. More than half of the participants answered questions related to the consent process correctly and multiple sets of questions were piloted (Horner-Johnson & Bailey, 2013). While the evaluation and report consents are slightly different, the lit-

Mental Capacity Components

Understanding	Ability to understand treatment-related and diagnostic information
Appreciation	Ability to relate treatment information to one's own situation, in particular to nature of the diagnosis and the possibility that treatment could be beneficial
Reasoning	Ability to rationally evaluate treatment alternatives by comparing risks and benefits in light of potential consequences and their likely impact on everyday life
Choice	Ability to communicate a decision about treatment

Fig. 12.1 Components involved in determining mental capacity (Adapted from Grisso & Appelbaum, 1998)

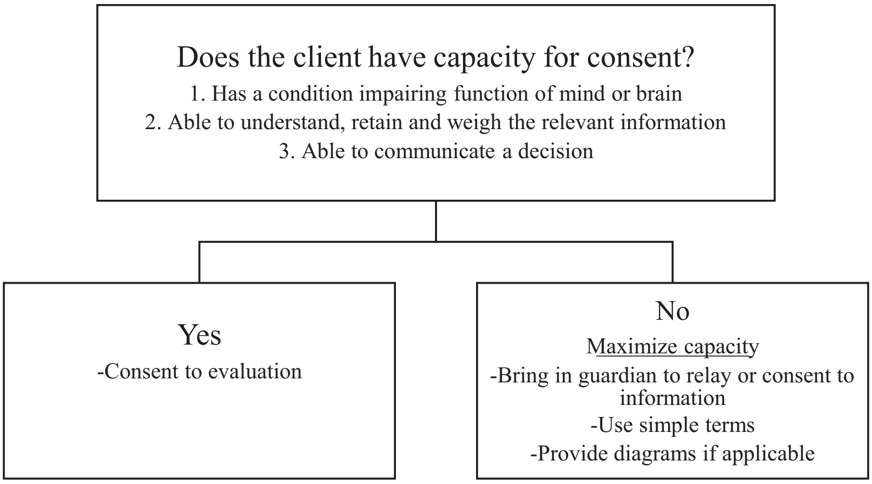


Fig. 12.2 Points to discuss when determining mental capacity (Adapted from Ministry of Ethics, 2014)

erature suggests that there is mental capacity to understand the overall process even with cognitive limitations. Determining mental capacity can be done by asking a variety of questions (see Fig. 12.2 adapted from the Ministry of Ethics, 2014).

There has been previous tension in the discussion of rights to those with intellectual disabilities and the caregiver roles. Moxley, Raider, and Cohen (1989) argued that family members are active agents in promoting the well-being of those with developmental disabilities. However, Qualls (1997) stated the need for balance between caregiver needs and the needs of the client to promote autonomy. The goals of the report should be the primary discussion when reviewing procedures at consent so that there is no confusion on what will be released in the report, particularly when the family and client are seeking services for intellectual disability.

Qualls (1997) stated that at any point in the life span of a client diagnosed with ID, families may be involved in providing support (e.g., emotional, financial, decision-making, direct care) for those who lack the full capacity to meet their own needs autonomously. The issue arises when families become fully enmeshed in the assessment process and lines begin to blur for the clients within the system (Ramisch & Franklin, 2008). Therefore, confidentiality should be discussed with caregivers by outlining what clinical aspects can or have to be reported. This discussion is especially important if a client only has legal guardianship in some, but not all, aspects of their life.

When producing a report for a client with a diagnosis of intellectual disability, there are multiple aspects to consider related to confidentiality, privacy, and privilege. The American Psychological Association Ethics Code states in 4.01 that psychologists have a primary obligation and take reasonable precautions to protect confidential information while recognizing the limits of confidentiality as regulated by law. The ethics code continues to say in 4.02 that psychologists should discuss confidentiality with the client including, to a reasonable extent, people who are legally incapable of giving informed consent and their legal representatives. If you are assessing a client with ID, the possible uses of the report, including who would have access to that information if consented and what could happen if there was possibly a subpoena issued for the report, must be discussed with the client and the legal guardian if possible. The discussion of confidentiality should occur before the start of the evaluation and well before any report is generated, typically during the informed consent process (American Psychological Association, 2017). Once consent and assent have been discussed, obtained, and sufficiently documented, it becomes important to consider the reason for the evaluation and subsequent report.

Rational Behind the Report

The purpose of writing a psychological assessment report is to create a comprehensive and integrated summarization and interpretation of the data generated during the assessment process, as well as place this developmental snapshot in the broader context of a client's varied background and developmental history. Moreover, the report should detail an assessment of the presenting problem and offer recommendations for the client. A report typically includes assessment data, behavioral observations, and relevant history combined using the informed judgments of an experienced clinician (Grills-Taquechel, Polifroni, & Fletcher, 2009). For clients with an intellectual disability, reports can be a way to communicate problems and challenges, and accommodations and recommendations to relieve difficulties occurring in their life. Research has shown that high-quality early intervention before the age of five leads to better long-term outcomes, making the early and accurate identification of ID and other developmental disabilities important (Guralnick, 2005). A concise and comprehensive report can help clinicians tailor interventions for those with ID, or even communicate concerns about a potential developmental issue before the condition is fully expressed for a subsequent examiner to consider. Therefore, an evaluator must report clearly and specifically as they can determine the trajectory of the particular client and the potential assistance they can gain access to.

Report writing addresses the client's presenting problem(s). Psychological reports allow a cohesive, linear timeline of the client's mental history, particularly for those with a lifelong diagnosis of ID. Reports are the ideal framework to parse out the concerns and tie them together with aspects of the client. Information should be gathered from a wide variety of sources in order to make comprehensive diagnostic determinations and treatment recommendations. As cited in Lichtenberger, Mather, Kaufman, and Kaufman (2004), Ownby (1997) described four purposes of an assessment report:

1. Answering the referral question as explicitly as possible
2. Providing the referral source with additional information when it is relevant
3. Creating a record of the assessment for future use
4. Recommending a specific course of action

Regardless of the evaluation purpose, the main goals of report writing are to generate hypotheses regarding a client's concerns, support or refute that hypothesis(es), and ultimately provide a conceptualization and recommendations for the client's specific difficulties. Reports that only focus on the data collected instead of a comprehensive interpretation of that data generate lackluster documents that disregard the intended purpose (Lichtenberger et al., 2004). O'Neill (1995) described three levels of clinician interpretations including the concrete level (e.g., only reporting scores), the mechanical level (e.g., reporting scores and the classification), and the individualized level (e.g., integrated reporting of scores). Reports written at the individualized level include the integration of history, behavior, and test results to detail the person instead of only focusing on the quantitative data. Reports interpreted at the individualized level are explanatory and the most useful (O'Neill, 1995).

In order to provide a comprehensive report, evaluations must be conducted in a concise, productive manner. Assessments for suspected ID can include an increased likelihood of test spoilage and noncompliance compared to other populations due to the nature of the existing problems. For instance, when administering an intelligence test, examiners must ensure that it is adequately assessing the client's abilities with regard to their current level of fine motor skills, vision/hearing issues, medical issues, and/or speech/language impairments. Professional judgment should be used to determine which intelligence test will correctly assess the client's cognitive abilities. All deficits should be mentioned in the report in relation to the client's performance on the intelligence testing (Reschly, Myers, & Hartel, 2002). More information on the types of intelligent tests available to assess for ID can be found in Chap. 22.

Critical Elements of a Report

There are various elements that are pertinent to a report in order to maintain a clear, comprehensive summary of the client. With a diagnosis of ID, omitted information could deny individuals needed services. The report should be based on a template that is organized in a logical manner.

Reports usually begin with a title so that readers of the report know what type of evaluation was conducted. Typically, the title is along the lines of "Psychological Evaluation" or "Psychoeducational Assessment." The title will depend purely on the type of evaluation given. Following the title, there should be identifying information which is usually presented in block format to avoid confusion as to whom the report belongs. The identifying information will often include the following:

- Name of examinee
- Date of birth
- Chronological age
- Grade (if testing a student) or occupation (if relevant)
- Name of educational or vocational placement
- Date (s) of testing
- Date of report
- Examiner's name
- Supervisor's name (if relevant)

Other information may be included in the identifying information section of the report depending on the purpose of the evaluation. The clinician may feel that other identifying information is necessary for the reader and can use clinical judgment to include this in the report. As long as the information is clear and concise, the details can be adjusted per the preferences of the assessor (Lichtenberger et al., 2004).

Reason for Referral

The reason for a referral is typically the next portion of the report. The primary concern of the client, his or her caregiver, and/or the family is outlined in this section, which sets the stage for the rest of the report. A person with ID may be referred for testing by his/her family, school districts, or a number of mental health agencies. These may include screening to determine eligibility for certain programs, a targeted assessment that focuses on a particular problem, a diagnostic assessment to assess strengths and weaknesses in an area, a counseling assessment with an emphasis on a client's ability to manage daily responsibilities, mandatory re-evaluations per guidelines, and/or progress evaluations to assess current interventions and progress (Sattler, 2001).

Understanding the referral question will ultimately determine the type of evaluation conducted and the specific recommendations for the client moving forward.

The reason for the referral section should ideally be no longer than one or two paragraphs. This section should include the name and positions (e.g., physician) of the referral source. The referral question may vary wildly. For example, referral questions may be in regard to whether the client is able to live independently, if the client can transfer to a group home/supervised location, or what academic environment and curriculum would best meet the client's strengths and abilities. After stating the problem succinctly, providing brief anecdotes or examples may help to get the point of referral across to the reader. Most referrals will concern the client's academic, adaptive, linguistic, cognitive, behavioral, or social development (Lichtenberger et al., 2004).

Background Information

Background information may be the next section of the report and is intended to provide a comprehensive summary of the client, while detailing overarching concerns. Collecting information from a variety of sources will aid in pinpointing the concerns and contributions from different perspectives. The information from each informant should be relevant to their first-hand knowledge (e.g., a physician giving medical or developmental history, teacher giving academic history). Information should come from those who interact with the client on a regular, if not daily, basis such as family members, caretakers, and/or group home workers. Background information may include history of development from medical, familial, educational, behavioral, and social perspectives. However, if the background information is not relevant to the referral concern, it is likely not needed within the report. Only include information that pertains to the primary concern and the test data interpretations, or that is necessary to clarify outstanding developmental concerns (e.g., a conflicting and confusing diagnostic history). Information that is personal and not pertaining to the purpose of the evaluation should also be left out of the report (Kamphaus & Frick, 1996).

The background section should be organized from topic to topic in a logical progression, preferably in chronological order. By keeping the information in order, the duration and the intensity of the concerns become salient. Each paragraph in the background section should be a separate topic (e.g., one paragraph on medical history, one on development, one on adaptive behavior). For reports involving suspected ID, developmental history is a critical topic to cover in the background information section and may include possible genetic or developmental diagnoses, relevant family history, as well as the mother's pregnancy, delivery, after birth complications, and developmental milestones throughout the client's life. Client medical and developmental history for an ID assessment can often be a muddled, error-ridden cluster of information that should be clarified in the report. All of the developmental and medical milestones and delays should be detailed in chronological order (Lichtenberger et al., 2004).

Given the complexity of assessing clients with suspected ID and the potential for different individuals and environments to elicit different behaviors and symptoms, multiple informants should contribute to the information provided in the report (Handen, 2007). Because there may be a limited capacity in those with severe ID, evaluating the symptoms of ID may not be possible beyond what is observed and reported upon by others. Multiple informants may include parents/guardians, teachers, aides, home workers, and/or social workers. Since most informants are seeing symptoms in different environments and/or perspectives, information is likely to conflict. The level of discrepancy should be discussed in the report with a hypothesis provided for why discrepancies or congruences may exist (e.g., behaviors only presenting in a certain setting or those that present across settings). Frequently, a problem behavior described by one informant will not be described by another informant, as agreement has been reported to be about 68.1% for some symptoms (Handen, Feldman, & Honigman, 1987). Such findings suggest that clinicians gather additional information when two or more informants disagree to determine the clinical utility of reported information (Handen, 2007). If there is any discrepant information among the sources, the examiner should try to account for the discrepancy with a logical explanation, which will provide a more balanced picture of the client.

Behavioral Observations

Behaviors observed during assessment as well as other settings should be included in the behavioral observations section of the report. Assessment settings are controlled to maximize attention and performance while one-on-one interactions occur. Because the setting is meant to optimize the performance of the client, the behaviors may not be congruent with behaviors displayed in other, albeit similar, settings (e.g., school). If the behavior in the session seems to generalize to multiple environments, it is best practice to confirm the behavior with other sources from various settings. Compiling a list of the behaviors observed in session can help to keep this section of the report organized. Interpreting behaviors in the report can help the reader to better understand the behaviors by combining hypotheses and observed behaviors into integrated statements. For instance, the evaluator must interpret and define behaviors in the context of the client conceptualization; if clinical symptoms of attention-deficit/hyperactivity disorder (ADHD) are reported, but a diagnosis of ADHD is not given, the evaluator must provide the interpretation that led to that decision (e.g., inattention symptoms were not reported to be clinically significant beyond what would be expected given the client's intellectual abilities). Reports should not consist of a string of behaviors observed in the session without interpretation nor should a list of hypotheses be presented without giving examples of behaviors (Lichtenberger et al., 2004).

It is important to observe and elicit adaptive behaviors for clients referred for ID. There are specific adaptive behavior scales and interviews that can be used to reflect an individual's ability to independently meet the needs and social demands on the environment. This behavior is one criteria required to make a diagnosis of ID. The operational definition of "limitations of adaptive behavior" from the AAIDD is to be at least two standard deviations from the mean for one of the three adaptive behaviors or for the overall score. Observing this behavior in session will give a general insight on the level of ID and functional impairment of the client.

Adaptive behaviors for an individual presenting with possible ID should be structured around the three domains of conceptual, social, and practical skills so that the report can parse out the symptoms for the diagnosis or diagnostic trends. Figure 12.3 explains the three domains of adaptive behavior adapted from the *Diagnostic and Statistical Manual of Mental Disorders Fifth Edition* (2013) under intellectual disability.

The first is the conceptual domain including skills in language, reading, writing, math, reasoning, knowledge, and memory (Parekh, 2017). These items can be tested using a variety of intelligent quotient (IQ) tests where a total score is derived from a standardized test to assess human intelligence. Achievement tests can also assess the conceptual domain which includes the application of the specific domain aspects. The conceptual domain can also be discussed in the test results section of the report. When reporting IQ and achievement scores, major discrepancies should be noted. The bigger the discrepancy as well as the lower the IQ score can help determine severity level (mild, moderate, severe, profound) and, in turn, the level of accommodations required.

The second domain is the social domain which includes empathy, social judgment, interpersonal communication skills, the ability to make and retain friendships, and similar capacities (Parekh, 2017). The social domain can appear in the report in a number of places, including behavioral observations, assessment measures, and a variety of informants (e.g., client, guardian, teacher, social worker). Depending on the severity level of the client, details on the social domain would differ in the report from something less problem-

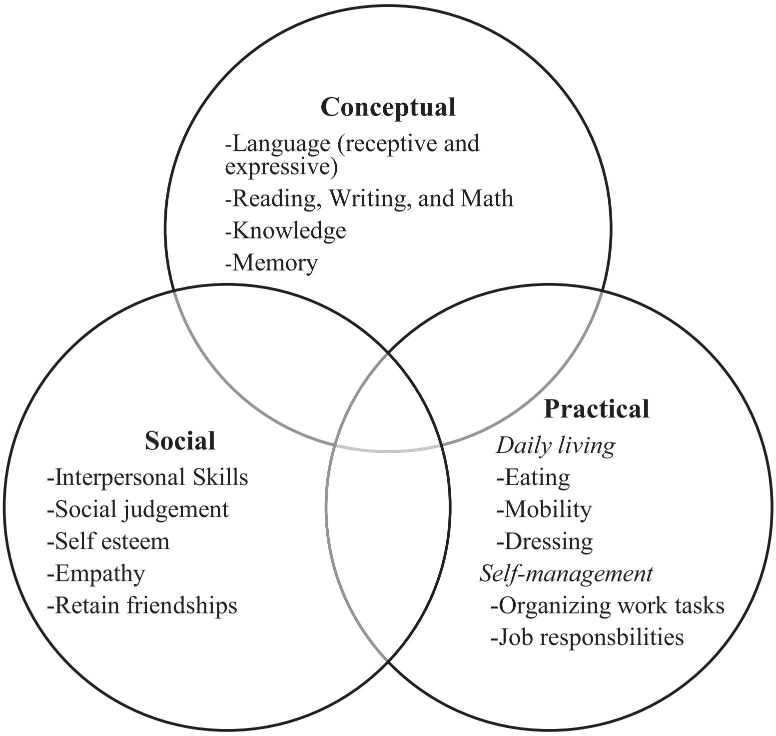


Fig. 12.3 The three domains of adaptive behavior that should be assessed (Adapted from APA, 2013)

atic (e.g., client being immature in social interactions) to more extreme (e.g., limited understanding of communication in speech and gesture).

The last domain is the practical domain centering on self-management in areas such as personal care, job responsibilities, money management, recreation, and organizing school or work tasks (Parekh, 2017). All three domains of adaptive functioning should be considered. An individual diagnosed with ID with a low IQ, but high adaptive functioning, would likely be considered mild severity, as they have the ability to care for themselves in a functional way. If adaptive functioning is not a strength (e.g., cannot feed oneself, get dressed, shower), the designation would likely be more severe ID. When including this domain in the report, an emphasis on the client's ability should be stressed in order to defend the diagnosis and severity level. Adaptive functioning can be found in its own section, under behavioral observations, or even under informant interviews. Placement of this information in a report depends on how the report is being organized (e.g., chronologically, etc.), which will be discussed later in this chapter. Further information regarding adaptive functioning testing can be found in Chap. 23.

Physical appearance can also be included in the behavioral observation section of the report, using a sentence or two about how the client appeared during the session (e.g., dressed appropriately, disheveled). Level of grooming and hygiene can be indicative of adaptive behavior and/or the amount of help the client needs (or currently receives). Client communication should be included in the report (e.g., how well the client engaged, whether they were nonverbal, and/or body language). If probed for response to failure or response to feedback during the session, reactions can be included in the observed behavior. Mood and temperament should also be included. People have varying temperaments in response to environment, styles of learning, and strengths and vulnerabilities (Brooks & Goldstein, 2001). If any factors were influencing the behavior in the session, it should be included in the report (e.g., the client was tired or hungry). Some writers include a statement of validity and reliability at the end of this session based on the information observed (e.g., the observed behavior is reliable since it was observed in all sessions).

Test Results and Interpretation

The test results and interpretation section of the report contain the tests administered, the data from the assessments, and interpretation of the data. This section of the report accounts for all of the data throughout the sessions: background history, behavioral observation, and test results. Choosing a basic format keeps the report organized in a logical manner. Format is typically found in three ways: domain by domain, ability by ability, or test by test. Whichever format is chosen should continue through the results section of the report. All three formats should include qualitative and quantitative data to inform the reader of the main findings (Lichtenberger et al., 2004).

The use of subheadings can help to keep this section organized and should move from broad to specific information about the client. For example, with intelligence testing, global IQ scores are reported first and then broken down into subtest scores and the respective strength and weaknesses. The main conclusions of the test interpretations can be in the first or last paragraph depending on the preference of having succeeding paragraphs describe why or having the paragraphs add up. The results should also move from standardized test data to information collected during observations and background history. Information can also be organized by themes, either with performance or with abilities in cognitive, linguistic, academic, social, or emotional domain (Lichtenberger et al., 2004).

The findings of the assessment should be considered to see how the data fit together. Lichtenberger et al. (2004) suggested several questions that can be asked to decide how the data should be incorporated:

1. How are the findings consistent?
2. What patterns of data support the common theme?
3. Which data are contradictory?
4. How can you explain the contradiction?

By looking at the consistencies and contradictions between assessment results and observations, a comprehensive picture can be formed of the individual ultimately making interpretations and recommendations easier (Lichtenberger et al., 2004). Interpreting should include examining the data, focusing on the person, describing what the person could and could not do, and reporting scores so they are concise and clear. Diagnoses should be included in this section of the report using diagnostic codes. When writing diagnostic impressions, data should be described to support the conclusion. Diagnosis can be the most important part of the report and should be considered carefully (Wolber & Carne, 2002).

Summary and Recommendations

Often (and unfortunately), the summary and recommendations sections of the report are the only sections read, as it recaps the valuable points of the report and guides the reader to services relevant to the diagnoses or concerns. Each section previously discussed in the report must be touched on in the summary; however, no new information should be brought forward in this portion of the report. Key points from the reason for referral, background information, behavioral observations, clinical impressions, and results sections should be included in the summary. The same wording should not be used so that the reader does not feel they are just re-reading the report. Instead, the information should be rephrased and shortened to give a quick overview. By highlighting the main points of the report in the summary, the reader can then lead into the recommendations knowing all the pertinent information (Lichtenberger et al., 2004).

The recommendations are based on the initial referral question, as well as the data and information collected during the assessment. For example, if the referral question focused on motor skills, the recommendation section of the report would focus on providing services or interventions to help improve motor skills, with attention paid to age and/or developmental level, degree of skill deficit, and intensity of impairment. Prior interventions should be considered when providing recommendations. For instance, if a client has a history of not responding to a particular intervention (with reason to believe it was well implemented), the clinician should explore alternative, empirically supported interventions. Recommendations should be informative and comprehensive, especially since the diagnosis of ID is for life (Lichtenberger et al., 2004). In formulating specific recommendations for a person with ID, Sattler (1998) suggests considering:

1. Do the results indicate a diagnosis of intellectual disability?
2. Is the person eligible for special services?
3. What are the least restrictive options for programming?
4. If mainstreaming or inclusion is possible, what additional supports will be needed?
5. What are the person's social skills strengths and weaknesses?
6. What skills will be needed for productive employment?
7. What types of supports will he or she require to live independently? Or what type of living arrangement will be needed?

The levels of required support needed for an individual diagnosed with ID should be discussed in the summary and recommendations sections as well. The AAIDD developed a classification system based on the amount of supports needed by the client (Schalock et al., 2010). The four levels of support include intermittent, limited, extensive, and pervasive. Depending on the functioning of the individual and the severity level of the ID diagnosis, the support may be as simple as brief targeted interventions such as social support (intermittent) or as extensive as continuous, daily life support (pervasive) in all aspects of their life (Carr & O'Reilly, 2016). While intermittent support is brief, limited support is more constant but only in certain situations (e.g., entering a new environment). Extensive support is a continuous support, but not life dependent, such as pervasive support with the hope that the individual will become more independent over time (Sigafoos, Lancioni, Singh, & O'Reilly, 2017). Any changes in support needed should also be discussed. While ID is a lifelong condition, the amount and types of support needed may change over time. When explaining the changes, clinicians should be careful to be sensitive to hopeful questions (e.g., eventually living independently or becoming more functionally independent) and would be well advised to temper feedback with an individual client's strengths as well as weaknesses. The amount of the support may wax and wane, but the severity of the diagnosis will not.

Once the data is gathered, the clinician should determine what is beneficial to be included in the report to address the main concerns. The major findings should be included as well as diagnostic trends that are applicable to the area of concern (e.g., does the client have developmental delays across all areas of life?). Background information should be consolidated to relevant items instead of including every word spoken in session. For people referred for possible ID, it is important to thoroughly examine adaptive functioning, achievement levels, skill levels (e.g., motor skills, language skills), behavioral concerns, functions of behavior, and possible comorbid disorders. By analyzing the data, the report will be able to integrate assessment results with information about the client. Crnic (1988) discusses a list of points to consider when determining what information to integrate in the report:

1. Is there significant test scatter across skills areas?
2. How do the results compare with previous assessment?
3. Did changes in the structure of testing (e.g., testing limits or allowing the use of alternating strategies) affect performance?
4. What behavioral factors were problems and what factors were strengths? Did they affect the assessment outcome?
5. Did performance or behavior vary across settings?

Are historical variables (e.g., developmental, medical, behavior) relevant to the client's performance?

Intended Audience

The intended audience of a report will vary depending on a myriad of factors (setting, purpose, etc.), and may not only be the parent/guardian. While the primary referral question will function as the framework of the report, the language used will depend primarily on who the report is written for. Reports written in a concise manner are more likely to be read in full. Writing a report with technical jargon will lead to confusion particularly for the family of the client. For those seeking medical help, especially those being assessed for possible ID, the report may be geared toward physicians who are more interested in reading about the technical or neurological underpinnings. The report may also be written for another psychologist, for example, someone who recommended the evaluation. In a case like this, including more data may be helpful; however, care should be taken as it is typically not good practice to include raw data in the actual report. An authorization to release information should be used in order to deliver data to other psychologists. Data are ordinarily not provided to non-psychologists or those inexperienced in their interpretation.

For reports meant to be used in school settings, tailoring the report to school interventions is ideal. It is important to note that schools offer different interventions and not all schools will offer the same services. Therefore, the examiner should enquire about the applicable resources available to the client to ensure the recommendations made are feasible. Different audiences will evaluate reports in wildly different manners and should be written as such.

One should also be careful of the language used to report a diagnosis of ID. Clinicians should use "people first" language (i.e., a person with an intellectual disability instead of an intellectually disabled person), as an individual's diagnosis does not define them. This can be even more significant to a family who is just learning about the diagnosis of ID. It is best practice to be sensitive to the reaction and feelings of a client and their family especially regarding diagnoses. Using "people first" language can also teach the client and family that while there might be developmental delays, there are strengths and positive attributes that make up the person. Because of the stigma surrounding ID (and unfortunately most other mental health diagnoses as well), correct terminology should be used. Intellectual disability was previously called "mental retardation," which has since been deemed offensive. Handen (2007) stated that this should be explained since the family may come across this label among other agencies. Some families may have never come across the labels "intellectual disability" or "mental retardation" even after having assessments with similar findings; however, the terms should be explained (Handen, 2007). Careful consideration should be taken when discussing the diagnosis.

The feedback session is an ideal opportunity to explain the scope and implications of the diagnosis of ID to the client and/or family, especially if the report is written for a different intended audience. The success of a feedback session can be based on how well the clinician probed for the initial referral question. Being on the same page about concerns can promote a productive discussion about the findings and the implications. While it is relevant to explore the scope of a diagnosis of ID, the strengths of the client should be stressed, leaving the session with a positive note. Intervention areas and services available should also be discussed to ward off any feelings of helplessness or stress. The client or advocate needs to understand that ID is a lifelong diagnosis and not something that will go away or be fixed; however, there are multiple resources available. In a case where a person with ID does not have family or an advocate there to comprehend the scope of the diagnosis, the discovery should be explained using the appropriate language if appropriate. The intellectual level of discussion should be based on the level of functioning of the client so that they leave comprehending what ID means and what resources can be of help.

In some ways, one of the most difficult feedbacks to conduct are with the family, advocates, and/or an individual with an ID. It is important that the client and/or their advocate understand what a diagnosis of ID means. One way to achieve this goal is to discuss the report in detail. The results found in the assessment can be discussed by going over strengths and weaknesses in certain areas such as the subscales of the IQ assessment or the adaptive behavior assessment. Providing figures or visual aids to understand the test scores (e.g., bell curves) can also help get the point across in less technical terms. Bell curves are based on statistical analysis and cannot perfectly fit every demographic. The bell curve figure should be used as a general illustration to make explanations more comprehensible but is not meant to be the actual data of the results section or representative of all populations.

By dividing up the information into tangible data, the individual can better understand what equates delays. This also provides the opportunity to discuss what qualifies someone for accommodations or programs that can benefit the client with ID. Sometimes the news of a diagnosis of ID can leave families feeling overwhelmed and unable to comprehend the implications during the feedback session. By providing them with a written report and feedback, family members have another chance to react to the news and possibly contact the clinician later with additional questions. Clinicians should always offer another chance to discuss findings or answer any questions to ward off confusion (Handen, 2007).

Confidentiality

Ensuring client confidentiality should be a priority when creating a psychoeducational report. The rights of those with ID can change quite drastically with severity levels. Whether the client has the legal or mental capacity to uphold their own legal standings does not determine confidentiality; however, it can determine who can be provided the confidential information. Under the Health Insurance Portability and Accountability Act and various state and federal laws (seek your own counsel for advice and applicable law), private health care information remains confidential, and privacy laws are to be upheld by health providers (and their business associates) including clinicians conducting psychological evaluations.

Shared information should be kept to a minimum by only providing the minimal details that is relevant to the purpose of the disclosure (Office of Civil Rights, 2017). For example, if a client was seeking services for ID through a government agency, providing the diagnosis would be relevant, but providing the client's religious affiliation would not be relevant. Due to the possibility of the client's consent to release the report to other agencies, the report should be written with only applicable details pertaining to or defending the diagnosis of ID or any other comorbid disorder. Psychologists should only include in written and oral reports information pertinent to the purpose of the communication (American Psychological Association, 2017).

Providing too many details on the families' life can also be a breach in confidentiality. The report is centered on the client, which should include family history; however, the report should not reveal confidential information on specific family members. For example, the report should include family history of previous psychological diagnoses but should likely not state, "Her sister, Jane, was diagnosed with schizophrenia." By divulging details of someone else's diagnosis, confidentiality has been broken for a person who is not even a client.

An authorization form should be used to obtain or release information to and from others for the sole purposes of receiving information to write the report or release the report to other agencies for services. The authorization form is a specific consent form that details what information is being released or obtained and to who. One of the main reasons a client with ID may seek an evaluation may be to provide various agencies a legal document detailing their diagnosis. The report should be written so that there is no violation of privacy when the report is released (Office of Civil Rights, 2017). Confidentiality and privacy laws are mostly governed at the state level. Any confusion about content of the report breaching confidentiality or when or to whom information should be shared should be directed to appropriate legal counsel in the state under which the evaluation is being conducted.

Safeguarding the Data and the Report

Once the evaluation is complete and the report is generated, the priority shifts to safeguarding the data. Confidentiality should not end once the report is finished and the client is no longer seen. Confidentiality of data and reports not only covers how records are kept but also who owns and has access to data, where the data are stored, how the data are protected, how long the data should be kept, and when the data can be destroyed (Cooper, 2016).

Release of raw data and psychological test materials affects the reliability and validity of the test procedures and results and also often has overlooked entanglements with copyright violations (e.g., making copies of protocols). Currently, federal courts and approximately 20 states promote the protection of psychological tests while some states also enact a nondisclosure privilege and duty to safeguard test materials and raw data from disclosure (Kaufmann, 2009). With the current shift from paper to electronic sources of record keeping comes new guidelines. HIPAA guidelines state that the health provider must maintain reasonable and appropriate administrative, technical, and physical safeguards when storing private health information, which would include raw data, test materials, and reports (HIPAA, 1996). The Department of Health and Human Services (HHS) provided new security standards in 2003, stating that psychologists who transmit private health information electronically take security precautions to prevent a breach in data and conduct risk analyses (for details contact your legal representation).

In 2007, the American Psychological Association produced record-keeping guidelines (see https://www.apa.org/practice/guidelines/record-keeping.aspx). Guideline 3 details confidentiality, stating that practitioners should be aware of the current regulatory and legal requirements regarding client records. Guideline 6 explains the security measures that should be established for practitioners with client records. If client paper records are created, they must be stored in a cabinet with a lock and key. The same can be said with electronic records which should also be properly stored. Records should be stored in a location where they are protected from damage (e.g., fire, water, mold, insects). Guideline 9 goes in detail on how to store electronic records stating that practitioners should be careful with the use of e-mail and other communication tools due to the possibility of breach of confidentiality. Electronic records should be protected via passwords, firewalls, data encryption, and authentication (Lustgarten, 2015). Standard 6.02 of the code of ethics establishes that in any form, record creation and storage must be confidential. Moreover, if a practitioner must use shared information, the minimum amount of information needed should only be used (APA, 2017). The completed report should be stored under lock and key to prevent tampering or stealing of sensitive information and should only be distributed under the client's authorization. Even then, only pertinent information should be shared with authorized agencies.

There are a number of extra steps that can be taken to further protect confidential records. Lee (2013) suggested creating a threat model to asses each client's associated risk. The *Electronic Frontier Foundation* (2014) suggested asking five questions in developing a threat model:

1. What do you want to protect?
2. Who do you want to protect it from?
3. How likely is it that you will need to protect it?
4. How bad are the consequences if you fail?
5. How much trouble are you willing to go through to try and prevent those consequences?

Stronger protections can also be applied by encrypting all electronic files so that getting to the client's record entails multiple encryptions. For example, a file storing device can be encrypted as well as the documents contained on the device. Storing the information on a file system that can be locked in either a lockbox or cabinet can further protect client information. There are ethical guidelines that must be followed if a breach in security for client records occurs. These guidelines overall are constantly evolving and so consultation with competent legal counsel is a must.

The APA Ethics Code states that informed consent must include the practices of storing and protecting client data and reports. Clients and legal guardians should be notified of how the information is stored (e.g., paper or electronic) and steps taken to protect confidentiality (Devereaux & Gottlieb, 2012). By not informing clients before services begin, practitioners are left with the possibility of monetary and disciplinary consequences (Lustgarten, 2015). Clients should also be notified during informed consent about retention of records. APA Ethics Code suggests retaining full records until 7 years after the last date of service for adults or until 3 years after a minor reaches the age of majority, whichever is later. When conducting evaluations for clients with a diagnosis of ID, the retention time of reports should be heavily weighed. Since this disorder is a lifelong diagnosis, the report may be needed for a longer period of time than that of a different diagnosis.

Client records of an assessment for ID include sensitive information discovered in interviews, behavioral observations, and test administration. All pieces of client information should be safeguarded by the practitioner to protect the client from harm. The APA Ethics Code and relevant state laws should be the guidelines followed when storing and protecting client records, and any confusion or questions should be resolved with knowledgeable legal counsel.

Conclusion

While writing a psychological report may seem like an overwhelming task, staying organized and concise can be the key to accomplishing the job. The importance of a psychological report with an ID diagnosis can be monumental considering the disorder is a lifelong disability. Establishing the groundwork during assessment to ensure the relevant information is covered removes the doubt of the diagnosis in the report. A detailed report indicating the diagnostic criteria the client meets (or does not meet) is valuable to the client and their family in getting services and accommodations. Concrete, feasible recommendations, some of which that can be started while waiting for more intensive services, are essential to the report. Writing a valid, comprehensive, integrated report that answers the initial referral question while simultaneously addressing any additional problems (e.g., comorbid disorders) will have the most positive impact on the client and their family.

References

American Psychiatric Association. (2013). *Diagnostic and statistical manual of mental disorders* (5th ed.). Arlington, VA: American Psychiatric Publishing.

American Psychological Association. (2017). Ethical principles of psychologists and code of conduct (2002, Amended June 1, 2010 and January 1, 2017). Retrieved from http://www.apa.org/ethics/code/index.aspx

Brooks, R., & Goldstein, S. (2001). *Raising resilient children: Fostering strength, hope, and optimism in your child*. Lincolnwood, IL: Contemporary Books.

Carr, A., & O'Reilly, G. (2016). Diagnosis, classification, and epidemiology. In A. Carr, C. Linehan, G. O'Reilly, P. Noonan Walsh, & J. McEvoy (Eds.), *The handbook of intellectual disability and clinical psychology practice* (pp. 3–44). London: Routledge.

Cicciola, E. (2008). Alfred Binet e le prime "misure" dell'intelligenza (1905–1908) [Alfred Binet and the first "measures" of intelligence (1905–1908)]. *Physis, Rivista internazionale di storia della scienza, 45*, 165–203.

Cicciola, E., Foschi, R., & Lombardo, G. P. (2014). Making up intelligence scales: De Sanctis's and Binet's tests, 1905 and after. *History of Psychology, 17*(3), 223–236.

Cooper, H. (2016). Data collection and handling of data sets. In *Ethical choices in research: Managing data, writing reports, and publishing results in the social sciences* (pp. 51–70). Washington, DC: American Psychological Association.

Crnic, K. A. (1988). Mental retardation. In E. J. Mash & G. Terdal (Eds.), *Behavioral assessment of childhood disorders* (2nd ed., pp. 317–354). New York, NY: Guilford Press.

Department of Health and Human Services. (2003, February 20). Health insurance reform: Security standards; final rule. *Federal Register, 68*, 8334–8381.

Devereaux, R. L., & Gottlieb, M. C. (2012). Record keeping in the cloud: Ethical considerations. *Professional Psychology: Research and Practice, 43*, 627–632.

Devi, N. (2013). Supported decision-making and personal autonomy for persons with intellectual disabilities: Article 12 of the UN convention on the rights of persons with disabilities. *The Journal of Law, Medicine & Ethics, 41*, 792–806.

Electronic Frontier Foundation. (2014). *Keeping your data safe*. Retrieved from https://ssd.eff.org/en/module/keeping-your-data-safe

Fesko, S. L., Hall, A. C., Quinlan, J., & Jockell, C. (2012). Active aging for individuals with intellectual disability: Meaningful community participation through employment, retirement, service, and volunteerism. *American Journal on Intellectual and Developmental Disabilities, 117*, 497–508.

Grills-Taquechel, A. E., Polifroni, R., & Fletcher, J. M. (2009). Interview and report writing. In J. L. Matson (Ed.), *Assessing childhood psychopathology and developmental disabilities* (pp. 55–88). New York, NY: Springer.

Grisso, T., & Appelbaum, P. S. (1998). *Assessing competence to consent to treatment: A guide for physicians and other health professionals*. New York, NY: Oxford University Press.

Guralnick, M. J. (2005). Early intervention for children with intellectual disabilities: Current knowledge and future prospects. *Journal of Applied Research in Intellectual Disabilities, 18*, 313–324.

Handen, B. L. (2007). Intellectual disability (mental retardation). In E. J. Mash & R. A. Barkley (Eds.), *Assessment of childhood disorders* (pp. 551–597). New York, NY: Guilford.

Handen, B. L., Feldman, R. S., & Honigman, A. (1987). Comparison of parent and teacher assessments of developmentally delayed children's behavior. *Exceptional Children, 54*, 137–144.

Horner-Johnson, W., & Bailey, D. (2013). Assessing understanding and obtaining consent from adults with intellectual disabilities for a health promotion study. *Journal of Policy and Practice in Intellectual Disability, 10*(3), 1–10.

Kamphaus, R. W., & Frick, P. J. (1996). *Clinical assessment of child and adolescent personality and behavior*. Needham Heights, MA: Allyn and Bacon.

Kaufmann, P. M. (2009). Protecting raw data and psychological tests from wrongful disclosure: A primer on the law and other persuasive strategies. *The Clinical Neuropsychologist, 23*(7), 1130–1159.

Kohn, N. A., Blumenthal, J. A., & Campbell, A. T. (2013). Supported decision-making: A viable alternative to guardianship? *Penn State Law Review, 117*(4), 1111.

Larson, S. A., Lakin, K. C., Anderson, L., Kwak Lee, N., Lee, J. H., & Anderson, D. (2001). Prevalence of mental retardation and developmental disabilities: Estimates from the 1994/1995 National Health Interview Survey Disability Supplements. *American Journal on Mental Retardation, 106*, 231–252.

Lee, M. (2013). Encryption works: How to protect your privacy in the age of NSA surveillance. Retrieved from https://freedom.press/sites/default/files/encryption_works.pdf

Lichtenberger, E. O., Mather, N., Kaufman, N. L., & Kaufman, A. S. (2004). *Essentials of assessment report writing*. Hoboken, NJ: Wiley.

Lustgarten, S. D. (2015). Emerging ethical threats to client privacy in cloud communication and data storage. *Professional Psychology: Research and Practice, 46*(3), 154–160.

Macmillan, D. L., Siperstein, G. N., & Gresham, F. M. (1996). A challenge to the viability of mild mental retardation as a diagnostic category. *Exceptional Children, 62*, 356–371.

McSherry, B. (2015). Decision-making, legal capacity and neuroscience: Implications for mental health laws. *Laws, 4*, 125–138.

Ministry of Ethics (2014). Consent and Confidentiality. Retrieved from http://ministryofethics.co.uk/?p=6

Moxley, D., Raider, M., & Cohen, S. (1989). Specifying and facilitating family involvement in services to persons with developmental disabilities. *Child and Adolescent Social Work, 6*, 301–312.

O'Neill, A. M. (1995). *Clinical inference: How to draw meaningful conclusions from psychological tests*. Brandon, VT: Clinical Psychology Publishing.

Office of Civil Rights. (2017). HIPPA for Professionals. Retrieved from https://www.hhs.gov/hipaa/for-professionals/index.html

Ownby, R. L. (1997). *Psychological reports: A guide to report writing in professional psychology* (3rd ed.). New York: Wiley.

Parekh, R. (2017). *What is intellectual disability?* Washington, DC: American Psychiatric Association. Retrieved from https://www.psychiatry.org/File%20Library/.../APA_DSM-5-Intellectual-Disability.pdf

Public Law 104-191— 104th Congress. Health Insurance Portability and Accountability Act of 1996.

Public Law 106–402 — 106th Congress. Developmental Disabilities Assistance and Bill of Rights Act of 2000.

Qualls, S. H. (1997). Transitions in autonomy: The essential caregiving challenge an essay for practitioners. *Family Relations, 46*(1), 41–45.

Quinn, G. (2011). *Rethinking personhood: New directions in legal capacity law and policy or how to put the 'shift' back into paradigm shift*. Galway, Ireland: National University of Ireland, Center for Disability Law & Policy. Retrieved from http://citizenship.sites.olt.ubc.ca/files/2014/07/Gerard_Quinn_s_Keynote_-_April_29__2011.pdf

Ramisch, J. L., & Franklin, D. (2008). Families with a member with mental retardation and the ethical implications of therapeutic treatment by marriage and family therapists. *The American Journal of Family Therapy, 36*, 312–322.

Reschly, D. J., Myers, T. G., & Hartel, C. R. (Eds.). (2002). *Mental retardation: Determining eligibility for social security benefits*. Washington, D.C.: National Academics Press.

Sattler, J. M. (1998). Clinical and forensic interviewing of children and families: Guidelines for the mental health, education, pediatric, and child maltreatment fields. La Mesa, CA, US: Jerome M Sattler Publisher.

Sattler, J. M. (2001). *Assessment of children: Cognitive applications* (4th ed.). San Diego, CA: Author.

Schalock, R. L., Buntinx, W. H. E., Borthwick-Duffy, S., Bradley, V., Craig, E. M., Coulter, D. L., … Yeager, M. H. (2010). *Intellectual disability: Definition,* *classification, and system of supports* (11th ed.). Washington, DC: American Association on Intellectual and Developmental Disabilities.

Schalock, R. L., & Luckasson, R. (2013). *Clinical judgment* (2nd ed.). Washington, DC: American Association on Intellectual and Developmental Disabilities.

Sigafoos, J., Lancioni, G. E., Singh, N. N., & O'Reilly, M. F. (2017). Intellectual disability and social skills. In J. L. Matson (Ed.), *Handbook of social behavior and skills in children* (pp. 249–271). New York, NY: Springer.

Sigelman, C. K., Budd, E. C., Spanhel, C. L., & Schoenrock, C. J. (1981). When in doubt, say yes: Acquiescence in interviews with mentally retarded persons. *Mental Retardation, 19*, 53–58.

Tasse, M. J. (2016, September). Defining intellectual disability: Finally we all agree…almost. *American Psychological Association Newsletter.* Retrieved from: http://www.apa.org/pi/disability/resources/publications/newsletter/2016/09/intellectual-disability.aspx

Wolber, G. J., & Carne, W. F. (2002). *Writing psychological reports: A guide for clinicians* (2nd ed.). Sarasota, FL: Professional Resource Press.

13

Agencies and Professions in the Provision of Care

Karen L. Nankervis and Maria V. Vassos

Background

With the move away from institutionalisation to community participation, inclusion, human rights, and person-centred supports, the need for systems and services to meet the needs and aspirations of people with intellectual disability (ID) and their families is essential. However, supporting people with ID who are described as having complex needs is often regarded as challenging to systems, services and individuals, leading to difficulties in having those needs met effectively. Supporting people with ID and complex needs requires multiple safeguards as well as service coordination between multiple agencies and service systems. This chapter provides an overview of the issues associated with complex needs for people with ID, and best practices in supporting them.

K. L. Nankervis (✉)
University of Queensland, St Lucia, QLD, Australia
e-mail: k.nankervis@uq.edu.au

M. V. Vassos
University of Queensland, St Lucia, QLD, Australia

Endeavour Foundation, Cannon Hill, QLD, Australia

What Is Meant by Complex Needs?

There is inconsistent understanding of what is meant by "complex needs" and it is often used to describe people with ID who have additional needs (Cooper & Ward, 2011) or multiple needs that span issues, services and systems (Rankin & Regan, 2004). *Complex needs* is a term described by Rankin and Regan (2004) as:

> *"A framework for understanding multiple, interlocking needs that span health and social issues. People with complex needs may have to negotiate a number of issues in their life, for example learning disability, mental health problems, substance abuse. They may also be living in deprived circumstances and lack access to suitable housing or meaningful daily activity. As this framework suggests, there is no generic complex needs case. Each individual with complex needs has a unique interaction between their health and social needs and requires a personalised response from services."* (p.i)

Accordingly, there is no universal understanding or definition of the term complex needs. For instance, a person may have complex *disability support* needs that are associated with impairment, such as having profound intellectual and multiple disabilities. In other cases, a person may have complex needs that are associated with having needs that span services and systems, not just disability services. As proposed by Rankin and Regan (2004) complexity of need can involve multiple and interlocking problems that have

J. L. Matson (ed.), *Handbook of Intellectual Disabilities*, Autism and Child Psychopathology Series, https://doi.org/10.1007/978-3-030-20843-1_13

depth (severity of need) and/or *breadth* (range of need). Thus, complexity of need can arise from: (1) the severity of impairment and disability for the individual and/or (2) needs that require system and sector coordination due to multiple needs (such as co-morbidity), circumstances (abuse, neglect, trauma), or complex situations (such as inadequate appropriate accommodation). Table 13.1 presents some examples of the depth and breadth of complex needs for people with ID.

However having breadth or depth of need alone may not be complex. While some people with ID have severe and pervasive disability support needs, or multiple needs that span organisational boundaries, where adequate, appropriate and coordinated services and supports are readily available meeting those needs may not be complex. A person's support needs can also become complex unpredictably especially where crisis arises such as carers' or services unexpectedly no longer able to provide care or where challenging behaviour results in placement breakdown, leading to potential homelessness or inappropriate accommodation. Thus, complexity of need is associated with any difficulties in having those needs met. Such difficulties include the timely availability of required supports and services or systems that are able to work together in a responsive and integrated fashion.

Table 13.1 Examples of complex needs associated with severity of need (depth) or range of need (breadth)

Severity (depth)	Range (breadth)
Intense and pervasive level of support needs associated with profound intellectual and multiple disabilities	Residing in inappropriate accommodation such as young people in residential aged care, long stay hospital settings or precarious and unstable accommodation
ID and high physical, and/or medical needs	Multiple, socio-economic disadvantage and social isolation
ID and challenging behaviour	Social exclusion with few opportunities for engagement in meaningful activities
Multiple and co-occurring disabilities and conditions	Co-existing conditions with ID such as mental health, alcohol, and/or drug misuse
Mild to moderate ID and a history of offending (forensic disability)	A background of trauma or neglect
Children with ID in out of home care with high physical disability or extreme challenging behaviours	

Service and System Responses and Outcomes for People with Intellectual Disability and Complex Needs

While the vision for people with complex needs is the same as for everyone—inclusion and participation, independent living and paid work, people with ID and complex needs often experience lower levels of quality of life, adverse life outcomes, and social isolation. They are disadvantaged in health, housing, independent living and employment (Cooper & Ward, 2011) and there is an overrepresentation of children with disabilities in child protection systems (Hill, 2012) with a contributing factor being increased likelihood of being abused and neglected (Horner-Johnson & Drum, 2006; Spencer et al., 2005).

Rankin and Regan (2004) state that those with the greatest (most complex) needs are at greatest risk of getting the least services. Also, that where care systems fail to recognise the interconnected nature of people's complex needs, they fall through the gaps between services. For people with complex needs there can be a lack of supports coordination, fragmentation of services, and poor information flow at government and local service level (Rosengard, Laing, Ridley, & Hunter, 2007). Accordingly, service systems and processes need to ensure that complex needs are met in an integrated fashion.

There are a number of factors that contribute to poor outcomes for people with ID and complex needs (Collings, Dew, & Dowse, 2016; Mansell & Beadle-Brown, 2012 ; Rosengard et al., 2007). These include:

- People with complex needs and their families/carers being daunted by complex service systems that span numerous agencies and are difficult to navigate, know what is available and how to go about gaining access (Mansell & Beadle-Brown, 2012; Rosengard et al., 2007). Gaps in required services and supports can occur where there is a lack of collaboration and integration by services and systems or inability to meet complex needs such as co-occurring mental health and intellectual disability. Other gaps and challenges people with ID and complex needs face arise where services and systems are too inflexible to respond to a breadth and depth of needs (Collings et al., 2016) and have administrative or bureaucratic barriers that must be crossed.

- Not being able to access the services or ending up in inappropriate services due to the nature of the person complex needs. Complex needs associated with severe disability, co-occurring mental health problems, autism and/or extremely challenging behaviours can lead to higher levels of out of area placement, associated with higher costs, lack of choice and control, social isolation and difficulties in transitioning back to their community of origin (Mansell & Beadle-Brown, 2012; Pritchard & Roy, 2006; Rosengard et al., 2007). People with complex needs are more often in long-stay hospital settings, "stuck" due to a lack of supported housing that meets their needs, level of risk and level of support required to live in the community (Sergeant & Brown, 2004). Additionally, they can find themselves in group accommodation where there is incompatibility between co-residents including resident to resident physical and verbal assault resulting in people with ID reporting that they feel unsafe in their home environment (Gatfield, 2016).
- Barriers to accessing required services and systems due to inflexible service criteria that may use ID or challenging behaviour as exclusion criteria (Mansell & Beadle-Brown, 2012; Rosengard et al., 2007) or have obscure or inconsistent entry criteria. Fragmented and siloed service systems that lack coordination and integration limit the person's ability to receive coordinated service provision and have their range of needs met (Mansell & Beadle-Brown, 2012; Rosengard et al., 2007).
- Resource constraints that prevent early and proactive supports, leading to crisis driven responses. Examples of such resource constraints include lack of respite care or short breaks for families or early intervention for younger children with challenging behaviour (Mansell & Beadle-Brown, 2012; Rosengard et al., 2007).

As can be seen, services and systems are critical to the quality of life outcomes for people with ID and complex needs. Therefore disability agencies and professions in the provision of supports to people with complex needs must be able to assist people with ID and their support networks to negotiate service systems to ensure needs are met and human rights are safeguarded.

Special Case: Family Crisis Resulting from Poor Service Response

While caring for a person with ID (regardless of age) can be a rewarding experience for parents and families (e.g.Corman, 2009 ; Grant, Ramcharan, McGrath, Nolan, & Keady, 1998 ; Hastings, Beck, & Hill, 2005), it can also be challenging for a multitude of reasons including the restrictions placed on the family's opportunities for social engagement (e.g.Burton-Smith, McVilly, Yazbeck, Parmenter, & Tsutsui, 2009 ; Edwards, Higgins, & Zmijewski, 2007), and the long-term impact on the physical and/or mental health of parents in particular (e.g.Burke, Patton, & Taylor, 2016 ; Burton, Lethbridge, & Phipps, 2008 ; Emerson, Robertson, & Wood, 2004 ; Miodrag & Hodapp, 2010). Consequently, some parents and families can experience a "crisis of caring" resulting in a request for a disability service to take over the day-to-day care of their family member with ID (*out-of-home care*).

Typically families requiring out-of-home care for their child with ID will go on to a waitlist for accommodation services in their jurisdiction. When a placement becomes available, the family usually works in collaboration with the service provider to implement planned transition to the accommodation arrangements.

However, there are instances where families seek immediate, unplanned emergency out-of-home care for their family member with ID as a result of family crisis. In these cases, the family member is often placed in temporary, emergency accommodation until the family agrees to resume the caring role, or an accommodation service is sourced if the family is unwilling to provide care on a continual basis.

Illustrative Case Study

Sam, a 12-year-old boy with autism and Down syndrome, lives at home with his mother. His mother wants him to continue to live at home; however, she needs assistance due to Sam's high levels of challenging behaviours. In-home supports are funded as well as some clinical services, including speech therapy to increase his communication skills. However his aggression has been escalating and Sam's mother does not believe that she can keep her other children safe from harm. While Sam has been supported in the family home by a disability service provider, that provider withdrew involvement due to assaults of workers and a lack of behaviour assessments or supports in place. Despite considerable efforts, a new service provider could not be engaged. A serious incident where Sam injured his mother led to the police being called and Sam being removed from the family home. Sam's mother refused to have him return home and crisis meetings were called by the government agency to find emergency accommodation. Sam lived in a number of short-term respite accommodation houses for 6 months until a longer term, more stable accommodation placement could be found.

Multiple studies have been conducted to investigate the reasons why families have sought out-of-home care for a family member with ID, with only a handful of studies specifically focusing on families who have sought unplanned, emergency out-of-home care during a period of family crisis. There is a noticeable overlap in the reasons reported by families who sought planned out-of-home care, and the families who sought immediate, emergency out-of-home care, with three groupings evident across the identified reasons. The first grouping relates to the *family member* with ID, who typically presents with complex needs due to severe disability, limited communication, challenging behaviour, and/or health problems (e.g.Ellem, Wilson, & Chenoweth, 2016 ; Essex, Seltzer, & Krauss, 1997 ; Grey, Griffith, Totsika, & Hastings, 2015 ; Llewellyn, Dunn, Fante, Turnbull, & Grace, 1999 ; McConkey, Nixon, Donaghy, & Mulhern, 2004 ; Nankervis, Rosewarne, & Vassos, 2011). The second grouping specifically relates to the *parent and other family members*, with parent inability to cope with the parenting role (e.g.Alborz, 2003 ; Mirfin-Veitch, Bray, & Ross, 2003 ; Victorian Equal Opportunity and Human Rights Commission [VEOHRC], 2012), poor parent health and/or mental health (e.g.Duvdevany & Vudinsky, 2005 ; Ellem et al., 2016 ; Nankervis et al., 2011), parental concern for their other children (e.g.Hostyn & Maes, 2007 ; Llewellyn, McConnell, Thompson, & Whybrow, 2005 ; VEOHRC, 2012), family conflict (e.g.Ellem et al., 2016 ; Hostyn & Maes, 2007 ; Llewellyn et al., 2005 ; McConkey et al., 2004 ; Nankervis et al., 2011), and financial strain (e.g.Chui & Hung, 2006 ; Ellem et al., 2016) cited as reasons leading to the decision to seek out-of-home care (or emergency out-of-home care).

The last grouping specifically relates to the *supports available* to the family to upkeep the caring role. Within this grouping, what is evident is that families who have sought out-of-home care (or emergency out-of-home care) are *reliant* on formal services available through the disability service system in order to cope with the caring role, mainly as a result of social isolation or a lack of social supports available to the family (e.g.Hostyn & Maes, 2007 ; Mirfin-Veitch et al., 2003 ; Nankervis et al., 2011). Because of this overreliance on formal supports (especially respite care), these families report significant unmet need for services, which then influences their ability to cope with the caring role on a long-term basis (e.g.Ellem et al., 2016 ; Llewellyn et al., 2005 ; McConkey, McConaghie, Barr, & Roberts, 2006 ; Mirfin-Veitch et al., 2003 ; VEOHRC, 2012). In addition, many families who have sought out-of-home care (or emergency out-of-home care) indicated that the services made available to them were of poor quality in their opinion (e.g. poor support practices in use, poor staff), resulting in more stress for the family as opposed to a sense of relief and assistance (e.g.Mirfin-Veitch et al., 2003 ; VEOHRC, 2012).

Coupled together, the known reasons for families to seek out-of-home care (or emergency out-of-home care) paint a picture of families who are experiencing many stressors, demands and hardships related to providing not only for the family member with ID, but the entire family. However, it should be noted that not all families caring for people with ID who have high support needs will seek out-of-home care. This is where formal service delivery and access to supports is crucial to families, and cannot be underestimated. Resilience is a notion widely discussed in the psychology research literature, with *parental resilience* recently defined by Gavidia-Payne, Denny, Davis, Francis, and Jackson (2015) as a parent's capacity to deliver quality parenting and care to their children despite adverse circumstances within the family. Gavidia-Payne et al. (2015) identify many protective and risk factors that are associated with parental resilience, with social connectedness being a crucial protective factor for the family (i.e. parental resilience is associated with greater levels of support). Also, resources such as social support and access to effective formal services allow a family to maintain their desired family social system, and if these resources are limited, family crisis can ensue (McCubbin & Patterson, 1983), Given the above, when it comes to conceptualising family crisis within the disability service system, the role of the service system and the supports available to a family cannot be neglected.

So what can agencies and professionals do to prevent family crisis? Recommendations from the research literature around the early identification of parents at-risk of seeking emergency out-of-home care (Nankervis et al., 2011; VEOHRC, 2012) have merit. Specifically, the VEOHRC (2012) advocate that systems must be put in place to accurately *identify* families at-risk of seeking emergency out-of-home care, with a focus on providing coordinated, early intervention supports to prevent these families from reaching a stage where they feel that crisis-driven, out-of-home care is their only viable option to cope. These recommendations for future practice were endorsed by parents who had sought emergency out-of-home care in the past (Vassos, 2017). Early identification of families at risk of crisis constitutes a proactive and prevention-based approach to ensure assessment of need processes that inform service allocation and support planning.

The Role of Agencies and Professions in the Provision of Care to People with ID Who Have Complex Needs

Systems, agencies and professionals are critical in supporting families and ensuring that people with ID's high and complex needs are appropriately met. The challenges in accessing the supports and services required must be addressed to avoid poor outcomes such as social isolation and poor quality of life. This necessitates an understanding of the depth and breadth of a person's needs and the implementation of a carefully targeted approach that understands and addresses the interaction between those needs and those that span systems (Rankin & Regan, 2004). An integrated approach to understanding complex needs and spans systems and processes enables the early identification of supports and interventions required to prevent crisis or other adverse outcomes.

The essential elements for such an integrated and coordinated approach to meeting complex needs are listed below (Carnaby & Pawlyn, 2009; Hudson, Dearey, & Glendinning, 2004; Rankin & Regan, 2004; Rosengard et al., 2007), and will be discussed further in this section:

- Understanding the needs of the person, especially their complexity.
- Access to information and advice.
- User and carer empowerment and co-design.
- Single point entry and access to required supports.
- Coordinated service delivery.
- Integrated systems.
- A proactive and preventative approach.
- Monitoring and measuring outcomes.

Understanding the Needs of the Person

In order to understand the person, a comprehensive, person-centred approach is essential, as well as understanding what constitutes complexity for the individual and their support network. An understanding of the whole person needs to be at all stages of service delivery and requires an exploration of the breadth, depth and interaction between needs (Rankin & Regan, 2004). By understanding the person and the basis of their complex needs, services can be tailored to the person, their circumstances and their needs. This requires comprehensive needs assessment that considers the whole person, and enables a person-centred approach where services are designed around the person rather than the person "fitting in" with the services that are on offer (Hudson et al., 2004; Rankin & Regan, 2004; Rosengard et al., 2007). Service planning and delivery should be designed to meet the needs of the person, not those of the service or system.

Access to Information and Advice

In order for people with ID to have their needs met, they need to be aware of what is available to them, both disability specific and mainstream. Commonly there is a lack of accessible information for people who have complex needs, and they have low awareness of what is available (Rosengard et al., 2007). Therefore information and advice for people with ID and their families must be accessible and understandable (e.g. written in Easy English, using plain language; translated into other languages), as well as readily available.

User and Carer Empowerment and Co-design

With human rights generally, and the UN Convention on the Rights of Persons with Disabilities (2006) specifically, giving primacy to a person's will and preferences must be actively supported (Carney, 2015). The best outcomes are where the person with ID is involved in planning their supports recognising and building on their strengths as well as those of their support network. As such, users of social services need to be recognised as co-producers of their own care (Rankin & Regan, 2004), and the experts about their own lives (Hudson et al., 2004). Involving people with ID and their carers at all levels of service planning, development and delivery as well as facilitating access to advocacy as required recognises the contribution the person with ID can make, and views them as the experts (Rosengard et al., 2007).

For people with complex needs and ID, choice, control and decision-making are important; however, due to the complexity of their needs (including severity on ID), decisions about the person's life may be undertaken by others, often under formal arrangements for substitute decision-making such as guardianship. Supporting a person with ID to participate in decision-making includes involving them right at the start; avoiding a tokenistic approach; allocating the necessary time, resources and commitment; and being clear about what exact decisions can be made (Hudson et al., 2004).

There is a growing focus on supported decision-making in response to overreliance on formal systems of substitute decision-making (Jameson et al., 2015). Supported decision-making aims to empower people with cognitive impairments, including ID, by recognising that they are the decision-maker, but are provided support for decision-making from one or more others from their network of support, giving them the assistance they need (Kohn, Blumenthal, & Campbell, 2012). This support is individualised to the person (Jameson et al., 2015) and may involve explaining issues and options in ways that are understandable to the person, or interpreting the individual's words or behaviours to identify preferences (Kohn et al., 2012). People with ID who exercise greater self-determination and have more control over their lives have greater independence and quality of life (Jameson et al., 2015). A reciprocal model emphasises the contribution that can, or could be made by the people in receipt of services and support (Hudson et al., 2004), recognising them as valued persons who have valued social roles. A person's level of participation in decision-making can range from being provided with information about decisions to be made, services and supports available, through to the individual having the authority and ability to make decisions for themselves (Hudson et al., 2004).

Single Point Entry and Access to Required Supports

Navigating complex and fragmented service systems is a significant barrier to people having their complex needs met. Users of service systems should be able to access the entire system of support through a single point of entry (Rankin & Regan, 2004). Single point entry can involve the ability to access appropriate services to meet different needs, information about available services (Rosengard et al., 2007) without the need to identify and then have contact with multiple agencies. Additionally, multiple legitimate routes to a variety of services from a variety of sectors that reflects diverse needs and circumstances are required (Hudson et al., 2004).

Coordinated Service Delivery

For a person with ID and complex needs in the context of service fragmentation, the roles of "service navigator" (Rankin & Regan, 2004; Rosengard et al., 2007) or "systems wranglers" (Madden, Fortune, Collings, & Madden, 2014) are needed to assist the person with ID and their family to identify needs and goals, plan their supports, and coordinate access to the required supports and services. Such a role needs to have knowledge of both specialist and mainstream services, as well as knowledge of specific issues, such as dual disability (Madden et al., 2014; Rankin & Regan, 2004). Case management is used to address situations where a person has complex needs and a single service response will not be adequate (Gursansky, Kennedy, & Camilleri, 2012). An essential element is a skilled coordinator, working across sectors and services, who is an active negotiator who understands the needs of the person who acts as the central point of linking services and other sectors (Madden et al., 2014).

Integrated Systems

An integrated systems approach is based on a holistic view of the person and a comprehensive, whole system model where all relevant services are involved in joint processes for information sharing (with the person's/family's permission) to prevent service users having to repeat information, facilitate pathways through services (Rosengard et al., 2007). Joined up and integrated service systems do more than just share information. They serve to enhance the supports provided by each other as well. Joint working is essential in order to have a "whole systems" approach (Hudson et al., 2004) rather than partnerships that are ad hoc or fragmented. In an integrated system working relationships are built around shared service user goals rather than organisational frameworks and existing systems (Hudson et al., 2004).

A Proactive and Preventative Approach

Early and proactive supports to meet identified needs and prevent escalation and crisis is essential. This implies identification and provision of supports and services to meet the needs of adults and children with ID in a timely manner rather than crisis driven responses. For example, children with intellectual disability are at increased risk of developing emotional and behavioural difficulties at an early age and carers experience stress related to the behaviours of the child (Gore, Hastings, & Brady, 2014; Nankervis et al., 2011). An early, preventative approach to providing behaviour supports, including parent education, can have a positive impact on the child's behaviour as well as carer stress and coping (Hudson et al., 2003). Without an early intervention approach, emotional and behavioural difficulties will persist over time into adulthood (Gore et al., 2014).

Additional to early interventions, timely and proactive identification of a person with ID as having complex needs and the provision supports is required. Systems, agencies and personnel need to reconsider approaches that prioritise access by urgency of need (crisis-driven) to ensure that reasonable and necessary supports are provided in a timely manner.

Monitoring and Measuring Outcomes

Continuous monitoring of how services are being provided and the outcomes for the person is critical so that progress in meeting their needs can be monitored and acted upon. Monitoring enables the identification of difficulties and barriers, identifying what is working well and achieving desired outcomes, which is essential to informing any reassessment of the person's needs and objectives. Monitoring the implementation of planned actions avoids implementation gaps that lead to poor outcomes, such as inappropriate accommodation or homelessness.

Monitoring should also focus on outcomes that are meaningful for the individual, be measured and the tools and ways of measuring should be individualised to the person's needs and goals (Carnaby & Pawlyn, 2009; Rosengard et al., 2007).

Hudson et al. (2004) draw on the evidence of the outcomes that service users contribute to a good quality of life as:

(a) Being healthy.
(b) Staying safe.
(c) Enjoying and achieving, including feeling valued and having access to social contact and company.
(d) Making a positive contribution.
(e) Economic well-being.

Of course, the role of agencies and professions in the provision of integrated and effective supports for people with ID who have complex needs is dependent on the skills, knowledge and experiences of the staff involved. The next section of this chapter will concentrate on the required competencies for the professions involved in service provision to people with ID and complex needs.

Professional Competencies

All professionals involved in planning and service delivery for people with ID who have complex needs require training to ensure that they are able to recognise and relate effectively to people with complex needs and their families, and assist them in navigating systems of services and supports available (Rankin & Regan, 2004). Given this, the concept of professional competence is fundamental to any discussion regarding service quality in the disability service sector, and will receive extensive focus.

In 1999, Hoffman stated there are three main approaches when it comes to defining *competencies:* (1) *observable competence,* (2) *standard of performance,* and (3) *underlying attributes*. In the first notion of observable performance—one must be able to witness performance on a task in order to attribute if someone can competently complete the desired task. The second approach is to conceptualise competency as a standard, i.e. a minimum standard of performance a person need to demonstrate in order to be deemed competent to complete the desired task. The third and last approach is to conceptualise competency in terms of a person's underlying attributes, i.e. the knowledge, skills or abilities that allow competent performance on the desired task. Hoffman also indicated that the approach to competency adopted will depend on context and requirements. When exploring the notion of professional competencies in the disability service system, the second and/or third conceptualisations will be focused on specifically.

When it comes to supporting people with complex needs, three professionals working in the disability service system have key roles to play to ensure quality service for people with ID—disability support workers, behaviour support practitioners, and case managers. The essential competencies associated with these three work roles, starting with disability support workers is discussed further.

Disability Support Workers

With regard to disability support workers, also referred to as disability support professionals, consistent conceptualisation of worker competency is difficult due to the variability of this role. Generally speaking, the role of a disability support worker is to ensure that support plans related to client personal care, health, behaviour, community access, and other relevant needs are effectively implemented on a consistent basis (Bogenschutz, Hewitt, Nord, & Hepperlen, 2014; Hewitt & Larson, 2007; Iacono, 2010). This may involve varying levels of direct care/support, ranging from supporting clients to complete daily living tasks to the best of their ability, to undertaking daily living tasks for their clients. Additionally, disability support workers can also be employed within various service settings such as supported accommodation, day programs and respite services, or within the clients' home. Given the heterogeneity of the intellectual disability population and the various service options available, the disability support worker role is diverse, with individual work duties and tasks typically determined by the worker's place of employment and the support needs of their clients (Bogenschutz et al., 2014). Given this, defining a universal framework for support worker competency may be difficult.

However, some researchers have attempted to piece together an all-encompassing set of competencies related to the disability support worker role. In line with Hoffman's (1999) third conceptualisation of competency, Taylor, Bradley, and Warren (1996) focused on understanding the skills, knowledge, abilities and personal characteristics that disability support workers require in order to effectively perform their role. Taylor et al. (1996) conducted focus groups with support workers working within various support services, and identified 144 relevant skills, knowledge and abilities within twelve competency areas (see Table 13.2). These competency areas are also reflected in a more recent competency analysis conducted by the National Disability Authority (2018) (see Table 13.2 also). The National Disability Authority (2018) reviewed ten competency frameworks regarding the disability support worker role from various countries, and uncovered 18 common competency areas. Eight of Taylor et al.'s (1996) competency areas were directly comparable with competencies reported by the National Disability Authority (2018). In comparison to Taylor et al. (1996), the National Disability Authority (2018) emphasised additional competencies related to staff behaviour (e.g. ethical practice, respect, resilience, safety and cultural awareness/practice, innovation) and specific work tasks (e.g. focus on client health/wellness, providing specific clinical support, working from a person-centred practice perspective, focusing on quality improvement). While the National Disability Authority (2018) mentioned staff behaviours as specific areas of competency, Taylor et al. (1996)

Table 13.2 Disability support worker competency areas (National Disability Authority, 2018; Taylor et al., 1996)

Taylor et al. (1996)	National Disability Authority (2018)
• Communication skills (to engage with clients)	• Communication (to engage with clients)
• Participant empowerment (to support decision-making, choice) • Advocacy (knowledge of issues facing people with a disability, skills to facilitate advocacy)	• Empowerment and advocacy (promoting clients to advocate for themselves, and supporting client advocacy)
• Assessment skills (in order to identify client needs)	• Evaluation, observation and assessment (regarding client need and support)
• Education, training and self-development (to maintain skills, knowledge and abilities)	• Education, training and self-development (to maintain skills, knowledge and abilities)
• Community living skills and supports (support to build client skills around independent living)	• Community living skills and supports (appropriate assistance to support clients to engage in meaningful activities and other services)
• Community and service networking (skills to facilitate service use, knowledge of services and supports available to clients)	• Community inclusion and networking (knowledge of services and supports available to clients, supporting client inclusion in the community and within their family and social networks)
• Crisis intervention (e.g. problem solving skills, conflict resolution skills)	• Crisis prevention and intervention
• Facilitation of services (i.e. creating support plans, implementing them)	• Planning and organisation (to promote partnerships, access to services for clients, and effective work practices)
• Vocational, educational and career support (to enable employment and participation in educational courses)	• Professionalism and ethics (ensuring all practice is considered ethical and professional)
• Organisational participation (i.e. knowledge of their service, participation in the processes of the service)	• Specific clinical support (related to behaviour support, mental illness and other clinical issues)
• Documentation (completion of)	• Health and wellness (supporting clients to maintain good health)
	• Quality (focus on service improvement, and evaluating quality of care provided to clients)
	• Person-centred practice (providing supports in line with this framework)
	• Safety (ensuring that the environment around the client is safe and predictable, and using safe work practices at all times)
	• Resilience, positive attitude and openness to change
	• Cultural (values and respects diversity, and supports clients in accordance to cultural customs and beliefs)
	• Respect, dignity and privacy (regarding their clients)
	• Innovation, creativity and problem solving (to ensure positive experiences for clients)

identified these behaviours as *ideal* support worker characteristics.

Further work in developing a universal competency framework for the disability support worker role is needed, based on the views of people with ID themselves. When asked to discuss the qualities of an effective support worker, people with ID place more emphasis on support worker skills and characteristics related to *interpersonal interaction* with clients, whereas service managers emphasise skills and characteristics that allow the worker to effectively complete their work duties (Dodevska & Vassos, 2013; Hatton, Wigham, & Craig, 2009).

Behaviour Support Practitioners

Behaviour support practitioners specialise in providing support to people with ID who exhibit challenging behaviour. According to Emerson (2001), challenging behaviours include behaviours that threaten the quality of life and/or the safety of a person with ID, or others who support the person (e.g. family members, support staff). Examples of such behaviours include self-injurious behaviours and aggressive or destructive behaviours. Behaviour support practitioners work in accordance to the principles of positive behaviour support. Positive behaviour support has its developmental roots in person-centred practice and the normalisation, social role valorisation and inclusion movements, while also intersecting the principles of applied behaviour analysis (Carr et al., 2002). Behaviour support practitioners work with people with ID, their families and their disability support workers to develop and implement a coordinated evidence-based intervention strategy that aims to enhance the person's quality of life while also minimising their challenging behaviour by addressing the function of that behaviour (Carr et al., 2002). Intervention approaches adopted in positive behaviour support include functional behavioural assessment, skill development using behavioural principles, and systematic environmental change.

Often behaviour support practitioners working with people with ID are trained in a discipline other than positive behaviour support, such as psychology, social work, nursing, occupational therapy or speech pathology. These professions will often have competency standards for registration to practice within the discipline. Because of this, behaviour support practitioners are likely assessed for competency based on the frameworks outlined by their relevant registration body, as opposed to a specific competency framework related to competently implementing positive behaviour support. Nevertheless, documenting competency to provide behaviour support services is starting to gain some traction within the disability service system, with the Positive Behaviour Support Coalition (2015) in the United Kingdom introducing a competence framework for providing best practice positive behaviour support services. In addition, to align with the introduction of the National Disability Insurance Scheme (NDIS) in Australia, the NDIS Quality and Safeguards Commission (2018) has introduced a national Behaviour Support Competency Framework. The competency framework of the Positive Behaviour Support Coalition will now be discussed in some detail.

The Positive Behaviour Support Coalition (2015) framework focuses on informing what knowledge behaviour support practitioners need to know, and the tasks/duties they will need to implement in order to deliver best practice positive behaviour support. The framework is divided into three main sections that describe specific competencies that need to be achieved by behaviour support practitioners, disability support workers and supervisor/management staff. The first section of the framework relates to *creating high quality care and support environments*, the second section relates to *functional, contextual and skills-based assessment*, and the last section of the framework relates to *developing and implementing a behaviour support plan*. Table 13.3 presents the summarised Positive Behaviour Support Coalition (2015) competency framework, with the detailed framework (not shown) breaking down each individual component in the framework into knowledge and work tasks/duties relevant to the three workers. The Positive Behaviour Support Coalition (2015) indicate that their framework can be used to support the development of a whole organisation approach to positive behaviour support, to develop curriculums related to training staff in positive behaviour support, and to support staff recruitment.

It can be expected that by undertaking competent practices that ensure the implementation of evidence-based, person-centred positive behaviour support plans, by behaviour support practitioners, in conjunction with the client and their supports, will result in positive outcomes. These outcomes include (1) enhanced quality of life for the person with ID, including active participation in meaningful daily life activities, (2) increased development of skills and positive behaviours

Table 13.3 The summarised Positive Behaviour Support Coalition (2015) competency framework

Creating high quality care and support environments	Functional, contextual and skills-based assessment	Developing and implementing a behaviour support plan (BSP)
• Ensuring that services are values led • Knowing the person • Matching support with each person's capabilities and with goals and outcomes that are personally important to them • Establishing clear roles and effective team work • Supporting communication • Supporting choice • Supporting physical and mental health • Supporting relationships with family, friends and wider community. • Supporting safe, consistent and predictable environments • Supporting high levels of participation in meaningful activity • Knowing and understanding relevant legislation • A commitment to behaviour skills training	• Working in partnership with stakeholders • Assessing match between the person and their environment and mediator analysis • Knowing the health of the person • Understanding the principles of behaviour (the four term contingency); understanding the function of behaviour • Supporting data driven decision-making • Assessing the function of a person's behaviour • Assessing a person's skills and understanding their abilities • Assessing a person's preferences and understanding what motivates them	• Understanding the rationale of a BSP and its uses • Synthesising data to create an overview of a person's skills and needs • Constructing a model that explains the functions of a person's challenging behaviour and how those are maintained • Devising and implementing multi-element evidence-based support strategies based on the overview and model (antecedent strategies, alternative behaviours, increasing skill and communication, systems change and contextual interventions) • Devising and implementing a least restrictive crisis management strategy (arousal curve, reactive strategies) • Developing the plan; outlining responsibilities and timeframes • Monitoring the delivery of the BSP (procedural/treatment fidelity/integrity) • Evaluating the effectiveness of the BSP • The BSP as a live document

that are functionally equivalent and replace the challenging behaviours, (3) reduction and elimination of the use of restrictive practices (such as chemical, physical or mechanical restraint, seclusion and containment), and (4) reduction of risk for the person and their supports, including decreasing risk of placement breakdown.

Case Managers

As previously discussed in this chapter, people with ID and complex needs are likely to require support from case managers to assist them in identifying and gaining access to required services and supports. People with ID and complex needs often require high intensity case management from case managers who have specialist skills and competencies (Cambridge, 2008).

The Case Management Society of America (CMSA;, 2016, p. 11) defines case management as "a collaborative process of assessment, planning, facilitation, care coordination, evaluation and advocacy for options and services to meet an individual's and family's comprehensive health needs through communication and available resources to promote patient safety, quality of care, and cost effective outcomes". The CMSA definition is an internationally recognised definition, with other case management professional bodies using this definition as the basis of their definition of the role, for example the Case Management Society United Kingdom (CMSUK;, 2015). Case management competency frameworks that are specific to people with ID and complex needs are scarce, and for that reason the more generic but widely recognised and adopted case management competencies of the CMSA (2016) and CMSUK (2015) will be the focus of this section. These frameworks are summarised in Table 13.4.

In line with Hoffman's (1999) second conceptualisation of competency as a set of minimum standards for workers, the CMSA (2016)

Table 13.4 A summary of the CMSA (2016) and CMSUK (2015) competency standards for case managers

CMSA (2016)	CMSUK (2015)
Client selection process—Screening referrals to identify those who will benefit most from case management	Professional practice—Engaging in regular supervision of practice, practicing autonomously and within professional boundaries, self-awareness and reflective practice, and continuing professional development regarding evidence-based practice
Client assessment—The completion of a thorough, person-centred assessment that takes into account the cultural and communication needs of the client and any other relevant stakeholders	Ethical conduct—Knowledge of relevant ethical codes of conduct, practicing ethically and with integrity, advocating on behalf of clients where appropriate, implementing procedures to adequately deal with client complaints, and working in accordance to relevant legislation
Care needs and opportunities identification—The identification of client care needs that would benefit from case management intervention	Communication—Providing information to clients and relevant stakeholders, employing appropriate communication strategies, developing rapport with clients and relevant stakeholders, and building skills to deal with difficult conversations or negotiations
Planning—In collaboration with the client and other relevant stakeholders, work to identify intervention goals and document them within the client's case management plan	Assessment—Using different methods to gather relevant information, applying relevant assessment measures for the client, and sharing (confidentially) knowledge and information for the benefit of others
Monitoring—Ongoing assessment of the client's response to their case management interventions	Goal setting—Identifying SMART goals, setting objectives to support the achievement of goals, and measuring outcomes relevant to the goals
Outcomes—Positive health and well-being outcomes are maximised as a result of case management intervention	Planning and prioritising—Evaluating different forms of information to identify issues and draw conclusions, organising information to support goal planning, identifying client priorities for intervention, producing case management plans relevant to the client's priorities and goals, demonstrating creativity and innovation to create the best possible plan for the client, and problem solving when faced with challenges
Closure—Undertake relevant procedures to close off case management services when appropriate	Implementing plans—Appropriate delivery of interventions, ensuring continuity in intervention, and coordinating activities in support of the client's objectives
Facilitation, coordination and collaboration—With clients and relevant stakeholders in order to achieve client goals and positive client outcomes	Monitoring and evaluation—Monitoring client progress to achieve goals, identifying effectiveness of interventions using appropriate measures, and applying continuous improvement principles (regular monitoring, etc.)
Qualifications—Maintaining certification to practice within a health or human services discipline or in the absence of a need for certification, evidence of qualification in a relevant health or human services discipline with a relevant fieldwork component as part of the qualification	Record keeping—Maintaining objective and accurate records, ensuring confidentiality and usability of information, presenting information in clear and concise reports
Legal—Adherence to all relevant legislation when providing case management services to clients	Management—Use appropriate referral avenues, engage in teamwork to support clients, assess and manage any client risks, managing resources appropriately and managing client expectation, and ensuring all support provided abides by organisational policy
Ethics—Ensuring all practice is ethical and adheres to ethical code of conduct governing the case manager's discipline of practice	Leadership—Making effective decisions, acting in a leadership capacity, and work in accordance to the principles of evidence-based practice in an effort to shape case management practice
Advocacy—Advocate on behalf of the client where appropriate	
Cultural competence—Maintain awareness of, and be responsive the cultural and communication needs of each client	
Resource management and stewardship—Integrating factors related to quality, safety, access, and cost-effectiveness when supporting clients	
Professional responsibilities and scholarship—Ongoing professional development to maintain knowledge and competency, and working in accordance to the principles of evidence-based practice	

and CMSUK (2015) competency frameworks provide a set of standards for case managers, with both frameworks also making explicit statements regarding how a case manager can demonstrate that they meet each standard. For example, regarding the CMSA's (2016) *monitoring* standard, case manager can demonstrate their competency by: (1) documenting their efforts to collaborate with their clients and relevant stakeholders to review client progress, (2) initiating changes to the proposed case management interventions in response to any changes to a client's circumstances, and (3) undertaking and documenting care plans reviews, and making efforts to ensure that updated intervention plans are acceptable to the client and relevant stakeholders. Comparing the two frameworks, there is overlap in content, implying that competency standards for case managers may have some universality. Of interest, competence in the areas of *assessment* of client need, *goal setting* and *outcome measurement*, intervention *planning* and ongoing *monitoring*, effective client *collaboration* and *communication*, and *ethical/professional conduct* are stressed by both frameworks.

In summary, the available evidence and information regarding professional competency for three key professionals who support people with ID who have complex needs is lacking. This is concerning given that staff competency plays a crucial role in ensuring service quality within any human service organisation. Researchers and policy makers need to prioritise the development, validation and evaluation of universal competency frameworks for key professionals working in the disability service system. This is especially the case when working with people who have complex needs as this cohort has a higher likelihood of adverse outcomes. Given that many countries are implementing significant changes to how disability services are provided to people with a disability, this task takes on extra importance as a method of ensuring consistent and effective staff practice to ensure high quality disability services that are focused on outcomes for individuals.

Concluding Remarks

Meeting complex needs for people with ID can be challenging. However these challenges can be overcome if services and systems work in ways that ensure integrated and coordinated service provision that meets human rights, provides safeguards, and improves quality of life. Often complexity of need is not associated with the individual or the need itself. Complexity arises where the right supports are not available or accessible, where service systems are fragmented and difficult to navigate, or where access to services is only prioritised where there is imminent crisis.

This chapter has highlighted the need for the early identification of families who are supporting children with ID and complex needs and are at risk of requiring out-of-home placement due to stress and inability to continue to care for their child. Such early identification enables the early and timely provision of the supports required to prevent escalation of the depth of the needs of the child and supports the family's ability to continue to care for their child in the family home.

Also discussed has been the essential elements required to be in place for social service systems, service provision agencies and professionals for best practices in providing care for people with ID who have complex needs. Having in place competent systems, agencies and professionals that understand an individual's complex needs will enable responses that result in positive outcomes that are meaningful and individualised to the person with ID and their support network.

References

Alborz, A. (2003). Transitions: Placing a son or daughter with intellectual disability and challenging behaviour in alternative residential provisions. *Journal of Applied Research in Intellectual Disabilities, 16*, 75–88. https://doi.org/10.1046/j.1468-3148.2003.00148.x

Bogenschutz, M., Hewitt, A., Nord, D., & Hepperlen, R. (2014). Direct support workforce supporting individuals with IDD: Current wages, benefits, and stability. *Intellectual and Developmental Disabilities, 52*(5), 317–329. https://doi.org/10.1352/1934-9556-52.5.317

Burke, M. M., Patton, K. A., & Taylor, J. L. (2016). Family support: A review of the literature of families of adolescents with disabilities. *Journal of Family Social Work, 19*(4), 252–285. https://doi.org/10.1080/10522158.2016.1214658

Burton, P., Lethbridge, L., & Phipps, S. (2008). Children with disabilities and chronic conditions and longer-term parental health. *The Journal of Socio-Economics, 37*, 1168–1186. https://doi.org/10.1016/j.socec.2007.01.032

Burton-Smith, R., McVilly, K. R., Yazbeck, M., Parmenter, T. R., & Tsutsui, T. (2009). Quality of life of Australian family carers: Implications for research, policy, and practice. *Journal of Policy and Practice in Intellectual Disabilities, 6*, 189–198. https://doi.org/10.1111/j.1741-1130.2009.00227.x

Cambridge, P. (2008). The case for a new 'case' management in services for people with learning disabilities. *British Journal of Social Work, 38*, 91–116.

Carnaby, S. & Pawlyn, J. (2009). Profound intellectual nd multiple disabilities: meeting complex needs through complex means. In J. Pawlyn & S. Carnaby (Eds.), *Profound intellectual and multiple disabilities: Nursing complex needs*. Malden, MA: Blackwell Publishing. https://doi.org/10.1002/9781444301526.

Carney, T. (2015). Supported decision-making for people with cognitive impairments: An Australian perspective? *Laws, 4*, 37–59.

Carr, E. G., Dunlap, G., Horner, R. H., Koegel, R. L., Turnbull, A. P., Sailor, W., … Fox, L. (2002). Positive behavior support: Evolution of an applied science. *Journal of Positive Behavior Interventions, 4*, 4–16. https://doi.org/10.1177/109830070200400102

Case Management Society of America. (2016). *Standards of practice for case management*. Little Rock, AR: Author.

Case Management Society United Kingdom. (2015). Case management framework. Retrieved from https://www.cmsuk.org/files/CMSUK%20General/Case_Management_Framework_Final_Conference%2026.10.15.pdf

Chui, M. Y. L., & Hung, R. C. H. (2006). Decision-making of Chinese caregivers for adult out-of-home placement. *Journal of Intellectual Disability Research, 50*, 678–689. https://doi.org/10.1111/j.1365-2788.2006.00826

Collings, S., Dew, A., & Dowse, L. (2016). Support planning with people with intellectual disability and complex support needs in the Australian National Disability Insurance Scheme. *Journal of Intellectual & Developmental Disability, 41*, 272–276.

Cooper, V., & Ward, C. (2011). *Valuing People Now* and people with complex needs. *Tizard Learning Disability Review, 16*, 39–43.

Corman, M. K. (2009). The positives of caregiving: Mothers' experiences caregiving for a child with autism. *Families in Society: The Journal of Contemporary Social Services, 90*, 439–445. https://doi.org/10.1606/1044-3894.3923

Dodevska, G. A., & Vassos, M. V. (2013). What qualities are valued in residential direct care workers from the perspective of people with an intellectual disability and managers of accommodation services? *Journal of Intellectual Disability Research, 57*, 601–615. https://doi.org/10.1111/j.1365-2788.2012.01565.x

Duvdevany, I., & Vudinsky, H. (2005). Out-of-home placement of children with intellectual disability: Israeli-born parents vs. new immigrants from the ex-USSR. *International Journal of Rehabilitation Research, 28*, 321–330. https://doi.org/10.1097/00004356-200512000-00004

Edwards, B., Higgins, D. J., & Zmijewski, N. (2007). The families caring for a person with a disability study and the social lives of carers. *Family Matters, 76*, 8–17.

Ellem, K., Wilson, J., & Chenoweth, L. (2016). When families relinquish care of a child with a disability: Perceptions from birthmothers. *Australian Social Work, 69*, 39–50. https://doi.org/10.1080/0312407X.2015.1015039

Emerson, E. (2001). *Challenging behaviour: Analysis and intervention in people with severe intellectual disabilities*. New York City, NY: Cambridge University Press.

Emerson, E., Robertson, J., & Wood, J. (2004). Levels of psychological distress experienced by family carers of children and adolescents with intellectual disabilities in an urban conurbation. *Journal of Applied Research in Intellectual Disabilities, 17*, 77–84. https://doi.org/10.1111/j.1360-2322.2004.00183.x

Essex, E. L., Seltzer, M. M., & Krauss, M. W. (1997). Residential transitions of adults with mental retardation: Predictors of waiting list use and placement. *American Journal on Mental Retardation, 101*, 613–629. https://doi.org/10.1352/1944-7558-119.6.496

Gatfield, O.R. (2016). *A conceptual model of quality service provision for people with intellectual disability and challenging behaviours: Enhancement and enrichment through service user and other stakeholder consultation.* PhD thesis, School of Psychology, The University of Queensland. doi: https://doi.org/10.14264/uql.2016.200

Gavidia-Payne, S., Denny, B., Davis, K., Francis, A., & Jackson, M. (2015). Parental resilience: A neglected construct in resilience research. *Clinical Psychologist, 19*, 111–121. https://doi.org/10.1111/cp.12053

Gore, N., Hastings, R., & Brady, S. (2014). Early intervention for children with learning disabilities: Making use of what we know. *Tizard Learning Disability Review, 19*, 181–189.

Grant, G., Ramcharan, P., McGrath, M., Nolan, M., & Keady, J. (1998). Rewards and gratifications among family caregivers: Towards a refined model of caring and coping. *Journal of Intellectual Disability Research, 42*, 58–71. https://doi.org/10.1046/j.1365-2788.1998.00079.x

Grey, J. M., Griffith, G. M., Totsika, V., & Hastings, R. P. (2015). Families' experiences of seeking out-of-home accommodation for their adult child with an intellectual disability. *Journal of Policy and Practice in Intellectual Disabilities, 12*, 47–57. https://doi.org/10.1111/jppi.12106

Gursansky, D., Kennedy, R., & Camilleri, P. (2012). *The practice of case management: Effective strategies for positive outcomes*. Sydney: Allen & Unwin.

Hastings, R. P., Beck, A., & Hill, C. (2005). Positive contributions made by children with an intellectual disability in the family. Mothers' and fathers' perceptions. *Journal of Intellectual Disabilities, 9*, 155–165. https://doi.org/10.1177/1744629505053930

Hatton, C., Wigham, S., & Craig, J. (2009). Developing measures of job performance for support staff in housing services for people with intellectual disabilities. *Journal of Applied Research in Intellectual Disabilities, 22*, 54–64. https://doi.org/10.1111/j.1468-3148.2008.00439.x

Hewitt, A., & Larson, S. (2007). The direct support workforce in community supports to individuals with developmental disabilities: Issues, implications, and promising practices. *Mental Retardation and Developmental Disabilities Research Reviews, 13*, 178–187. https://doi.org/10.1002/mrdd.20151

Hill, K. (2012). The prevalence of youth with disabilities among older youth in out-of-home placement: An analysis of state administrative data. *Child Welfare, 91*, 61–84.

Hoffman, T. (1999). The meanings of competency. *Journal of European Industrial Training, 23*, 275–286. https://doi.org/10.1108/03090599910284650

Horner-Johnson, W., & Drum, C. (2006). Prevalence of maltreatment of people with intellectual disabilities: A review of recently published research. *Mental Retardation and Developmental Disabilities Reszearch Reviews, 12*, 57–69.

Hostyn, I., & Maes, B. (2007). Parents' perspectives on grounds for out-of-home placement of young children with a disability. *The British Journal of Developmental Disabilities, 53*, 131–146. https://doi.org/10.1179/096979507799103423

Hudson, A. M., Matthews, J. M., Gavidia-Payne, S. T., Cameron, C. A., Mildon, R. L., Radler, G. A., & Nankervis, K. L. (2003). Evaluation of an intervention system for parents of children with intellectual disability and challenging behaviour. *Journal of Intellectual Disability Research, 47*, 238–249.

Hudson, B., Dearey, M., & Glendinning, C. (2004). *A New Vision for Adult Social Care: Scoping Service Users' Views*. University of York: Social Policy Research Unit. Retrieved from http://www.york.ac.uk/inst/spru/pubs.htm

Iacono, T. (2010). Addressing increasing demand on Australian disability support workers. *Journal of Intellectual and Developmental Disability, 35*, 290–295. https://doi.org/10.3109/13668250.2010.510795

Jameson, J., Riesen, T., Polychronis, S., Trader, B., Mizner, S., Martinis, J., & Hoyle, D. (2015). Guardianship and the potential of supported decision making with individuals with disabilities. *Research and Practice for persons with Severe Disabilities, 40*, 36–51.

Kohn, N., Blumenthal, J., & Campbell, A. (2012). Supported decision-making: A viable alternative? *Penn State law Review, 117*, 1111–1157.

Llewellyn, G., Dunn, P., Fante, M., Turnbull, L., & Grace, R. (1999). Family factors influencing out-of-home placement decisions. *Journal of Intellectual Disability Research, 43*, 219–233. https://doi.org/10.1046/j.1365-2788.1999.00189.x

Llewellyn, G., McConnell, D., Thompson, K., & Whybrow, S. (2005). Out-of-home placement of school-age children with disabilities and high support needs. *Journal of Applied Research in Intellectual Disabilities, 18*, 1–6. https://doi.org/10.1111/j.1468-3148.2004.00201.x

Madden, R., Fortune, N., Collings, S., & Madden, R. (2014). *Cross-Sector Service Coordination For People With High And Complex Needs: Harnessing Existing Evidence And Knowledge*. Centre for Disability Research Policy. Retrieved from https://ses.library.usyd.edu.au//bitstream/2123/13484/1/Cross%20Sector%20Service_FINAL.pdf

Mansell, J., & Beadle-Brown, J. (2012). *Active support: Enabling and empowering people with intellectual disabilities*. London: Jessica Kingsley.

McConkey, R., McConaghie, J., Barr, O., & Roberts, P. (2006). Views of family carers to the future accommodation and support needs of their relatives with intellectual disabilities. *Irish Journal of Psychological Medicine, 23*, 140–144. https://doi.org/10.1017/S0790966700009940

McConkey, R., Nixon, T., Donaghy, E., & Mulhern, D. (2004). The characteristics of children with a disability looked after away from home and their future service needs. *British Journal of Social Work, 34*, 561–576. https://doi.org/10.1093/bjsw/bch066

McCubbin, H. I., & Patterson, J. M. (1983). The family stress process: The double ABCX model of adjustment and adaptation. *Marriage & Family Review, 6*, 7–37. https://doi.org/10.1300/J002v06n01_02

Miodrag, N., & Hodapp, R. M. (2010). Chronic stress and health among parents of children with intellectual and developmental disabilities. *Current Opinion in Psychiatry, 23*, 407–411. https://doi.org/10.1097/YCO.0b013e32833a8796

Mirfin-Veitch, B., Bray, A., & Ross, N. (2003). "It was the hardest and most painful decision of my life!" Seeking permanent out-of-home placement for sons and daughters with intellectual disabilities. *Journal of Intellectual and Developmental Disability, 28*, 99–111. https://doi.org/10.1080/1366825031000147049

Nankervis, K., Rosewarne, A., & Vassos, M. (2011). Why do families relinquish care? An investigation of the factors that lead to relinquishment into out-of-home respite care. *Journal of Intellectual Disability Research, 55*, 422–433. https://doi.org/10.1111/j.1365-2788.2011.01389.x

National Disability Authority. (2018). *A review of competency frameworks for disability service staff.* Retrieved from http://nda.ie/Publications/Others/Research-Publications/Review-of-Competency-Frameworks-Disability-Service-Staff1.pdf

NDIS Quality and Safeguards Commission. (2018). *Behaviour support competency framework*. Retrieved from https://www.nds.org.au/images/resources/NDIS_Behaviour_Support_Competency_Framework_Draft_30_May_2018.pdf

Positive Behaviour Support Coalition United Kingdom. (2015). *Positive behavioural support. A competency framework*. Retrieved from https://www.skillsforcare.org.uk/Document-library/Skills/People-whose-behaviour-challenges/Positive-Behavioural-Support-Competence-Framework.pdf

Pritchard, A., & Roy, A. (2006). Reversing the export of people with learning disabilities and complex health needs. *British Journal of Learning Disabilities, 34*, 88–93.

Rankin, J. & Regan, S. (2004). *Meeting Complex Needs: The Future of Social Care*. London: Institute for Public Policy Research. Retrieved from https://www.ippr.org/files/images/media/files/publication/2011/05/Meeting_Complex_Needs_full_1301.pdf

Rosengard, A., Laing, I., Ridley, J., & Hunter, S. (2007). *A Literture review on Multiple and Complex Needs*. Edinburgh: Scottish Executive Social Research. Retrieved from https://www.gov.scot/Resource/Doc/163153/0044343.pdf

Sergeant, E., & Brown, G. (2004). Housing people with complex needs: Finding an alternative to traditional service models. *Housing, Care and Support, 7*, 25–30.

Spencer, N., Devereux, E., Wallace, A., Sundrum, R., Shenoy, M., Bacchus, C., & Logan, S. (2005). Disabling conditions and registration for child abuse and neglect: A population-based study. *Pediatrics, 116*, 609–613.

Taylor, M., Bradley, V., & Warren, R. (1996). The Community Support Skill Standards: Tools for managing change and achieving outcomes. Retrieved from http://files.eric.ed.gov/fulltext/ED400646.pdf

Vassos, M. (2017). *Developing a greater understanding of parental relinquishment of care in the disability sector* (unpublished doctoral dissertation). The University of Queensland, Brisbane, QLD.

Victorian Equal Opportunity and Human Rights Commission. (2012). Desperate measures: The relinquishment of children with a disability into state care in Victoria. Retrieved from http://www.humanrightscommission.vic.gov.au/index.php/our-projects-a-initiatives/children-with-disability-a-relinquishment

Part IV

Client Protections and Innovations

Informed Consent

14

Eileen Carey and Ruth Ryan

Values governing the need for informed consent 'promote and protect a person's well-being' and 'respect a person's self-determination' (Buchanan & Brock, 1990). Realising these values has proven to be problematic for people with intellectual disabilities, and those who support them, and as such, are a central focus of this chapter. While a vital component of supporting people with intellectual disabilities to make informed choices in their living, evidence suggests that informed consent means different things in different contexts and settings, variably practiced and rarely achieving the theoretical ideal. This chapter sheds light on and contributes to the theoretical and practical discourses of seeking, obtaining and facilitating informed consent for persons living with intellectual disability. Guided by the United Nations Convention on the Rights of Persons with Disabilities (UNCRPD) (2006) reasoning on 'rights' and 'disability' this chapter relates the notion of informed consent and activities of consenting with particular relevance to the application of CRPD's Article 12 (Arnardóttir & Quinn, 2009; Werner et al., 2012).

The chapter firstly traces the history of consent theory from pre-modernist, modernist and post-modernist perspectives. The evolving nature of informed consent is addressed and changing perspectives described noting the significance of evolving concepts of autonomy and self-determination specifically as relating to intellectual disability. Firstly historical reflections signify how deprivation and infringement of rights occurred (Kayess & French, 2008; Stein, 2007) often due to the absence of safeguards to insure that consent was informed rather than being coerced (Iacono & Carling-Jenkins, 2012). Secondly, aligned with the rise of bioethics, the endorsement of autonomy and self-determination of intellectually disabled individuals is demonstrated which aims to challenge readers to think of informed consent as more than personal rights but as augmenting inclusion in society. Thirdly, supported by two figures (Figs. 14.2 and 14.3) the principal processes and interactions of informed consent are presented which recognise both the simplicity and the multifactorial nature of informed consent communications, which require active involvement of those involved. Ultimately, this chapter demonstrates the enablement of intellectually disabled people to provide informed consent in all matters related to their life choices, and especially so for such examples as, receipt or use of a service, an intervention, or participation in research.

E. Carey (✉) · R. Ryan
Department of Nursing and Midwifery,
University of Limerick, Castletroy, Limerick, Ireland
e-mail: eileen.carey@ul.ie; ruth.ryan@ul.ie

J. L. Matson (ed.), *Handbook of Intellectual Disabilities*, Autism and Child Psychopathology Series, https://doi.org/10.1007/978-3-030-20843-1_14

Historical Developments of Consent

Despite the fact that numerous people from theology, law, anthropology, philosophy, medicine, nursing, psychology, sociology and psychiatry have debated the political, legal, ethical and moral issues and processes of consent, compared to many other areas in scientific enquiry, consent continues to be an evasive concept as no unifying theory exists. An overarching view of informed consent has been restricted. It is a widespread and varied phenomenon and despite extensive research and debate, remains deeply puzzling. In a capsulated review of consent theory, the literature abounds with studies representing theoretical perspectives that have influenced the study of informed consent, shaping and reshaping societies' understandings.

It is beyond the scope of this chapter to explore every theory or every writer; therefore, it is limited to works and writers taken as indicative of a particular era, and have influenced Western and Christian cultures. Three epochs of pre-modernism, modernism and post-modernism are utilised to provide a framework to debate the various discourses of consent. These three epochs produce three very different ways of understanding consent. More importantly, as the rise of consent theory has been diffuse, haphazard and a very slow process, it would be absurd to champion specific dates. To some extent these epochs overlap and it is the dominant philosophical system of the time and associated belief system as opposed to chronological time that is emphasised. The discourses presented highlight the different perspectives that prevailed and the manner in which permissions, prohibitions and alliances have been constructed based on each discourse. Table 14.1 provides an overarching framework, which has been conceptualised to guide some of the discussion in this chapter. Discussion of the various discourses lends insights into modern-day consent modes held by cultural institutions medicine, nursing and research.

Table 14.1 An overview of the discourses on consent

Pre-modernist discourses	Modernist discourses	Post-modernist discourses
A source of harmony	A source of liberalism	A legal-political concept
A source of conformity	A source of obligation	An ethical-legal concept
Control vs. autonomy	A question of morals	A basic human right
	An acknowledgement of rights	A communicative interaction

Pre-modernist

The pre-modern epoch extends from approximately the beginnings of western civilisation, in ancient Greece, through to the beginning of modernity in the early fifteenth century. The primary epistemology of pre-modernism emphasised the use of mythical thinking and myths to explain and interpret the world by the first civilisations of ancient Mesopotamia and Egypt. In exploring pre-modern discourses, including reference to consent in ancient Greece and consent codes of practice described by theologians with the Christian church, an enabling understanding of consent is conceptualised as 'a source of harmony' and 'a source of conformity' identifying informed consent as an ethic of subjection to religious authority and social order.

In this era, many writers spoke in terms of hierarchy and consensus, unity and subordination where social order required one unified hierarchy, with such matters involving counselling of the rulers and consent by the ruled (Herzog, 1989). Early pre-modern history therefore offers a concept of 'tacit consent', which is not necessarily verbalised, explicit or formalised. Regarded as implicit, 'tacit consent' denotes the tranquil and serene adhesion of the community to any determination presented as such, to a specific form of action, which was perfectly integrated within the customs and habits of that community and thus legitimated by norms and tradition' (Nerves 2004). The state of things was generally seen as unchanging, and the social order was strictly enforced. Consent evoked a common sense of feeling confirmed by habit or tradition rather than being understood as an individual prerogative, nor as the expression of the exercise of any (Fletcher 1983). Influenced by an enmeshment of religious and political activity, cultural and societal habit and tradition dominated routine choice. In other words, 'consent' referred to what was in conformity (what agrees) with the customs and habits of that community without being requested or granted explicitly or formally (Fletcher 1983). Such practices greatly reduced opportunities for understanding individual will and preference.

It was the Greeks who instigated ideas of political freedom, whereby thinkers, such as Plato and Aristotle developed ideas of statehood noting attributes such as the ability to reason and to exercise intelligence as being distinctively human. At the beginning of the tenth century, as the Church began expanding and secular kingdoms arose in supremacy, both church and kingdom struggled with issues of power over ultimate authority. The end of this era depicted the emergence of *rivals* to consent such as freedom and liberalism which still dominate consent discussions to the present day.

Modernist

The Modern era extends from approximately the early fifteenth century to the middle of the twentieth century. Modernism is known as the Age of Enlightenment, which emphasised a belief in human progress through rationality and the methods of science (Rolfe, 2006). The Age of Faith gave way to the Age of Reason. Discourses within this era focused efforts to rationalise behaviour on a scientific footing. Enmeshed in the generation of scientific knowledge and understanding and of new perspectives consent theory developed. Such perspectives began a change in focus from mythical beliefs, religious supremacy and moral constraints to concepts of liberty, freedom, obligations, morals and eventually rights.

The major theorists in this era Locke, Kant and Montesquieu presented discourses of consent which implied normative assumptions in that individuals behave according to the norms of the societies in which they live. In contrast to the pre-modernist control and conformity thinking around consent this paradigm shift marked the cornerstone of modern liberalism viewing society as a collection of free individuals (Herzog, 1989). Early modern theorists clearly supposed that individuals had basic normative powers over themselves before they entered into social contracts, which brought questions of political obligation to the fore. In the early stages of this era the main thrust of discourses surrounding consent focused on liberalism and obligation versus issues of morality and rights. Arguing that individuals were rational and freethinkers challenged previous mythical beliefs, religious supremacy and moral limitations. But due to nature that such individuals were living in societies which were governed by religious and political authority which determined social order these discourses of the modernist era became entangled in controversy. The main debate being that when individuals have the power to bind themselves by exercising the normative power, then the upshot of the social contract becomes obligation.

Kant challenged Locke's theory of natural law and natural rights emphasising morality as a general term for an individuals or a society's standards of conduct, both actual and ideal, and of the character traits that determine whether people are considered 'good' or 'bad' (Levine, 2008). In its simplest formulation, an act that is moral brings more good consequences than bad ones. This gave rise to a consideration of how morals and ethics impact consent. Kant believed that because of the constitution of the mind of an individual, they presuppose a relationship of cause and effect in all experiences with the objects of this world.

Kant (1795) claimed that consent was not fundamental to a social contract view but rather that the inner voice of individuals, the conscience, was the source of morality, which informed individuals of what was right and commanded them to do their duty. Often called deontological, this approach has been increasingly called Kantian, because of its origins in the theory of Immanuel Kant (1724–1804) (Beauchamp, Walters, Kahn, & Mastroianni, 2008).

The conceptualisation of consent in this era introduced notions of rights and the role consent plays in determining and enacting the legitimate ends that governments can pursue. Since Governments existed by the consent of the people in order to protect the rights of the people and promote the public good, governments that failed to do so could be resisted and replaced with new governments. Montesquieu (1689–1755) alleged the study of political and social behaviour not to be an exercise in abstract thought, but rather to be undertaken in relationship to geographic, economic, and historic conditions. Montesquieu claimed that such conditions, along with customs, habits, religions and institutions, gave each nation a particular character, and therefore each society required constitutional forms and laws that reflected and supported the character of its inhabitants. The consequences reflected in this paradigm shift in the change of thinking about liberalism as individual rights taking precedent over collective rights has major implications for adults with intellectual disabilities (discussed in section-rise of bioethics). Modern theorists' contribution to consent theory primarily insisted consent be deliberate, voluntary arising from self, all-be-it either individual or group consent.

Post-modernist

In the context of consent, post-modern theorists challenged previous discursive constructions of consent and questioned the taken for granted assumptions that were used as a basis for knowledge construction. Post-modernism adopted a critical approach to all knowledge, highlighting the inter-relationship between truth, knowledge and power (Kaye, 2007). Post-modernists rejected many of the principles of the scientific revolution arguing that society is characterised by differences as opposed to commonalities. Rather than trying to find a common theory or narrative to guide all actions, post-modernist theorists developed and consolidated consent by integrating it within a coherent and unitary system, which acknowledged historical and social contributions while integrating legal, political, moral and ethical principles grounded in appropriate communicative transactions.

Post-modernists argued consent to be a complex process founded in historical and cultural social constructions. The focus of these theorists was on deconstructing concepts that had been used to construct consent theory and to develop new approaches to informed consent. The outset of the post-modernist era coined the term ‘informed consent’ as a legislative concept that marked the beginning of an explosion of discourses and the production of a variety of modes of informed consent, which became instruments in the surveillance, regulation and control in maintaining human rights. Major theorists exploring consent in the initial part of this era were Ramsey (1930), Jonas (1969), Veatch (1972) and Fletcher (1978). Further developments are reflected in seminal works of Beauchamp and Childress (1979), Fadan and Beauchamp (1986), Beauchamp and Childress (2001), Berg, Appelbaum, Lidz, and Parker (2001) and Manson and O’Neill (2007). Informed consent began to be viewed as an ongoing process, a safeguard of personal statute, with communicative interaction as being recognised as one of the most critical elements involved.

The key contribution of the post-modernists era is the manner in which legal, political and ethical standards focused on respect for individual autonomy and inalienable rights. In other words, ‘informed consent’ theory was borne out of respect for individual autonomy, paralleled with the rise of the bioethical movement and debates around issues of gaining consent particularly relevant to minority groups such as people with intellectual disabilities. This focus served to promote a person’s right to self-determination, often happening within the context of the need for safeguards and complex communicative transactions within the decision-making process. Some of these pertinent issues are highlighted in the following sections.

Rise of Bioethics and Informed Consent

Noting historical developments of informed consent, it is clear to see how the expansion of informed consent practices and regulations have occurred in tandem with the broader evolution of society, noting bioethics and ethical participation of human subjects in research as significant movements in which the discourse of informed consent has given rise to contentious debate. Bioethics is integral to these considerations because it examines the ethical, legal and social issues of advances in bioscience in a number of disciplinary domains, namely ageing and end of life, clinical ethics, life sciences, disparities in health and healthcare and disability.

A notable abuse of humanity reflected in modernist times refers to the eugenics era, a movement purporting to 'improve the genetic composition of the human race'. Given the extent of the subject area, the brief outline of it here is inevitably illustrative rather than comprehensive. Basically, between the years of 1907 onto 1957 the eugenics campaign sought to promote a genetic purity which was guided by a White supremacist colonial project designed to maintain the purity, superiority, and health of the default, that being the White race (Llyes, 2018). Psychological and intellectual assessments played an essential and powerful player in this eugenic project (Winston, 1998) with new ways of classifying people, through a brief consideration of an essential psychological concept, 'the normal distribution', culminating in a statistical visual organising tool for populations (Hacking, 1995). And in rounding up the very tail of the normal distribution of population level intelligence quotient (IQ) scores, psychologists classified many people as feebleminded, imbeciles, idiots, and institutionalised. And for those who fell within this remit, many were subjected to atrocities such as forced sterilisations and inhumane institutionalisations.

Forced Sterilisations (Early 1900s)

Advancing this ideology of the eugenics perspective, societies' obligation was to prevent those considered 'feebleminded' from procreating so as to prevent 'them' from 'continuing their kind'. In the early 1900s the United States of America was the first country to undertake sterilisation for eugenic purposes, unleashing a global wave of forced sterilisations across many countries including Canada, Australia, Germany, Belgium, Scandinavia, Japan, China, India, Taiwan and South Africa. And by 1963 over 60,000 people in the USA had been sterilised without their consent (Roy, Roy, & Roy, 2012). However, Universal Human Rights have subsequently driven agendas to uphold the rights of people, by proclaiming that the necessity for each individual to provide informed consent on such decisions. More recently in 2014, in conjunction with a number of organisations the World Health Organization issued a joint statement: 'Eliminating forced, coercive and otherwise involuntary sterilization, An interagency statement' (WHO, 2014) which references the involuntary sterilisation of a number of specific population groups including women with intellectual disabilities.

The Vipeholm Sugar Trial (Sweden 1947–1955)

At the beginning of the twentieth century, Vipeholm, in Lund Sweden, was the only state hospital which catered for the needs of approximately 800 individuals who were intellectually disabled, classified at that time as the 'uneducable mentally deficient'. During this time, 'tooth decay' had become increasingly prevalent in the Swedish population. In order to establish an effective preventative public health strategy, the government commissioned the Folktandvåren (Public Dental Service) and Medical Board to establish an interdisciplinary team at Vipeholm to identify the cause of dental caries.

In 1947 progressing the Vipeholm Dental Caries Study, adding sugar to the basic (standard) diet of 436 patients in various controlled formats over the course of each day resulted in 2125 new dental caries been induced by the end of the study in 1953. Findings from this study have advanced our understanding related to oral health, and considered the most significant contributions to the entire dental literature as it:

> *'…definitively established that the more frequently sugar is consumed, the greater the risk, and that sugar consumed between meals has a much greater caries potential than when consumed during a meal'* (Zero, 2004)

However, ethical issues were not discussed formally either before or during the study and none of the patients involved in the study had given their informed consent (Zero, 2004). Subsequently, legislation was introduced which prevented the use of Vipeholm patients as research subjects (Zero, 2004). Similar inhumane treatment of children with intellectual disabilities was evidenced in the Willowbrook studies.

The Willowbrook Study (United States of America 1963–1966)

In the United States of America, the Willowbrook Study reflects a significant case of advance in medicine (Krugman, 1986). In this period of 1963–1966 over 700 children at the 'Willowbrook State School' were involved in a 'successful' experiment that discovered a vaccination for hepatitis under the principal investigator Dr. Saul Krugman. Krugman states that parents were informed that their children were likely to have only a sub-clinical infection followed by immunity to the particular hepatitis virus, and only children with parents who gave informed consent were included. The research subjects were all children deliberately infected with the hepatitis virus. Critics of this research differ in opinion, related to unethical or questionably ethical procedures involving risks to the health and consent which portrayed cognitive privileges in dehumanising individuals with intellectual disabilities (Carlson, 2009) and questions related to the concept of 'informed consent' (Iacono & Carling-Jenkins, 2012).

Children's diets consisted of extracts of stools from infected individuals, while others were administered injections of more purified virus preparations. For example, coercion and pressure have been identified as concerns for parents of children with intellectual disabilities, a notable case in the early 1970s, the 'Willowbrook State School for Children with Mental Retardation' (Klotz, 2004, Carlson, 2009, Iacono & Carling-Jenkins, 2012).

However, if consent obtained through coercion or parents feared losing their children's placements in the school, the principle of voluntariness of consent had been violated. Moreover, due to crowding and long wait lists for admission to the school, minimal home supports, and available rooms for children on the experimental wing, thus influencing the decision of some parents who did not have the resources to care for their children (Iacono & Carling-Jenkins, 2012). Krugman re-visits the ethics of this research in his 1986 paper recognising '*the accomplishments of the research to be well documented in the medical literature*' arguing that the development of a vaccine would outweigh the anticipated 'minor harms' to these children (Krugman, 1986). It is clear to see how the polarisation of risks and benefits of informed consent gives rise to many debates and controversial considerations in health services and research.

Ethical Considerations for Human Experimentation and Research

In efforts to protect the rights of people from abuse, the first international code of ethics, The Nuremberg Code, was established in 1949. The central declaration of The Nuremberg Code (1949) was that the voluntary consent of every human subject would be obtained prior to research being undertaken (Beauchamp et al., 2008). In 1964 The Declaration of Helsinki advocated prior review of research protocols by an '*independent committee*', as well as making explicit provision for participation in research by legally incompetent persons. Early history of research ethics policy focused on the risks and on preventing subjects from being exposed to unacceptable or exploitive levels of risk (Fadan & Beauchamp, 1986).

The Belmont Report (1978) later outlined what was considered the three most important ethical principles (respect for persons, beneficence, and justice) that should govern the conduct of research with human beings. And the 1994 National Institute of Health (NIH, 1994) guidelines ensured that research inclusion gained momentum over research exclusion. Therefore, the presumption about research participation flipped from exclusion (protection from risk) to inclusion (enabling autonomous choice about participation).

More recently, legal and international human rights framework emphasise the capacity of the intellectually disabled for social, physical, emotional and intellectual development, and in doing so, concepts of autonomy and independence have become central features (UNCRPD, 2006). Furthermore, in Article 31 of The United Nations Convention on the Rights of Persons with Disabilities (2006) researchers are now required to include representative populations of minority groups unless there are special reasons for excluding them. Published ethical guidelines for conducting research with people with Intellectual Disabilities has now come to the fore Lai et al. (2006). While various philosophies in relation to research ethics are now widely debated, Biggs (2010) accepts that no single theory or principle can be adopted as a guiding philosophy for researchers or research ethics committees (RECs). Biggs proclaims that REC decision-making needs to balance various approaches to arrive at a consensus decision. Conversely, McDonald et al. (2009) have argued that blanket prohibitions imposed by Institutional Research Boards may actually limit the autonomy and self-determination of intellectually disabled individuals and thereby increase their marginalisation in society.

Evolving Definitions of Informed Consent

The concept of informed consent has been described as evolving over centuries by different schools of thought. The changing definitions of informed consent have reflected changing attitudes to how best to seek, obtain, and maintain consent when engaging people, especially those regarded as being vulnerable. Generally legal, political, moral, regulatory, philosophical, medical, and psychological literature favour components of informed consent to include: (1) disclosure; (2) understanding (3) voluntariness; (4) competence and (5) consent. For example, Appelbaum (2007) conceptualised consent as encompassing four components, which include the ability to: (1) understand relevant information; (2) appreciate the consequences of the information for one’s own situation; (3) reason about the available options; and (4) communicate a choice. And specifically with regard to people with intellectual disabilities, Dye, Hare, and Hendy (2007) identified three main components of informed consent as: (1) possessing sufficient information relevant to the decision to be made; (2) having the capacity to make a decision and to understand the consequences of the decision; and (3) making the decision voluntarily and free from coercion. While focus shifted to consent capacity as being the determining issue in obtaining informed consent, defining it as ‘ability to understand information relevant to making an informed, voluntary decision’ (National Institutes of Health (NIH), 2009), the need to regard informed consent as a ‘respectful relationship’, and as a ‘process’ rather than a one-off event placed more onus on advocates for facilitating resources to enhance informed consent experiences, especially for those with intellectual disabilities (DHSSPS, 2003). And more recently, the boundaries of what exactly constitutes ‘informed consent’ have been broadened by specifically constructing the concept from an ethics of care perspective, created in notions of relational autonomy-in-informed consent (RIAC), which when placed within the remit of care ethics attracts global perspectives so as to enrich current ideas of bioethics principles of autonomy and informed consent (Osuji, 2018). Figure 14.1 presents a Venn diagram showing the multiple overlapping purposes of informed consent (Hall, Prochazka, & Fink, 2012).

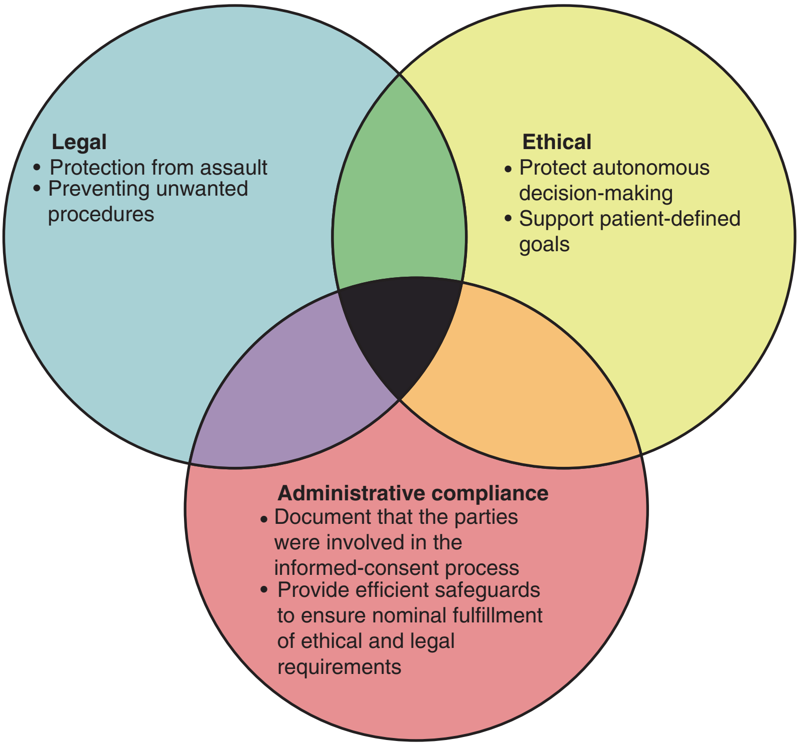


Fig. 14.1 Purposes of informed consent

Informed Consent Approaches

In asserting post-modernist theorists views of consent the following paragraphs and figures explicate the authors understanding of how informed consent can foster the autonomy and freedom of intellectually disabled people to make an informed decision. Infusing values of self-determination and choice can be challenging when people experience complex communication needs, impaired social interaction and limited experience of choice in addition to difficulty making informed choices for themselves (Bigby, Fyffe, & Ozanne, 2007), but not impossible. The following approaches Fig. 14.2: ‘Gaining Informed Consent: A Process’, and Fig. 14.3: ‘Informed Consent Gaining: An Interactive Cycle’ illustrate this perspective.

Gaining Informed Consent: A Process

The following Fig. 14.2 presents the standard ‘Gaining Informed Consent: A Process’, which portrays disclosure; understanding; voluntariness; competence and consent.

This framework has evolved from the initial focus whereby consent conveyed by an individual has been sought and obtained in culturally and linguistically appropriate language. Additionally, where written agreement has been utilised, integral in the act of signing and the existence of the signature is the credibility of the above-mentioned components of informed consent. Generally, the onus of proof of capacity has not been on the person seeking consent but rather on the consenting individual. In this standardised approach to gaining informed consent; the assimilation of and processing of information; the

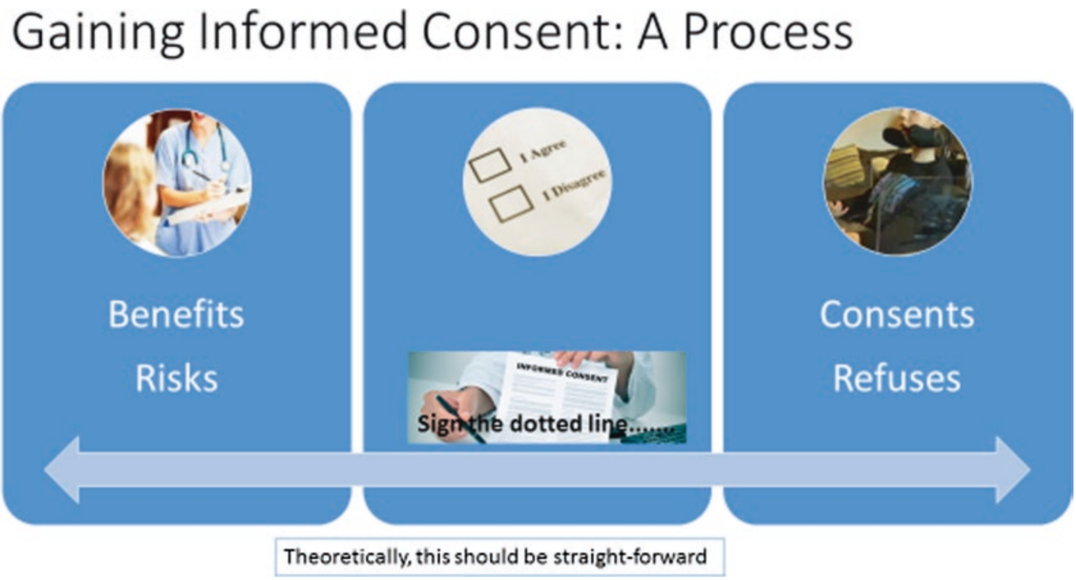


Fig. 14.2 Gaining informed consent: a process

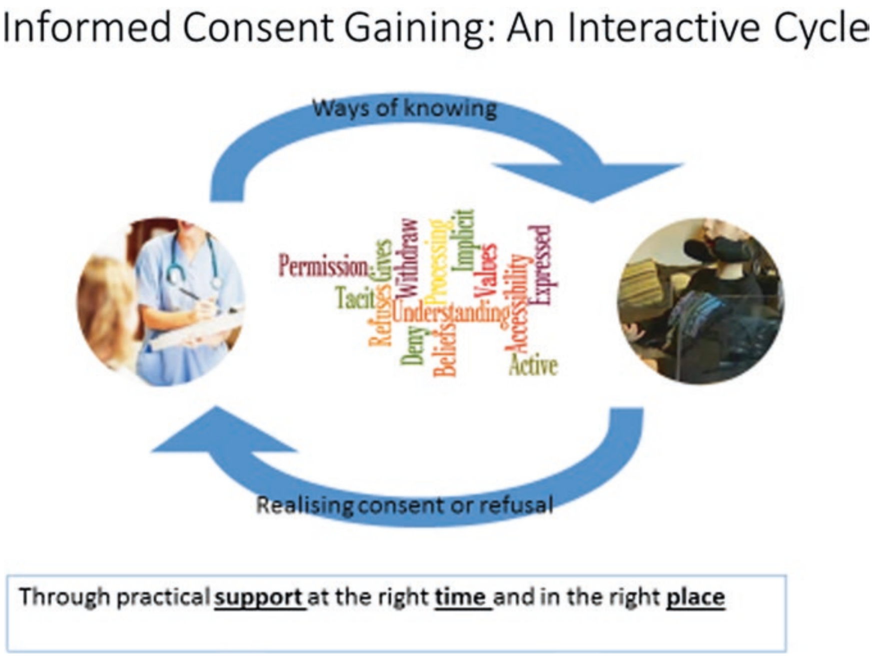


Fig. 14.3 Informed consent gaining: an interactive cycle

making of a judgement and the communication of a voluntary decision demonstrates the embodiment of theories of cognition and rational decision-making. Cautionary notes indicate such emphasis has rendered many people with disabilities, especially those with intellectual disabilities, disadvantaged in terms of communicating and explaining personal choices in these cognitive and rational domains. Particularly for those with moderate, severe and profound intellectual disability, the process of informed consent has traditionally been guided mainly by a status approach,[1] which determined that these individuals, by the nature of having an intellectual disability, did not have (legal) capacity. These people, subsequent to being deemed to lack capacity, were subjected to the enactment of guardianship laws. Reflective of individual and society responses to protecting those regarded as being vulnerable the introduction of guardianship laws aimed to demonstrate responsibility towards people at different periods in their lives, placing many people within custodial remits. And when rolled out in practical situations for people with intellectual disability, guardianship laws by the very nature of excluding them from matters of disclosure, voluntariness and choice rendered many unable to engage in the process of informed consent. Opportunities to make or engage in life choices was severely restricted for those who found themselves segregated by custodial care provisions. There is little doubt that the status approach obliterated the human rights of many people to engage and participate not only in having choices and making decisions for themselves as individuals but also in contributing to group and wider society decisions, erroneous thoughts in light of newer understandings of how this minority group of people can participate in making life choices.

Informed Consent Gaining: An Interactive Cycle

The 'interactional' perspective in Fig. 14.3 asserts the importance in developing meaningful relational interactions in time and over time to enhance mechanisms to develop engagement in disclosure (alternative and augmentative communication strategies); understanding (trust and relational); voluntariness (checking for signs of assent and dissent; comfort zones); competence (at ease to make a decision-with required supports) and consent (acceptance or refusal).

By gaining informed consent from people with intellectual disabilities, a 'respectful relationship', rather than a one-off event, is advocated enhancing interactions over a period of time. The impetus for this challenge reflects the beliefs in maximising human potential and services required to meet individual needs and outcomes while avoiding emphasis on limitations to understanding disability.

Underpinning these beliefs the value of communication displayed in 'Informed Consent Gaining—An Interactive Cycle' aims to present how meaningful and trustful relationships inherent within informed consent, placing emphasis on human potential, guided by a supports based approach. This philosophy aligns with the assumptions and understandings of the UNCRPD (2006) Handbook for Parliamentarians on the Convention which describes support in decision-making as:

> '*Those assisting a person may communicate the individual's intentions to others or help him/her understand the choices at hand. They may help others to realise that a person with significant disabilities is also a person with a history, interest and aims in life, and is someone capable of exercising his/her legal capacity*' pp.X

This interactive cyclical nature of seeking, obtaining, maintaining consent and/or respecting refusals or withdrawals to consent is underpinned by two principles, (a) *ways of knowing* people and (b) *maximising human potential*.

Ways of Knowing

A fundamental aspect of consent gaining interactions is that it challenges the '*ways of knowing*' people, from a pathological model where persons were perceived as objects or 'labels' in contrast to people with the potential to flourish as masters of their own lives, ability to self-determine, with a life plan and pathway, a life story or narrative (Bach & Kerzner, 2010; Barnes & Mercer, 2010; Department of Health, 2009; HSE, 2011; Lashewicz, Mitchell, Salami, & Samantha, 2014). In illuminating '*ways of knowing*' the practical implementation of supports, needs to take cognisance of individual values and principles that recognise contributions within communities.

---

[1] Status approach determining that capacity related to intellectual functioning and IQ singularly

With the right support, people with intellectual disabilities can exercise their right to make decisions and consent to life choices of their choosing. Willingness of support persons not only to listen to the person but to hear what the person is inferring via signs and expressions, recognising their individual and often unique mechanisms of communication. This lies at the heart of knowing the person. For this to be achieved personal supporters, support networks and/or circles of support should understand a person's life history, individual needs, routines, preferences and forms of communication. The value of familiarity and trust within any support for people with intellectual disabilities must be acknowledged since it evolves around supporters' willingness to develop connections, establish and sustain meaningful interactions (de Haas & Ryan, 2016).

Maximising Human Potential

The second principle of consent gaining interactions is that it must no longer consider intellectual disability as an absolute invariant trait of the person. Disability as a concept is viewed as a social–ecological construct exemplifying the interaction between the person and his or her environment. Therefore communications must focus on the role that meaningful interactions play in enhancing human functioning, self-worth and subjective well-being, so '*maximising human potential*'. Therefore when making a presumption that every person has the right to make decisions and be supported to exercise these rights (ACT, 2015; MCA, 2005), a movement beyond rational and cognitive theories therefore underpins the informed consent process, requiring recognition of affective theories (Cifor, 2016), which minimises the risk of 'absorbing the other'.

Expressing the changing understanding that intellectual disability needs to be recognised beyond an inherent pathological disorder to a more inclusive social ecological functioning, aids in reforming not only systems of support that people living with intellectual disabilities require but also the paradigm shift in medical, legal and familial understandings of delivering the practical functional supports required. In light of decisions to be made, taking cognisance of the timing of these decisions, people with intellectual disability should be supported to provide informed consent via a functional approach to decision-making in order to successfully engage in either or both of the above mentioned approaches.

Being Active Agents within Informed Consent

The following paragraphs demonstrate how people with intellectual disabilities can be supported to be active agents central to decision-making when providing informed consent. The aim being to diminish notions that people with intellectual disabilities are non-progressive and hold perpetually undeveloped cognitive capacities. Maximising decision-making capacity, supported decision-making models and circles of support demonstrate how people can be supported to be active agents within informed consent.

Maximising Decision-Making Capacity

It is now widely recognised that many individuals with intellectual disabilities are capable of providing informed consent with minimal, if any support. For others consent processes need to be tailored to maximise decision-making capabilities. Meaningful interactions need to be incorporated in order to provide support to individuals at the time the decision is to be made, taking cognisance of disclosure (of the procedure, expectations, benefits, risks); understanding (interpretation of information); voluntariness (having a choice, autonomy, self-determination, lack of coercion); competence (ability to make a decision-with required supports) and consent (acceptance or refusal).

For a person to be able to exercise decision-making capacity and make informed decisions, supported in a social and political environment, which recognises and promotes the value of autonomy allowing it to flourish is required (Atkinson, 2007). The terms autonomy and self-determination are sometimes used interchangeably but distinguishing between the two is helpful in relation to understanding 'informed consent'. Atkinson (2007) describes the difference between these terms as follows:

> *Autonomy is the capacity for self-government and self-determination: the ability to choose for oneself. (…/…) Self-determination requires an individual to have the capacity to formulate and carry out plans, desires and policies of their own devising. Self-government further requires the individual to take account of their own rules and values in making these choices* (Atkinson, 2007)

In maximising decision-making capacity, a recognition of the inherent values and respect for humanity in promoting autonomy and self-determination of all people, post-modernists move beyond the status approach of whether a person has capacity or does not have capacity, to acknowledging the wide range of applicability attributable to the moving statuses and functional approaches[2] to maximising capacity. The ability to understand the nature and consequences of a decision, when communicated in appropriate and respectful means, in the context of available choices, at the time the decision is to be made, with supports and accommodation ensures that a person is in a position to exercise their legal capacity.

As post-modernist perspectives of consent are adopted, and the philosophy and understanding of the United Nations Convention on the Rights of People with Disabilities the following definition of legal capacity supporting informed consent is fitting:

> *'Legal capacity to me is a continuum that connects with everything needed to enable the person to flourish—a right to make decisions and have them respected, a place to one's own, a life in the community connected to friends, acquaintances and social capital, whether in public or private settings. Personhood is broader than just capacity—and these broader connections serve to augment capacity in a virtuous circle'* (Quinn, 2011)

Therefore, this perspective flips the traditional belief that disabled people lack capacity to a belief in the unique ability of each person to express capacity by the nature of being a person. These changing beliefs represent a move in social processes from intellectually disabled people perceived as not having abilities, to favouring moving statuses and functional perspectives to viewing capacity as developing decision-making capability to self-determinate.

Supported Decision-Making Models

Supported decision-making models are now recognised as enablers to the implementation of practices facilitating people, especially those with intellectual disabilities, to provide informed consent when making life choices. The following addresses two prominent models of supported decision-making (SDM). Two models describing SDM, 'Maximising Decision-Making Abilities' (Bach & Kerzner, 2010) and 'Enabling Conditions' (Flynn & Arstein-Kerslake, 2014a), are conducive to the implementation of informed consent practices. From a Canadian perspective Bach and Kerzner (2010) conceptualise SDM as a process with outcomes, viewing SDM as:

> *'occurring when an individual with cognitive challenges is the ultimate decision-maker but is provided support from one or more persons who explain issues to the individual and, where necessary, interpret the individual's words and behaviour to determine his or her preferences through three different decision-making statuses'* (Bach & Kerzner, 2010)

The three statuses, (1) legally independent status, (2) supported decision-making status, and (3) facilitated decision-making status, are now presented.

Figure 14.4 demonstrates how providing meaningful supports and accommodations sufficient to the disabled person's unique needs and anticipated outcomes, over time, assists the disabled person in maximising their decision-making capability to exercise their legal capacity. This supports and accommodations approach significantly contrasts with the custodial nature of pervious historical approaches. Within this framework Bach and Kerzner identify decision-

---

[2] Functional approach

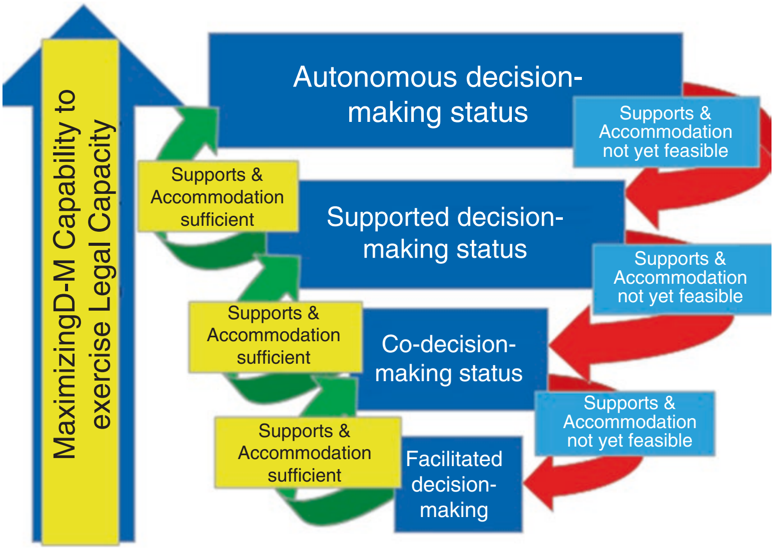


Fig. 14.4 'Maximising decision-making capacity' Bach and Kerzner (2010). This approach to 'decision-making capacity' advocated by Bach and Kerzner (2010) focuses on what accommodations and supports a person requires to manage the decision-making process in a way that maximises their decision-making capacity and thus exercise legal capacity

making statuses, which aim to be consistent with the person's identity. In general, while conceptualising distinct 'statuses' they acknowledge movement between legally independent and supported can be made, depending on the decision in question and the time the decision is made. The conceptualisation of SDM purported by Bach and Kerzner (2010) as a process with outcomes underpins decision-making capability has underpinnings in decision-making capability and seen as a relationship between decision-making abilities, decision-making supports and accommodations, and decision-making status. People move between statuses as abilities, supports and accommodations evolve reflected in Fig. 14.4.

The first autonomous decision-making status, familiar to most people, automatically recognises the person's capability to make decisions, by him/herself or with assistance, to understand and appreciate the relevant information, and the reasonably foreseeable consequences of, making a decision, therefore, exerting will and preference and exercising legal capacity. The first point on Bach and Kerzner continuum is the autonomous legal independent decision-making status, where an individual has the ability to make decisions on his/her own and is recognised as such. This may require reasonable accommodation to assist the decision-making process. One example of this would be ensuring that information relating to the decision is made available in a format which the person can understand (Goldbart & Caton, 2010), giving the person plenty of time to come to a decision, and enabling the use of informal support where the person can consult those closest to them in making the decision (The Office of the Public Advocate, 2014). This point reflects the ways in which most people make decisions by consulting others, while being free to accept or reject their advice.

In the supported decision-making status and co-decision-making status, a support person is appointed by the person with a disability, or by an administrative tribunal or court. This occurs specifically within a meaningful relationship between the person with a disability and his/her support person. The individual is provided with assistance in decision-making in any and all areas desired. This assistance can take the form of a circle of support, whereby the person chooses a number of trusted individuals to assist in the decision-making process (Atherton & Gates, 2007). These should be people who know the individual well, and can help interpret the person's will and preferences and communicate these intentions to third parties (Schalick, Westbrook, & Young, 2012), who are obliged to accept the decision as a valid one. The state should have a role in providing opportunities for support, enabling support

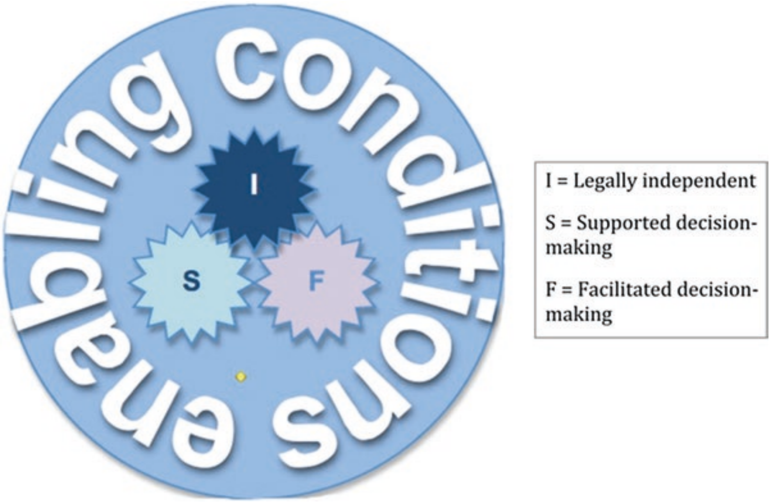


Fig. 14.5 Support model (Flynn & Arstein-Kerslake, 2014a). This model builds on the work of Bach and Kerzner (2010) and acknowledges the enabling conditions a person needs moving between the different statues of decision-making

agreements to be formalised and ensuring that decisions made through this mechanism are respected by third parties (Bach & Kerzner, 2010). Therefore, support person credibility to interpret the disabled person's will or intention, consistent with their personal identity becomes characteristic of supported decision-making, to exercise legal capacity.

In the final facilitated decision-making status a facilitator is appointed by administrative tribunal or advanced planning document (i.e., power of attorney). This decision-making status is applied as a last resort, where there is no circle of support or other person who could reasonably interpret the will and preferences of the individual. In this case, an appointed facilitator takes decisions on behalf of the individual, but does so with the will and preferences of the individual at the centre of the decision-making process and in the manner which best augments the person's autonomy and decision-making capability (Bach & Kerzner, 2010).

The benefits of Bach and Kerzner model is that is focuses understanding on the mechanism of SDM through 'decision-making capacity' when realising how accommodations and supports a person requires to manage the decision-making process in a way that maximises their decision-making capacity and thus exercise legal capacity, given their unique decision-making abilities. Building on the work of Bach and Kerzner (2010); Flynn and Arstein-Kerslake (2014c) purpose the 'Support Model' to SDM presented in Fig. 14.5.

Flynn and Arstein-Kerslake (2014a) SDM 'Support Model' image represents the overarching support model of legal capacity. The cogs in the centre of the circle represent the three statuses of decision-making: legally independent, supported and facilitated as previously outlined by Bach and Kerzner (2010). The major difference in the models relates to the area surrounding these statuses 'the enabling conditions' which include advocacy, reasonable accommodation, accessible information and communication, recognition of different forms of expression, advance planning tools, and so on.

The first principle of Flynn and Arstein-Kerslake model is that every person enjoys legal capacity regardless of his/her level of decision-making ability a principle of Article 12 UNCRPD, and each person should empowered to exercise his/her legal capacity through the expression of will and preference. Second, this requires the abolition of any assessment of decision-making ability (capacity assessment) which, if undertaken, may result in the loss of legal capacity. Any assessment which takes place in a support model of legal capacity should be centred on the support that is needed in decision-making to augment an individual's existing strengths, rather than the deficits of the individual (Flynn &

Arstein-Kerslake, 2014c). The facilitator's role is to imagine what the person's will and preferences might be and to make the decision on this basis. This distinguishes facilitated decision-making from substitute decision-making (Flynn & Arstein-Kerslake, 2014b).

Both models, Bach and Kerzner (2010) and Flynn and Arstein-Kerslake (2014a) are reflective of changing times, emphasising important aspects of the collaborative and relational aspects underpinning SDM in enabling a person to provide informed consent. The models require the abolition of substitute decision-making which subjects the will of an individual to the dominance of another's will or notion of what is in her 'best interest'. This does not prevent a representative from making a decision for another person who is not expressing her will and preferences in a way which anyone can interpret, rather, it requires representatives making such decisions to do so in a way which attempts to draw out the imagined will and preferences of the person (Flynn & Arstein-Kerslake, 2014c).

Recognising that many life choices can be complex, difficult to understand and challenging for some people, the following paragraphs introduce the concepts of circles of support providing for discussion of key elements required to support decision and enable a person provide informed consent.

Circles of Support

Popularity of circles of support in practice are emerging partly due to the philosophy and ideology surrounding the approach, perceived outcomes related to guidance (Macadam & Savitch, 2015), tangible resources and practical assistance (Lay & Kirk, 2011) and emotional support (Kaye-Beall, 2016). Circles of support originally designed to target social isolation and emotional loneliness have more recently been used as a support to intellectually disabled people to be autonomous in the decisions they make. Every circle is different because of the uniqueness of the individual at the centre of the circle and the uniqueness of the contribution of each circle member; therefore, it is important to recognise that one method will not suit all people who require support to make decisions, as everyone is different and will need different supports at different times (Browning et al., 2014).

Understanding person centeredness and circles of support contribute to understanding the ***ideology*** of SDM by facilitating individuals independence in decision-making, legal capacity and avoid having their right to control their decisions taken over by substitute decision-makers through co-production approaches. Perhaps worth noting however, even though a rights based approach to service provision is espoused, freedom and choices in identified service strategies to support this framework are currently more likely to revert support persons to charitable models and a dependency approach to support. For example, with family numbers reducing and family members more geographically dispersed, there is a need to look at natural support systems within local communities and this has been described as 'Circle of Friends' or 'Voluntary Workers' and 'Host Families'. These are generally unpaid and sustainability is challenging. These circles emulate principles similar to that of the social model of disability which recommends person-centred approaches of support to be achievable through assistance of local volunteers and often professional supporters, additional to paid workers (Lay & Kirk, 2011).

Circles have been given a variety of names and follow different models such as 'Self-Directed Support Corporations' and 'Circles of Support and Accountability' and are similar to methods of organising like 'Family Group Conferencing' (Neill & Sanderson, 2012). Therefore, the importance of basing care and support in addition to habitation practices and educational supports on substantial scientific evidence for practical, moral and legal reasons is acknowledged. Inclusion of evidence based practices contribute to the discussion of informed consent and intellectual disability. There is however a dearth of research and empirical literature in relation to circles of support notably in the areas of sustainability, productivity and cost effectiveness.

Trusting and Respectful Relationships in Gaining Informed Consent

Trusting and respectful relationships are pivotal constructs in the optimisation of autonomy pertaining to the life choices of people with intellectual disability. Yet consensus in the literature reveals that by the very nature of the label of intellectual disability, this population group are regarded as vulnerable (Parker, 2012; Parley, 2010) and subsequently their personal choices variably acknowledged and respected. The word 'vulnerable' is now widely used in health and social care, but its precise definition remains elusive, partly because of its universality and dynamism in that everyone becomes or feels vulnerable to a greater or lesser extent at different times in their lives (Pritchard, 2001; Rogers, 1997). Little, Paul, Jordens, and Sayers (2000): 495) identifies vulnerability as 'susceptibility to any kind of harm, whether physical, moral or spiritual, at the hands of an agent or agency' explicating the concept of vulnerability beyond the inherent disposition of an individual to (a) the role an external agent or agents, 9b) the notion of harm (either purposive or unintentional) and (c) the widening of the types of harm and safety beyond physical.

When vulnerability is recognised as being inherently human, perhaps a flaw with post-modernist conceptions of informed consent relate to the elevation of individualism at the expense of considering important relational and environmental considerations. This elevation of individualism minimises the interdependent nature of being and is perhaps not reflective of the experiences articulated by many intellectually disabled people. This chapter opened with the statement that values governing the need for informed consent 'promote and protect a person's well-being' and 'respect a person's self-determination' (Buchanan & Brock, 1990). Perspectives of both intellectually disabled people and those who support them should be established determining what exactly family, circles of support, citizenship, friendship and/or professional assistance looks like and how they work to ensure honesty, transparency and clarity when determining roles and functions. Therefore, the authors recognise how informed consent is based on the premises that all persons:

- Have decision-making abilities and make decisions.
- Require varying levels of support at different times.
- Receive appropriate communication to inform understanding.
- Experience trustful and respectful interactions.
- Have equal legal rights.

Building on the above premises, safeguards in post-modernist times especially for people with intellectual disability, provide that those who support this group of individuals are obliged to identify and act on the persons will and preference and to accept their determinations as valid decisions. It is the states' responsibility to ensure legal mechanisms are in place so as to explicate this will and preference. In the context of informed consent, the following sections present practical examples of access and participation in research and healthcare relevant to the lives of people with intellectual disabilities.

Informed Consent Gaining and Research Participation

As previously discussed, research was often undertaken about people with intellectual disabilities, rather than being inclusive of them, and often without their consent; therefore, such research participation was without safeguards (Taua, Neville, & Hepworth, 2014). Understandably, in the past number of years ethical regulation of health and social research has increased considerably (Wiles, 2013). Various international and national ethics guidance have now proliferated policies and procedures which enshrine the concept of 'informed consent' as the principal code to be adhered to protect the individual patient or healthy volunteer subject from possible exploitation and harm conceptually linking to discourses of human rights and autonomy. Particular challenges can present to researchers seeking to access population groups such as people with intellectual disability who are regarded to be some of the most vulnerable or marginalised in society, and researchers must ensure rigorous ethical safeguards are in place (Carey, 2010; Ramcharan, 2006). These people regarded to be vulnerable are entitled to participate in research while having their interests safeguarded (Smith, 2008).

Iacono (2006) describes the dilemma between the intentions of an ethical committee of protecting people with intellectual disabilities from potential harm when participating in research while guaranteeing them the autonomy to make their own decisions as they participate in research. Gilbert (2004) discusses the groups of people who may be involved and the influences they may have when research is undertaken in the field of intellectual disabilities. Diversity and complexity of lay and professional groups and individuals, along with knowledge, attitudes and power relationships are issues identified which can be influencing factors in research access and participation by people with intellectual disabilities. A prime example is provided by Carey and Griffiths (2017) who report that adults with intellectual disability were required to undergo physical and mental health assessments in order to be deemed to be fit to participate in a research study. These authors raised the question as to whether this was a reflection of principles of beneficence and non-maleficence or an act of discrimination?

Once ethical approval has been obtained potential participants with intellectual disability are often conscripted to research participation through intermediaries, such as their family, support workers, social workers, friends, unpaid carers or doctors (Nicholson et al., 2013). Formal gatekeepers are officially appointed within services whereas informal gatekeepers are people known to the person and who can influence the person's decision-making about research participation. For the duration of many research projects those who fulfil the role of formal gatekeeper are often managers nominated by organisations. Such gatekeepers are those people who control access to either participants or data (Gerrish & Lacey, 2007). Enlisting managers as gatekeepers can facilitate the research process and promote the best communication practices between the researcher, the participants and where necessary, families, about the nature and procedures associated with the research (Carey, 2010).

Conversely, undertaking such dual roles of gatekeeper and manager may also result in issues of power impacting the research, as gatekeepers have been regarded as people who can either help or hinder research depending upon their personal beliefs (McDonald & Kidney, 2012). Lee (1993) found that in some research, frequent attempts were made by gatekeepers to frustrate and circumscribe the studies, while in other research projects, gatekeepers had preconceived expectations of the appropriate methodology and refused access to the study population. Carey and Griffiths (2017) recommend to those proposing to recruit and undertake research with people with intellectual disabilities to consider:

- *The need to build and maintain trusting relationships with relevant formal and informal gatekeepers.*
- *To set time aside to understand the attitudes and concerns gatekeepers may have with regard to a research study, principally how it may impact on the potential participants with an intellectual disability, support persons and the service involved and to work to alleviate such concerns.*
- *To set time aside to get to know and understand potential research participants.*
- *To engage in reasonable accommodations to support decision-making with and for adults with intellectual disabilities so that they can make decisions about participating in research.*
- *Ensure the person understands what they need to do to participate, how the research actually applies to them.*
- *Endorse the activity of ongoing consent by ensuring the each participant is appropriately informed in a timely manner and where necessary offered reminders and repetitions of the processes involved while being vigilant and observant for signs of assent and dissent.*

- *Affording time for participants to process and assimilate information regarding the nature and procedures associated with the research study and their associated feelings and thoughts.*

Carey and Griffiths (2017) advocate the benefits of familiarity in enhancing research experiences for researchers, gatekeepers and people with intellectual disability. Familiarity of the ways of knowing the person can illuminate routine ways the person communicates, for example, how do they express how they want to opt in or opt out of ordinary life choices. By understanding the ways of knowing of the person the researcher can tailor the appropriateness of information sharing to the identified timely and decision-specific needs of the person. If researchers are to engage meaningful interactions with potential participants in order to identify their will and preferences and to gain informed consent then researchers need to spend time understanding their ways of knowing as expressed by these individuals themselves, an element which must be factored into ethics applications. Currently this is challenging as researchers have limited access to such groups prior to gaining their informed consent. Many researchers are not identifying frameworks which assist those undertaking research with people with intellectual disability who do not have time spent with the person prior to undertaking the research. For example, Taua et al. (2014) describes a qualitative research study undertaken in New Zealand. Taua et al. (2014) included individuals with a dual disability (intellectual disability and mental health issues). The overarching aims of the study were to understand what people with a dual disability and carers experienced during the time of an inpatient admission to a mental health service. University ethics approval granted in Australia and multi-region ethical approval granted by a national ethics committee in New Zealand. Making minor adaptations and additions to those originally developed by Dougall and Fiske (2008, pp. 73–75), Taua et al. (2014) presented a framework for researchers to follow during the consent gathering process taking cognisance of the following:

- *Communicate slowly and clearly, quiet location with minimal interruptions.*
- *Concepts and questions introduced one point at a time.*
- *Use an appropriate level of language for each individual.*
- *Provide a written copy of the key points (use colour where possible).*
- *Combine words and pictures wherever possible.*
- *If possible, encourage the person to take their own notes so they can check back with you.*
- *Explore other ways of providing information if written and/or verbal information do not work (*e.g. *video, audio, pictorial).*
- *Use diagrams and flowcharts wherever possible (at least have several different formats prepared).*
- *Ask the person to repeat back frequently to confirm understanding.*
- *Utilise a support person/translator, as necessary.*
- *Allow enough time.*

Furthermore, for researcher perusal these author provided a list of reflective questions:

- *Did I provide all the information needed?*
- *Could the information be presented in a way that is easier to understand (*e.g. *visual aids, or with simpler language)?*
- *Have I fully explored all the different methods of communication?*
- *Did I ensure the right supports/people were in place in order to prevent coercion?*
- *Have I explained all the risks and benefits?*
- *Did I allow enough time for questions?*
- *Did I check back frequently enough?*
- *Was the environment appropriate/conducive?*
- *Have I documented the consent process thoroughly?*

Taua et al. (2014) recognise how the basic frameworks for seeking informed consent from potential participants should not differ dramatically for any person, as there is a level of risk for all participants in research. It is critical, however, that researchers recognise the higher levels of vulnerabilities in the intellectually disabled population, especially those with a dual diagnosis and ensure that their recruitment processes are rigorous and respectful. Research with people with intellectual disability should maximise the benefits of the research and minimise the risks, and those who are to conduct the research should be competent to do so, and if needed, be appropriately supervised. Researchers must not only be vigilant in listening to the communications of their research participants but they must hear what they are saying. Researchers must ensure that their research approaches can be flexible enough to concur with changing needs of research participants.

Informed Consent Gaining and Healthcare

In terms of the prevalence of health conditions, individuals with an intellectual disability have poorer health than others (Emerson, 2009). Entitled to be treated with dignity, respect, honesty and be involved in decisions about their health and well-being, it is well known that people with intellectual disabilities experience challenges in this regard (Krugman, 1986; Resch et al., 2010). Specifically healthcare access and healthcare participation arise as prominent issues of vulnerability for this population group (Philips, 2016; Wark et al., 2017). For those supporting people with intellectual disabilities there is a necessity to place more attention on care needs, access to healthcare, preventive care and health promotion (Lafferty, O'Sullivan, O'Mahoney, Taggart, & van Bavel, 2016; Sheerin, 2011).

Wark et al. (2017) sought to understand informed consent when supporting individuals ageing with intellectual disability to participate in developing their own health and support programs. Indeed, challenges in the provision of oral healthcare to people with intellectual disability have been acknowledged as physical dexterity, impaired sensory functioning, communication and behavioural problems (Knibb, 2010). And notably, in the provision of healthcare, the presence of cognitive disorders have traditionally been synonymous with critical impairment of decision-making abilities. Subsequently, a long-standing culture of paternalistic practices have existed and for some people remain evident, especially so when those providing services to people with intellectual disabilities have limited experience of intellectual disability.

The following demonstrates how the attitudes and practices of healthcare professionals can impact the informed consent experience of people with intellectual disabilities. In aiming to identify the main approaches adopted by dentists in a French study, Camoin, Dany, Tardieu, Ruquet, and Coz (2018) found a 'reassuring attitude' to be the main approach that ensued when treating an anxious child with intellectual disability. Four topics were discussed, first contact with the patient, information, attitude towards the patient and outcome from the practitioner's viewpoint. The coding procedure used thematic content analysis. In all twelve dentist participant's, eight women and four men (mean age = 42.5), three had an exclusively hospital practice, three had a private practice, and six practiced in both settings. Regarding experience, four practitioners had less than 5 years' experience, six practitioners had over 15 years' experience, and two practitioners had between 5 and 15 years' experience. All practitioners interviewed declared that they informed the patient about what was happening, from the start and throughout the session. This information was conveyed by various means (verbal and non-verbal communication) and various approaches (distraction, tell-show-do, transitional object). Different communicational strategies used were:

- ***Tone of voice***: *'I mainly use a soft and empathetic tone', 'my voice is calm, composed and even' (male, more than 15 years' experience, private practice).*
- ***Tell-show-do***: *'I show the patient the mirror, I make them touch it with their fingers, and then together we put it in the patient's mouth' (female, more than 15 years' experience, private practice); 'I always show the patient the*

instruments and let the patient touch them before I put them in their mouth' (*female, less than 5 years' experience, both private and hospital practice).*
- ***Use of a transitional object*** *(cuddly toy): 'If the patient brings a cuddly toy, it can also be used to show that the mirror is harmless' (female, between 5 and 15 years' experience, private practice).*
- ***Distraction****: 'Did you have a good holiday?' (female, between 5 and 15 years' experience, both private and hospital practice). 'I tell him a story to distract him, with a knight or a princess for example' (male, between 5 and 15 years' experience, hospital practice).*

These strategies demonstrate a variety of approaches and various mechanisms promoting relations between practitioners and children with intellectual disabilities. Worryingly, the practitioners within the study reported that if an ethical dilemma arose, and the patient refused care or had to be restrained, ethical values of patient autonomy would be sacrificed, under the realms of beneficence and non-maleficence, when clinical decisions were made.

Similarly, Goldsmith, Skirton, and Webb (2008) conducted an integrative literature review examining how consent was obtained from people with intellectual disabilities for healthcare interventions. Time limits set were publication between January 1990 and March 2007, encompassing 17 years of research with adult populations. The data reflected the opportunities and processes that intellectually disabled people demonstrated within consent to research, consent to treatment or (indirectly) consent to taking medication, while some data reflected hypothetical vignettes and some 'real-life' situations. Overall, the integrative review findings highlight the negative attitudinal responses as a main barrier to obtaining informed consent by healthcare staff. Several of the included studies reported that staff assumed that individuals with intellectual disabilities were unable to consent to treatment, and one study demonstrated that staff were unaware of guidelines relating to consent. Not surprisingly, assumptions of staff in this review demonstrate the prominent attitude of the status approach, all or nothing attitude to gaining informed consent. The review recommends a functional approach to assessing capacity for the purpose of obtaining informed consent recognising the complexity and nature of the decision need to be taken into account (Goldsmith et al., 2008).

Therefore, whether or not capacity to consent is achieved depends on the effort to 'tailor' the relevant information to the abilities and needs of the individual concerned. A significant point raised in this integrative review recognises how professionals are obliged to comply with their country's legal requirements; in practice, however, this may cause problems due to the time needed to maximise the potential for capacity in many people with intellectual disabilities. Future research into informed consent in people with intellectual disabilities is needed using real-life situations which are more likely to be familiar to the participants than hypothetical vignettes.

In 2013, further developing the theme of gaining informed consent with real-life situations, Goldsmith, Woodward, Jackson, and Skirton (2013) conducted a primary qualitative study using an ethnographic approach. People with intellectual disabilities were eligible for this study if they were aged 18 years or over, able to consent to participate in research, not affected by acute physical health problems, and not currently under the care of the local mental health team. The final inclusion criterion was the requirement for a routine blood test (e.g. for monitoring blood sugar control) within a few months of recruitment.

Participants' age ranged from 27 to 65 (mean 45). Half (50%) lived in a shared house (supported living), with the remainder either living at home with parents, living independently with support, or living along with informal family support. Participants with severe or profound intellectual disabilities were excluded, and this limits the transferability of the findings. Twelve of the 14 participants were single.

Observations and interviews formed the methods of data generation. Six participants were observed having one blood test, the observation period ranged from 3 to 6 min. All participants observed having the blood test were due to have blood tests as part of their routine medical care. All six participants were well known to the health professionals who performed the blood test. Health professionals included phlebotomists, healthcare assistants and registered general nurses. One semi-structured interview was conducted with 14 participants lasting between 12 and 35 min. All participants were offered the opportunity to bring a supporter to the interview and ten did so.

Consultations and interviews were video- and audio-recorded to ensure accurate transcription and enable observation of non-verbal behaviour. Data analysis was approached in an inductive way, considered appropriate for an exploratory, descriptive study guided by Braun and Clarke frameworks (Braun & Clarke, 2006). In all, the researchers identified six key themes; the patient in the healthcare context, information and knowledge, the consent process, behavioural characteristics, strategies and coping mechanisms, and 'the self'. Significantly, researchers noted that the consent process did not appear to be fully followed in any of the consultations observed in the taking of blood. In some cases, adequate information was provided. However, in the majority of cases, the health professional did not check that the participant understood the information given; this is an essential requirement for informed consent.

Goldsmith et al. (2013) reiterate the strength of their 2008 findings adding how personal qualities and experiences may influence the consent process; factors such as a person's place of residence, previous health experience and opportunities to make their own life choices all contribute to the individual's ability to consent to healthcare interventions.

In this study, the health professionals knew all participants described as 'experienced' patients. This may have led the health professionals to make assumptions concerning consent. As this study was conducted in the UK Goldsmith et al. (2013) noted that these assumptions would not be sufficient to fulfil the requirements of the Mental Capacity Act (2005), as it is the responsibility of the person carrying out the procedure to ensure that valid consent is in place at the time and this should include provision of relevant information. The right to self-determination and the right to full disclosure are major components on which informed consent is based (Grove, Burns, & Gray, 2014; Polit & Beck, 2004).

In addition, in the Goldsmith et al. (2013) study, there are two points worth highlightening (1) people with intellectual disability lack experience in decision-making, and the affect this has on their functional capacities with regard to informed consent and (2) clearly expressed their information needs with regard to their healthcare; they required clear, accessible information, with any verbal explanations being simple and without jargon.

Most notable, in Goldsmith 2013 from the 2008 study is the change in attitude of staff, in the belief that people with intellectual disabilities seek information and consent to healthcare procedures. These studies demonstrate the need to ensure there is greater awareness of increasing opportunities for effective communication to ensure that intellectually disabled people have choice, are in control and active in decision-making regarding their life choices. Nevertheless, even with the global and philosophical impetus of supporting decision-making, particularly with the implementation of the UNCRPD, current research reports continue to highlight challenges of implementing such moral and ethical principles.

Conclusion

Over the years consent theorists' have and continue to strive to illuminate and develop both the simplicities and intricacies of informed consent. An overview of historical contexts of consent theory, the impact of the bioethics and the evolving definitions informed consent lead to a current representations of informed consent both as a process and as an interactive cycle.

This chapter has presented informed consent as a dynamic, contextually driven, respectful approach to viewing people with intellectual disability primarily as decision-makers, inextricably linking informed consent with supported decision-making and rights.

This chapter presented a new way of thinking about informed consent and the role of communication and interaction. Figures 14.2 and 14.3 present an understanding of the components of informed consent asserting the importance in developing meaningful relational interactions in time and overtime to enhance mechanisms developing engagement in disclosure, understanding, voluntariness, competence and consent.

Building on such premises, the authors acknowledge the need for safeguards ongoing process and maximising human potential in supporting people with intellectual disability to identify and act on their will and preference.

Recommendations

There is a need for persons with intellectual disabilities, policy makers, legislators, service providers and supporters, to recognise how processes and interactions of informed consent maximises human potential.

People with intellectual disabilities need to have meaningful involvement at all levels in all aspects of decision-making and informed consent which affects their life choices.

- Persons with intellectual disabilities be included in education, research and philosophical debates exploring what informed consent means to them in their lives.
- Perspectives of both intellectually disabled people and those who support them to be established determining what exactly family, circles of support, citizenship, friendship and/or professional assistance looks like and how they work to ensure honesty, transparency and clarity when determining roles and functions in supporting people to make informed decisions.

As policy and legislation continues to develop in response to the needs of people with disabilities progress needs to be monitored and evaluated from the perspectives of all stakeholders. To continue driving the human rights agenda, building on the work of the UNCRPD to enable people with intellectual disabilities to take their rightful place in society as active citizens.

Service providers ensure that reasonable accommodations are afforded to people with intellectual disabilities so that they are equal members of their society.

In maximising decision-making support services need to ensure that meaningful interactions are incorporated in practices central to successful processes and interactions.

There is an onus to ensure explicit structures and procedures are utilised to enable the person to make decisions.

Individuals who support persons with intellectual disabilities must engage trustful relationships, aiming to support the person to explicate their will and preference to maximise human potential in decision-making. And when necessary ensure the availability and delivery of information about reasonable accommodations to enable the person in providing informed consent.

References

Appelbaum, P. S. (2007). Assessment of patients' competence to consent to treatment. *The New England Journal of Medicine, 357*, 1834–1840.

Arnardóttir, O. M., & Quinn, G. E. (2009). *The UN convention on the rights of persons with disabilities: European and Scandinavian perspectives* (Vol. 100). Leiden/Boston, FL: BRILL.

Assisted Decision Making (Capacity) Act. (2015). Number 64 of 2015, Dublin: Stationary Office, [online], Retrived from http://www.irishstatutebook.ie/eli/2015/act/64/enacted/en/print.html. Accessed 5 May 2017.

Atherton, H., & Gates, B. (2007). *A history of learning disabilities*. Edinburgh: Churchill Livingstone.

Atkinson, J. M. (2007). *Advance directives in mental health: Theory, practice and ethics*. Jessica Kingsley Publishers.

Bach, M. and Kerzner, L. (2010). A New Paradigm for Protecting Autonomy and the Right to Legal Capacity Prepared for the Law Commission of Ontario. The Ontario Law Commission, Canada, [online], Retrived from https://www.lco-cdo.org/wp-content/uploads/2010/11/disabilities-commissioned-paper-bach-kerzner.pdf. Accessed 6 Apr 2018.

Barnes, C., & Mercer, G. (2010). *Exploring disability a sociological introduction*. Cambridge: Polity Press.

Beauchamp, T. L., & Childress, J. F. (1979). *Principles of biomedical ethics*. Oxford: Oxford University Press.

Beauchamp, T. L., & Childress, J. F. (2001). *Principles of biomedical ethics* (5th ed.). Oxford: Oxford University Press.

Beauchamp, T. L., Walters, L. R., Kahn, J. P., & Mastroianni, A. C. (2008). *Contemporary issues in bioethics*. Wadsworth, OH: Thompson.

Belmont Report. (1978). *Ethical Principles and Guidelines for the protection of human subjects of research*. National Institutes of Health (NIH) Maryland: Office of Human Subjects Research (OHSR).

Berg, J. W., Appelbaum, P. S., Lidz, C. W., & Parker, L. S. (2001). *Informed consent legal theory and clinical practice* (2nd ed.). Oxford: Oxford University Press.

Bigby, C., Fyffe, C., & Ozanne, E. (2007). *Planning and support for people with intellectual disabilities: Issues for case managers and other professionals*. London: Jessica Kingsley Publishers.

Biggs, H. (2010). *Healthcare research ethics and law: Regulation, review and responsibility*. Abingdon: Routledge-Cavendish.

Braun, V., & Clarke, V. (2006). Using thematic analysis in psychology. *Qualitative Research in Psychology, 3*(2), 77–101.

Buchanan, A. E., & Brock, D. W. (1990). *Deciding for others: The ethics of surrogate decision making*. New York, NY/Cambridge: University Press.

Camoin, A., Dany, L., Tardieu, C., Ruquet, M. and Le Coz, P. (2018) Ethical issues and dentists' practices with children with intellectual disability: A qualitative inquiry into a local French health network. Disability and Health Journal 11(3):412-419.

Carey, E. (2010). Navigating the process of ethical approval: A methodological note. *The Grounded Theory Review, 9*(3), 19–32.

Carey, E., & Griffiths, C. (2017). Recruitment and consent of adults with intellectual disabilities in a classic grounded theory research study: Ethical and methodological considerations. *Disability and Society, 32*, 193–212.

Carlson, L. (2009). *The faces of intellectual disability: Philosophical reflections*. Bloomington, IN: Indiana University Press.

Cifor, M. (2016). Affecting relations: Introducing affect theory to archival discourse. *Archival Science, 16*(1), 7–31. https://doi.org/10.1007/s10502-015-9261-5

de Haas, C., & Ryan, R. (2016). Chapter 4 family centered care. In L. Rubin, J. Merrick, D. E. Greydanus, & D. R. Patel (Eds.), *Health care for people with intellectual and developmental disabilities across the lifespan* (3rd ed., pp. 49–58). Switzerland: Springer International Publishing.

Department of Health, Social Services and Public Safety (DHSSPS). (2003). Seeking consent: Working with people with learning disabilities, [online], Retrived from http://www.dhsspsni.gov.uk/consent-guidepart4.pdf. Accessed 31 Oct 2014.

Department of Health, U. (2009). *Valuing people now; A new three-year strategy for people with learning disabilities*. London: Department of Health.

Dougall, A., & Fiske, J. (2008). Access to special care dentistry, part 3. Consent and capacity. *British Dental Journal, 205*(2), 71–81.

Dye, L., Hare, D. J., & Hendy, S. (2007). Capacity of people with intellectual disabilities to consent to take part in a research study. *Journal of Applied Research in Intellectual Disabilities, 20*(2), 168–174.

Emerson, E. (2009). Estimating future numbers of adults with profound multiple learning disabilities in England. *Tizard Learning Disability Review, 14*(4), 49–55.

Fadan, R. R., & Beauchamp, T. L. (1986). *A history and theory of informed consent*. New York: Oxford University Press.

Fletcher, G. P. (1978). *Rethinking criminal law*. Boston, FL: Little, Brown and Company.

Fletcher, J. (1983). The evolution of the ethics of informed consent. In K. Berg & K. E. Tranoy (Eds.), *Research ethics* (pp. 187–228). New York, NY: A.R. Liss.

Flynn, E., & Arstein-Kerslake, A. (2014a). The support model of legal capacity: Fact, fiction, or fantasy. *Berkeley Journal of International Law, 32*, 124.

Flynn, E., & Arstein-Kerslake, A. (2014b). Legislating personhood: Realising the right to support in exercising legal capacity. *International Journal of Law in Context, 10*(01), 81–104.

Flynn, E., & Arstein-Kerslake, A. (2014c). The support model of legal capacity: Fact, fiction, or fantasy? *Berkeley Journal of International Law, 32*, 134–281.

Gerrish, K., & Lacey, A. (2007). *The research process in nursing* (5th ed.). Oxford: Blackwell Publishing.

Gilbert, T. (2004). Involving people with learning disabilities in research: Issues and possibiliites. *Health and Social Care in the Community, 12*, 298–308.

Goldbart, J. and Caton, S. (2010). Communication and people with the most complex needs: What works and why this is essential. *Research Institute for Health and Social Change. The voice of learning disability.* Mencap.

Goldsmith, L., Skirton, H., & Webb, C. (2008). Informed consent to healthcare interventions in people with learning disabilities – An integrative review. *Journal of Advanced Nursing, 64*, 549–563.

Goldsmith, L., Woodward, V., Jackson, L., & Skirton, H. (2013). Informed consent for blood tests in people with a learning disability. *Journal of Advanced Nursing, 69*(9), 1966–1976.

Grove, S. K., Burns, N., & Gray, J. (2014). *Understanding nursing research: Building an evidence-based practice*. St. Louis, MO: Elsevier Health Sciences.

Hacking, I. (1995). He looping effects of human kinds. In D. Premack & D. Sperber (Eds.), *Causal cognition: A multidisciplinary debate* (pp. 351–394). New York: Oxford University Press.

Hall, D. E., Prochazka, A. V., & Fink, A. S. (2012). Informed consent for clinical treatment. *Canadian Medical Association Journal, 184*(5), 533–540.

Health Service Executive (HSE). (2011). *Time to move on from congregated settings: a strategy for community inclusion-Report of the working group on congregated settings*. Dublin: Health Service Executive.

Herzog, D. (1989). *Happy slaves a critique of consent theory*. Chicago, IL: University of Chicago Press.

Iacono, T. (2006). Ethical challenges and complexities of including people with intellectual disability as participants in research. *Journal of Intellectual and Developmental Disability, 31*(3), 173–179.

Iacono, T., & Carling-Jenkins, R. (2012). The human rights context for ethical requirements for involving people with intellectual disability in medical research. *Journal of Intellectual Disability Research, 56*(11), 1122–1132.

Jonas, H. (1969). Philosophical reflections on experimenting with human subjects, ethical aspects of experimentation with human subjects. *Daedalus, 98*(2), 219–247.

Kant, I. (1795). Perpetual peace: A philosophical sketch [online], Retrived from https://www.mtholyoke.edu/acad/intrel/kant/kant1.htm. Accessed 29 Apr 2017

Kaye, D. H. (2007). Revisiting 'Dreyfus': A more complete account of a. *Trial by Mathematics.' Minnesota Law Review, 91*(3), 825–835.

Kaye-Beall, K. (2016). Centre for Autism Suport and Training. 'Circle of Support? This is the heart of Person-Centered Planning' [online], Retrived from http://centerforautismsupportandtraining.com/self-direction/circles-of-support/. Accessed 6 Apr 2018

Kayess, R., & French, P. (2008). Out of darkness into light? Introducing the convention on the rights of persons with disabilities. *Human Rights Law Review, 8*(1), 1.

Klotz, J. (2004). All in the mind intellectual disability; Communicating across the divide, All in the mind [video], [online], Retrived from http://www.abc.net.au/radionational/programs/allinthemind/intellectual-disability-communicating-across-the/3425430. Accessed 19 Feb 2014.

Knibb, D. (2010). Establishing a procedure for managing poor standards of oral care in vulnerable and dependent adults. *Journal of Disability and Oral Health, 11*(1), 14–16.

Krugman, S. (1986). The Willowbrook hepatitis studies revisited: Ethical aspects. *Review of Infectious Diseases, 8*(1), 157–162.

Lafferty, A., O'Sullivan, D., O'Mahoney, P., Taggart, L., & van Bavel, B. (2016). *Family carers' experiences of caring for a person with intellectual disability*. Dublin: University College Dublin.

Lai, R., Elliott, D., & Ouellette-Kuntz, H. (2006). Attitudes of research ethics committee members toward individuals with intellectual disabilities: The need for more research. *Journal of Policy and Practice in Intellectual Disabilities, 3*(2), 114–118.

Lashewicz, B., Mitchell, J., Salami, E. and Samantha, C. (2014). Understanding and addressing voices of adults with disabilities within their family caregiving contexts; implications for capacity decision making and guardianship, Law Commission of Ontario, [online], Retrived from http://www.lco-cdo.org/capacity-guardianship-commissioned-paper-lashewicz.pdf. Accessed 5 May 2017

Lay, J., & Kirk, L. (2011). Chapter 9: Person centred strategies for planning. In H. Atherton & D. Crickmore (Eds.), *Learning disabilities: Towards inclusion*. Croydon: Elsevier Health Sciences.

Lee, R. (1993). *Doing research on sensitive topics*. London: Sage.

Levine, C. (2008). *Taking sides clashing views on bioethical issues* (12th ed.). New York, NY: McGraw Hill.

Little, M., Paul, K., Jordens, C. F. C., & Sayers, E. J. (2000). Vulnerability in the narratives of patients and their carers: Studies of colorectal cancer. *Health, 4*(4), 495–510.

Llyes, E. (2018). Care-ful definition of consent. *Journal of Health Psychology, 23*(2), 277–288.

Macadam, A., & Savitch, N. (2015). Staying connected with circles of support. *The Journal of Dementia Care, 23*(1), 32–34.

Manson, N. C., & O'Neill, O. (2007). *Rethinking informed consent in bioethics*. Cambridge: Cambridge University Press.

McDonald, K., Kidney, C., Nelms, S., Parker, M., Kimmel, A., & Keys, C. (2009). Including adults with intellectual disabilities in research: Scientists' perceptions of risk and protections. *Journal of Policy and Practice in Intellectual Disabilities, 6*, 244–252.

McDonald, K. E., & Kidney, C. A. (2012). What is right: Ethics in intellectual disabilities research. *Journal of Policy and Practice in Intellectual Disabilities, 9*(1), 27–39.

Mental Capacity Act. (2005). Mental capacity Act (MCA) (2005) [online], Retrieved from www.legislation.gov.uk/ukpga/2005/9/pdfs/ukpga_20050009_en.pdf. Accessed 30 Sept 2014

National Institute of Health (NIH). (1994). Guideline on the inclusion of women and minorities as subjects in clinical research, [online], Retrived from http://grants.nih.gov/grants/guide/notice-files/not94-100.html. Accessed 31 Oct 2014.

National Institutes of Health (NIH). (2009). Research Involving Individuals with Questionable Capacity to Consent: Points to Consider' [online], Retrived from https://grants.nih.gov/grants/policy/questionablecapacity.htm. Accessed 26 Mar 2018

Neill, M. and Sanderson, H. (2012). Circles of Support and Personalisation, [online], Retrived from http://www.allenshea.com/wp-content/uploads/2017/03/circlesofsupportandpersonalisation-3.pdf. Accessed 06 Apr 2018

Neves, P. M. (2004). Cultural context and consent: An anthropological view. *Medicine, Health Care and Philosophy, 7*, 93–98.

Nicholson, L., Colyer, M., & Cooper, S. A. (2013). Recruitment to intellectual disability research: a qualitative study. *Journal of Intellectual Disability Research, 57*(7), 647–656.

Nuremberg Code. (1949). [Online], Retrived from www.nj.gov/health/irb/documents/nuremburg_code.pdf. Accessed 31 Oct 2014

Osuji, P. I. (2018). Relational autonomy in informed consent (RAIC) as an ethics of care approach to the concept of informed consent. *Medicine, Health Care and Philosophy, 21*, 101–111. https://doi.org/10.1007/s11019-017-9789-7

Parker, M. (2012). Caring for vulnerable ostomists: Learning disabilities and stoma care. *British Journal of Nursing, (Stoma Care Supplement), 21*(16), S4–S10.

Parley, F. (2010). What does vulnerability mean? *British Journal of Learning Disabilities, 39*(4), 266–276.

Philips, A. (2016). Vulnerable adults with diabetes: Improving access to care. *Practice Nursing, 27*(6), 287–290.

Polit, D. F., & Beck, C. T. (2004). *Nursing research: Principles and methods*. Philadelphia, PA: Lippincott Williams & Wilkins.

Pritchard, J. (2001). *Good practice with vulnerable adults* (Vol. 9). London: Jessica Kingsley Publishers.

Quinn, G. (2011). Rethinking Personhood: New Directions in Legal Capacity Law & Policy. Lecture, University of British Columbia, Vancouver, 21(7) [online], Retrieved from Accessed 29 April 2011.

Ramcharan, P. (2006). Ethical challenges and complexities of including vulnerable people in research: Some pre-theoretical considerations. *Journal of Intellectual and Developmental Disability, 31*(3), 183–185.

Ramsey, F. P. (1930). On a problem of formal logic. *Proceedings of the London Mathematical Society, 2*(30), 264–286.

Resch, J. A., Mireles, G., Benz, M. R., Grenwelge, C., Peterson, R., & Zhang, D. (2010). Giving parents a voice: A qualitative study of the challenges experienced by parents of children with disabilities. *Rehabilitation Psychology, 55*(2), 139.

Rogers, A. C. (1997). Vulnerability, health and health care. *Journal of Advanced Nursing, 26*(1), 65–72.

Rolfe, G. (2006). Validity, trustworthiness and rigour: Quality and the idea of qualitative research. *Journal of Advanced Nursing, 53*, 304–310.

Roy, A., Roy, A., & Roy, M. (2012). The human rights of women with intellectual disability. *Journal of the Royal Society of Medicine, 105*, 384–389. https://doi.org/10.1258/jrsm.2012.110303

Schalick, W., Westbrook, C., & Young, B. (2012). Communication with individuals with intellectual disabilities and psychiatric disabilities: A summary of the literature. *Michigan Retirement Research Center Research Paper*, 2012–2264.

Sheerin, F. K. (2011). Intellectual disability nursing–responding to health inequity. *British Journal of Learning Disabilities, 40*(4), 266–271.

Smith, L. J. (2008). How Ehtical is Ehtical research? Recruiting marginalized, vulnerable groups into health services research. *Journal of Advanced Nursing, 62*(2), 248–257.

Stein, M. A. (2007). Disability Human Rights. *California Law Review, 95*(1), 75–121.

Taua, C., Neville, C., & Hepworth, J. (2014). Research participation by people with intellectual disability and mental health issues: An examination of the processes of consent. *International Journal of Mental Health Nursing, 23*, 513–524.

The Office of the Public Advocate. (2014). A journey towards autonomy? Supported decision making in theory and practice. A review of the literature. Queensland: Queensland Government 22 November 2014.

United Nations Convention on the Rights of Persons with Disabilities (UNCRPD). (2006). [Online], Retrived from www.un.org/disabilities/convention/conventionfull.shtml. Accessed 15 June 2014

Veatch, R. M. (1972). Models for ethical medicine in a revolutionary age. *Hastings Center Report, 2*, 5.

Wark, S., MacPhail, C., McKay, K., & Müller, A. (2017). Informed consent in a vulnerable population group: Supporting individuals aging with intellectual disability to participate in developing their own health and support programs. *Australian Health Review, 41*(4), 436–442.

Werner, S., Tulchinsky, T. H., Gryga, I., Kovess-Masfety, V., Levav, I., & Pathare, S. (2012). Individuals with intellectual disabilities: A review of the literature on decision-making since the convention on the rights of people with disabilities (CRPD). *Public Health Reviews, 34*(2), 14.

Wiles, R. (2013). *What are qualitative research ethics?' In 'the what is?* (Research Methods Series). London: Bloomsbury Academic. https://doi.org/10.5040/9781849666558

Winston, A. S. (1998). Science in the service of the far right: Henry E. Garrett, the IAAEE, and the Liberty Lobby. *Journal of Social Issues, 54*(1), 179–210.

World Health Organisation (WHO). (2014). [Online], Retrived from http://www.unaids.org/sites/default/files/media_asset/201405_sterilization_en.pdf. Accessed 29 Apr 2017

Zero, D. T. (2004). Sugars: The arch criminal? *Caries Research, 38*(3), 277–285.

Research Priorities and Protections

15

Susana Gavidia-Payne and Mervyn Jackson

Introduction

Research can be simply defined as the establishment of a (new) knowledge base through the systematic application of agreed upon protocols. Approaches to research depend upon epistemologies, which vary considerably and require adaptation to specific questions as applied to particular populations. However, the creation of scientific knowledge should always follow ethical (or moral) principles. These principles govern the behaviour of researchers during all stages of the research process. They involve: the framing of research questions that benefit the target population; deciding upon a representative sample; ensuring participants have given informed consent (including the right to withdraw and protection from harm); using materials/test instruments that will generate reliable and valid knowledge; and, ensure responsible reporting of findings that lead to benefits for participants, the target population and the community/society as a whole. However, ever since research on humans began, problems with the ethical conduct of medical and behavioural researchers have continued. A clear example is highlighted by Beecher's (1966) paper on "Ethics and clinical research" (Jones, Grady, & Lederer, 2016). In his review of contemporary medical research, Beecher compiled a set of 50 empirical studies funded by US, Canadian or European Union Government agencies and published in leading medical journals (including the New England Journal of Medicine) that breached current ethical guidelines. While not specifically focusing on research involving people with intellectual disability, Beecher (1966) hoped that drawing attention to ethical problems would be sufficient for future researchers to address these issues (Jones et al., 2016). However, Jones et al. (2016) concluded that 50 years later, with ethical values changing over time, there is little consensus on what counts as ethical research; many conflicting interests in determining what is acceptable, ethical research still remain. The purpose of this chapter is to address the extra complexity of applying ethical principles to research involving people with intellectual disability, and the issues that continue to be posed for their participation in research that is ethical and significant to improve their quality of life. This chapter will review the history of abuse in the field, with a focus on ethical dilemmas emanating from past and contemporary research studies, including the implementation of safeguards used to protect people with intellectual disability. As research efforts are essential for improving the quality of life of people with intellectual disabilities, this chapter will discuss the prioritization of research themes in line with their level of acceptability.

S. Gavidia-Payne (✉) · M. Jackson
RMIT University, Melbourne, VIC, Australia
e-mail: susana.gavidia-payne@rmit.edu.au

J. L. Matson (ed.), *Handbook of Intellectual Disabilities*, Autism and Child Psychopathology Series, https://doi.org/10.1007/978-3-030-20843-1_15

The presentation of ethical dilemmas, conceptualized as those problematic situations where two or more principles or values conflict and more than one principle applies while there are good reasons to support mutually inconsistent courses of action (Kalvemark, Hoglund, Hanssona, Westerholmb, & Arnetza, 2004), has pervaded the history of human research. A classic illustration can be found in the series of social psychological studies carried out by Milgram in the 1960s where he suspended the participants' right to withdraw from experiments. To study obedience, Milgram developed a research paradigm where he placed subjects in a moral dilemma: Harm a fellow participant or defy the experimenter and withdraw from the experiment (Gridley, et al., 2017). To test defiance/obedience, Milgram provided the experimenter with standard prompts to be given to reluctant subjects ("the experiment requires you to continue, the next word is…"). This prevented subjects from terminating the experiment even though they clearly did not want to continue. The resolution of ethical dilemmas in research varies depending upon the dilemma and the experimental context. While the solution to some ethical dilemmas in research involves choosing the greater good/lesser evil solution, other ethical dilemmas are best resolved by case-based reasoning (Leake, 2001). Case-based reasoning can be applied to research involving people with intellectual disability where research has relied primarily on resolving the unique features arising from their diversity. In this way, Carlson (2013) has raised a number of ethical dilemmas associated with research involving people with an intellectual disability. These include: inclusion (and risk of exploitation) of people with intellectual disability vs. the exclusion (and discrimination) of people with an intellectual disability; the importance of informed consent vs. the inherent difficulty of obtaining informed consent; and the research focus on curing and preventing intellectual disability vs. affirming the right and quality of life of people with an intellectual disability to inhabit a shared world with people without disability.

Ethical dilemmas become more prominent as the *intellectual disability* classification (American Psychiatric Association, 2013) casts a wide net, encompassing individuals with a broad range of abilities and disabilities This complicates efforts to make generalizations about people with an intellectual disability as a single uniform group (Carlson, 2013). Hence, ethical considerations have to be adapted and qualified for each unique individual who is being considered as a research participant. The common characteristics of intellectual disability require that researchers need to carefully consider the challenges of working with this population, and with their participation, their various ethical issues that can arise. These may include: the capacity of the participant to make an independent informed decision to fully participate; the ability of the research team to simplify complex information (including research designs that involve placebos and no treatment controls); the manner in which this information is communicated (verbal, visual, etc.); the awareness that people with an intellectual disability may lack the experience of making independent decisions on their own behalf; the avoidance of any coercion or undue influence by researchers or other non-research people (parents, guardians) who may be involved directly (surrogate) or indirectly in decision-making; determining the risks/harm and benefits to the individual participant and not just the benefits to the research team or society; and, the continual monitoring through the whole research process to detect any discomfort/distress (resistance) that may directly or indirectly be displayed by the research participant indicating that they may no longer wish to participate (Bailey et al., 2014; Calveley, 2012; Iacono & Carling-Jenkins, 2012; Williams & Moore, 2011). This challenge is central as Carlson (2013) reported on a survey of clinical trials involving people with an intellectual disability on the US National Institute's registry. The survey revealed hundreds of studies involving participants with a variety of intellectual disability conditions (from broad designations of intellectual disability to specific conditions such as Down syndrome) using a wide variety of designs (random control trials, drug trials, genetic research and behavioural observations) into a variety of topics (including the nature and aetiology of disability to the impact of disability on various health conditions). With a wide variety of studies on a diverse population of people with intellectual disability, it is essential that all possible ethical concerns are explored and resolved.

Historical Context

There have been a number of remarkable reports on studies in which people with an intellectual disability have been subjected to unethical practices. These include (but not limited to) the following: World War II medical research; drug trials using live polio viruses; the failure to treat syphilis by the Tuskegee Institute; the purposeful infection of hepatitis at Willowbrook Sate Institution; and the use of children with intellectual disability in radioactive experiments. One of the most historically salient studies occurred during World War II in Nazi Germany, medical researchers completed many experiments on participants (including people with intellectual disability) without their consent and without any safeguards for their protection from harm (Annas & Grodin, 1992; Berg, 1996; Freedman, 2001). These medical researchers were put on trial after the war and as a consequence of these deliberations, the judges issued the Nuremberg Code of medical ethics, addressed further below. In another study in 1950, researchers reported feeding the live polio virus to 20 children with "mental retardation" at the Letchworth Village in New York State. The researchers did not request permission from the New York State Department of health (they later reported they believed the request would be denied) and there was no record in the original report of parental permission being sought or granted (Smith & Mitchell, 2001).

Originally called the Tuskegee study of untreated syphilis in the Negro male, this experiment began in 1932 by the US Public Health Service and the Tuskegee Institute, lasting for 40 years, and only terminated by a journalist whistle-blower (Dawson, 2008); it is considered one of the most unethical studies in history. The study comprised 600 participants (399 who had syphilis) and while promised free health treatment, none were treated even after the effective penicillin therapy became widely available. Participants were adult black males who were poor, had little education, were from rural areas, and many who were likely to be classified as having an intellectual disability.

Another infamous study took place at the Willowbrook State School (located on Staten Island, New York), which was a state supported institute housing children with an intellectual disability. In the first decade of operation (1950s) the institution become over-crowded and had frequent outbreaks of hepatitis. In order to understand the transmission and progress of this infectious disease, healthy children from Willowbrook were injected with various strains of hepatitis (Angell, 1992; Beecher, 1966; Krugman & Shapiro, 1971; Krugman, Ward, Giles, Bodansky, & Jacobs, 1959). While parents gave consent, later inquiries into this research found that they were not adequately informed about the risks/harm of these medical experiments (Freedman, 2001; Grodin & Glantz, 1994). Also at the level of state facilities, the (US) President's Advisory Committee on Human Radiation Experiments reported on a series of experiments using institutionalized children (with intellectual disability) at the Fernald and Wrentham Schools (West, 1998). The researchers exposed residents to radiation to investigate its effects on the human body's mineral absorption. While parents provided consent, they were not informed that the research involved ingestion of radioactive material (Advisory Committee, 1995: Freedman, 2001). Interestingly, the inquiry found that these experiments were conducted to the ethical standards of the day, but still violated the human rights of the participants (West, 1998). The direct result of each of these ethical abuses led to key inquiries with reports that helped frame current ethical guidelines into research involving people with intellectual disability.

Responses to Historical Events

In response to these ethical violations by researchers, a series of inquires reacted and responded by developing codes and guidelines for ethical researchers. These included the Nuremberg Code (Nuremberg Code, 1949); the Helsinki report (Declaration of Helsinki, 1964); the Belmont code (Belmont Report, 1978); and, the Common Rule. After World War II, a series of trials (known as the "Doctors' trial") were administered to hold German medical researchers accountable for conducting unethical medical procedures on prisoners (no informed consent, no voluntary participation, and no protection from harm). On advice from expert medical advisors to the prosecution, the three judges published a 10-point Nuremberg code of medical ethics. It included ethical principles such as need for informed consent; the absence of coercion; the principles of maleficence and benefice; properly formulated research paradigms; properly trained, qualified staff; and the participants' right to withdraw if they felt physically and mentally unable to continue. While the Nuremberg code was not officially adopted as law by any nation, it has been considered to be one of the most important documents in the history of clinical research ethics. It directly influenced the establishment of the Declaration of Helsinki and subsequent national regulations on research conduct in many nations including the USA, Canada, UK and Australia. Strict adherence to the Nuremberg Code could exclude the participation of many people with an intellectual disability from any scientific research, since the code clearly establishes that the voluntary and full informed consent of the human subject is absolutely essential (Glidden, 2008; Nuremberg Code, 1949). The powerful influence of researchers and parents/guardians, and the limited experience and understanding of participants with intellectual disability pose severe barriers to provide voluntary, informed consent (Berg, 1996: Clements, Rapley, & Cummins, 1999; Dalton & McVilly, 2004; Iacono, 2006; Weisstub & Arboleda-Flórez, 1997). While it must be recognized that some people may not be able to give informed consent, every effort should be made to ensure ethical practices are developed so that people with an intellectual disability will not be excluded from research (McCarthy, 1998).

The Declaration of Helsinki was developed by the World Medical Association and is a set of ethical principles regarding human experimentation. The declaration is morally binding on medical practitioners and its obligations override any national or local laws or regulations. The basic principles include: respect for the individual (participant); their right to self-determination; their right to make informed decisions; their right to fully participate or withdraw consent at any time; participant welfare must take precedence over the interests of science and society; and ethical considerations override national laws and regulations. There have been seven revisions (latest 2013) but many of the later revisions have been controversial and many nations (including USA and European Union) only recognise changes up to and including the third revision (1989) in their national legislation. The benefits of requiring researchers to follow internationally endorsed ethical standards such as the Declaration of Helsinki are evident. However, the Declaration of Helsinki states that some research populations are vulnerable and need special attention. Although vulnerability was never defined and there has been a tendency that the concern to protect vulnerable populations (for example, people with an intellectual disability) may lead gate keepers (ethics committees) preventing their inclusion in research studies. It is important that the protection of the rights of people with an intellectual disability is enhanced to the extent that it allows them to be ethically included in relevant research (Dalton & McVilly, 2004; Iacono, 2006).

In the USA, the Belmont Report (1978) summarizes ethical principles and guidelines for research involving human subjects and was created by the National Commission for the Protection of Human Subjects of Biomedical and Behavioral Research. Three core principles are identified: respect for persons (participants); beneficence; and social justice. The three primary areas of application are: Informed consent; assessments of risks and benefits; and, subject/

participant selection. In the USA, the Federal Policy for the Protection of Human Subjects (known as the "Common Rule") is based on the fundamental principles of the Belmont Report. However, community-based participatory researchers have criticized the Belmont Report because it is general in nature and does not take into account individual differences (including people with intellectual disability) (Shore, 2006).

The Nuremberg code (1949) and the Declaration of Helsinki (1964) form the basis for the ethical guidelines developed by various national government bodies including: National Institutes of Health (USA); National Health Service (UK); Medical Research Council (Canada); and, the National Health and Medical Research Council (Australia) (Iacono, 2006). However, these documents do not specifically address research involving people with an intellectual disability (Yan & Minur, 2004). Each document has separate sections for inclusion of people with an intellectual disability in research protocols, but none of the codes are sufficiently detailed to provide adequate guidelines to ensure the ethical inclusion of people with an intellectual disability in research. It must be noted that the rationale for these codes was in response to mainstream ethical dilemmas at the time and were not aimed to address the needs of specific populations such as people with an intellectual disability (Beecher, 1959; Curran, 1982; Musto, 1999; Perley, Fluss, Bankowski, & Simon, 1992; Yan & Minur, 2004).

Contemporary Ethical Dilemmas Involving People with Intellectual Disability

It has been widely acknowledged that people with intellectual disability played a very small to no role in decisions affecting their lives (Bailey et al., 2014; Freedman, 2001) with often incorrect assumptions about their cognitive capacity and functional competencies. It is then unsurprising that having a say about their participation in research endeavours may have also been considered problematic if not challenging to themselves, researchers and community alike, more particularly in the implementation of outcome-based research and genetic testing and therapy (Freedman, 2001). Investigations have then been fraught with difficulties, where consent, decision-making and risks and benefits of research participation of people with an intellectual disability have been amplified (Carlson, 2013; Iacono, 2006). The last 20 years, however, has seen an emphasis on self-determination, empowerment and self-advocacy by people with intellectual disability, all of which have brought to a head further dilemmas about whether, how and when people with an intellectual disability can become active research participants (Freedman, 2001; Stalker, 1998). While recent research has yielded further light in understanding these contentions from the perspectives of researchers and research participants with an intellectual disability (McDonald, 2012; Watchman, 2016), three issues remain complex and debatable, namely, decision-making, informed consent, and active participation in research endeavours. It must be noted that these ethical aspects of the research enterprise are closely interconnected in their execution, but for purposes of clarity, they will be treated separately in the present chapter.

Decision-Making

The process of decision-making is typically conceptualized as encompassing four components: (1) understanding relevant information; (2) appreciating a given situation and its consequences; (3) reasoning about available options; and (4) communicating a choice (Appelbaum, 2007). These features have been inextricably linked with the assessment of cognitive competence or capacity which may render an individual impaired and unable to understand information and act on the various available options. It is impairment associated with body function and structure as described in the International Classification of Functioning, Disability and Health [ICF; World Health Organization (WHO), 2013], that has been generally assumed to be the main barrier for people with an intellectual disability to make decisions about participating in research processes. This proposition is debatable considering that people with an intellectual disability do not constitute a homogenous group, and that intellectual disability is characterized by elements beyond cognitive impairment and affective states. As recognized by the ICF (WHO, 2013), it is possible that other experiences of a personal, social and environmental character such as lack of information and support, prior history in decision-making to participate research and some gate-keeping (McDonald, Conroy, Olick, and The Project ETHICS Expert Panel, 2017a) may be responsible for the limited participation of people with intellectual disability in research endeavours. This situation has given way to proposing forms of decision-making such as surrogate (Freedman, 2001), supportive (Iacono, 2006) to more actively participatory (Stack & McDonald, 2018). The latter has been favoured as more thoroughly serving the needs of people with an intellectual disability for inclusion, choice and participation as shown by McDonald, Conroy, Olick, and The Project ETHICS Expert Panel (2017b). The authors examined the perspectives of adults with intellectual disability, family members, researchers, disability service providers, and Institutional Review members about the perceived harmfulness of decision-making processes leading to research participation. All stakeholders agreed that surrogate decision-making was comparatively more harmful than other forms of decision-making. The authors concluded that the 'assent' expressed by people with intellectual disability in terms of their desires and thoughts should be a significant factor to be considered in the decision-making process to participate in research activities.

Variation in 'decisional capacities' for types of research has also emerged as a key point to consider beyond legal or other imperatives (Bailey et al., 2014). In their examination of parental ratings of decisional tasks of adults with Fragile X to participate in clinical trials, Bailey et al. (2014) reported great variability in decisional abilities, with 29% of adult males rated as not being able to complete decisional tasks as portrayed in a series of vignettes. Older people with Fragile X, whose parents were more willing for their child to participate in research studies, were predictors of greater participation in research. Recent studies also suggest that people with intellectual disability can and want to enact their preferences about participating in research with certain characteristics above others. They wish to make decisions about their participation, especially in research that involves positive social interactions, increased agency and confidentiality (McDonald et al., 2017a). It is then possible that a number of factors may be influencing the decisional capacity to participate in research, ranging from individual to more contextual in nature. Consensus on what constitutes an appropriate threshold for decisional capacity for varying types of research (Bailey et al., 2014; Freedman, 2001) is yet to be reached. Further research in this area can assist in providing a framework for 'degrees' of levels of participation in different types and aspects of research studies (e.g. medical vs. social research) according to the decision-making characteristics of people with intellectual disability. This may be a useful approach for ethics review boards to address not only ethical requirements and place appropriate safeguards in place, but also to be in line with the claims of people with intellectual disability for independence and autonomy.

Consent

Consent to participate in research has been inextricably linked to the notion of cognitive capacity or competency of people with intellectual disability, generating questions about whose responsibility it is to determine such capacity to provide informed consent (Iacono, 2006). While acknowledging that tensions and dilemmas around this issue continue to exist (Morgan, Cuskelly, & Moni, 2014), it is assumed that the right for autonomy and self-determination of people with intellectual disability is central to consenting to research participation. The question then arises, how would this look in practice? The operationalization of the process of consent can assist in teasing out how people with disability engage with consent provision. Specifically, this may mean ascertaining how people with intellectual disability vary in the extent to which they understand the information provided; appreciate different options and their consequences (risks and benefits), and are able to communicate a choice (Appelbaum, 2007). Horner-Johnson and Bailey (2013) tested these notions by examining the answers of people with intellectual disability to questions about key aspects of the consent process associated with a health promotion study. It was revealed that while more than half of participants correctly answered all of the questions, the remaining could not, with identifying potential risks of being in the study proving the most challenging. Horner-Johnson and Bailey concluded that many people with intellectual disabilities can provide their own consent to participate in low risk studies. However, the authors also raised the question of whether consent to participate in research studies is truly informed and understood as it has been argued elsewhere (Flory & Emanuel, 2004). A number of characteristics and elements of research studies, such as level of risk, design and topic, may be influencing the extent to which people with intellectual disability understand information that allows them to provide consent to participation in research events. Conceptualizing and operationalizing informed consent as a process, as opposed to a ‘one-off’ event, may be a more effective way of increasing its effectiveness.

The use of intervention strategies to improve understanding of informed consent has received some attention in the literature, particularly in regards to participation in clinical trials. Flory and Emanuel (2004) conducted a review of 30 studies covering 42 clinical trials which utilized interventions to improve consent understanding in various groups of hospital patients with a range of medical diagnoses, some of whom presented with cognitive impairment. In those studies, limited improvement in understanding using multimedia interventions and enhanced consent forms, and significant improvement through extended interactions between research staff and participants were reported. Participants with lower educational attainment and mental illness also showed lower understanding of the consent process. Similarly, in a recent review of audio-visual presentation of information for informed consent for participation in clinical trials with populations of children and people deemed not to be competent to give consent, Synnot, Ryan, Prictor, Fetherstonhaugh, and Parker (2014) found low quality evidence of the effect of such interventions on enhancing the information consent process.

It has then been proposed that in order to promote the understanding of informed consent, it may be necessary to utilise different tools according to an assessment of the functional characteristics of people with disability as well as the features of the research study. Horner-Johnson and Bailey (2013) suggest to diversifying the demands on consenting depending on the level of risk of the study, with lower risk studies requiring only some evidence of understanding as a sign for an individual to provide consent. Some authors assert (Bailey et al., 2014; Flory & Emanuel, 2004) that one-to-one encounters which are flexible and extended in their discussion of the study, while at the same time responding to the specific needs (emotional or otherwise) of participants, are most promising in facilitating understanding of consenting to research. More than action in itself, this premise implies that the provision of consent be conceptualized as a relational process so appropriate strategies are developed with deliberate attempts for environmental and contextual changes to facilitate consent (Bailey et al., 2014; Carlson, 2013; McDonald et al., 2017a).

Participation

Historically, the research participation of people with intellectual disability has been characterized by harm and abuse, a fact that inevitably turned the pendulum towards a view that rendered them vulnerable and in need of protection. This paternalistic premise, however, has been challenged by more contemporary efforts of people with intellectual disability for inclusion, self-determination and autonomy, with calls for their increased research participation (Nuwagaba & Rule, 2015). In discussing the risks of inclusion and exclusion of people with intellectual disability in research, Carlson (2013) maintains that researchers need to engage with critical disability perspectives, whereby common assumptions about what constitutes research involvement are challenged, and informed by careful deliberations about the interactions and power dynamics among people with disability, family members, researchers and ethics review boards. It follows that the various factors that can affect the research participation of people with intellectual disability, are similar to those influencing the process of consent, and therefore need to be evaluated in the 'real world'. This issue was examined by Cleaver, Ouellette-Kuntz, and Sakar (2010) who reported that greater research participation was associated with researcher's direct access to participants and where data collection procedures were non-invasive. These findings indicate, as has been suggested elsewhere (Nuwagaba & Rule, 2015), that diminished research participation may have more to do with contextual and cultural issues instead of the inherent characteristics of people with intellectual disability.

Risks, Benefits and Protections Associated with Participation

The inclusion and participation of people with intellectual disability in research events implies a weighing up of associated risks and benefits. Their identification, however, has been suggested to be a value-laden and contested process (Carlson, 2013). The diversity of views of key stakeholders such as families, researchers, ethics review boards, children and adults with intellectual disability themselves add complexity to the assessment of risks and benefits. Risk assessments in particular, despite requirements of ethical review board assessments, are not straight forward (Carlson, 2013; Iacono, 2006; Nuwagaba & Rule, 2015) as they are often at odds with the desires of people with intellectual disability to participate in research (McDonald, 2012). Within the context of a participatory research paradigm, Morgan et al. (2014) experienced unanticipated tensions and ethical dilemmas about conceptualizations of intellectual disability, role of the researchers, transparency, equality, and changes to original ethics agreements. Morgan et al.'s (2014) reflections highlight the unforeseen risks posed to both participants and researchers as they embark in the research process as well as the possibility that such risks can have detrimental effects on the well-being of all concerned.

Benefits of research participation, on the other hand, have strong support in the literature from a number of perspectives. First, participation of people with intellectual disability in research activities endorses their rights, dignity and autonomy, considering their inclusion a matter of justice (McDonald et al., 2016). Second, it adds value to the research process, providing enriched perspectives, as alternatives to the more academic and often considered rigid views of researchers (McDonald et al., 2016; Woelders, Abma, Visser, & Schipper, 2015). Third, research participation of people with intellectual disability contributes to their feeling valued and empowered, providing opportunities to demonstrate their abilities, learning from the research experience, and helping others (McDonald et al., 2016). Fourth, their representation, particularly in health research, is necessary to improve outcomes and ameliorate the influence of risk factors such as chronic conditions and poverty (Rios, Magasi, Novak, & Harniss, 2016). All these benefits can be achieved when characterization of the research process is effectively inclusive and collaborative, and where people with intellectual disability are considered research team members as opposed to research subjects (Woelders et al., 2015).

Discussion about the protection of people with intellectual disability when participating in research events has inevitably raised the notion of vulnerability. This concept is generally used as the main premise to protect them against risk, and offering of guarantees of well-being during their research participation. Safeguards against potential risks are crucial in this context, and informed consent has generally been the tool through which protections have been enforced by ethical review boards. This regulatory process, however, has been called into question (Iacono, 2006; McDonald et al., 2016); Nuwagaba & Rule, 2015) as decisions based on perceived risk, benefits and safety will vary according to whom is defining them, and may not necessarily represent the views of the community, including people with intellectual disability. For example, people with intellectual disability see 'gatekeeping' as more harmful than family members and ethical review board members (McDonald et al., 2017b).

Recent evidence also indicates that perceptions of safety converge and diverge on a number of fronts, while people with intellectual disability concur with others on focal safeguards for decision-making, they do not on issues of recruitment. Specifically, they perceive to be in greatest safety when recruited by somebody they trust, and when researchers ensure participants' own decision-making (McDonald et al., 2016). It can then be argued that people with intellectual disability may not necessarily feel they are in a vulnerable position, and that they are capable of making decisions about their participation in research on their own right. Researchers could support demystifying the issue of vulnerability by educating ethical review boards and others in the community, as well as introducing modifications to certain elements of the research process to ensure safety in keeping with ethical principles. Ethical and methodological dilemmas can be best navigated with flexibility in data collection and communication methods with people with intellectual disability (Watchman, 2016). On a similar vein, ethical review boards could promote disability ethical procedures that are disability friendly and contextualized (Carlson, 2013; Nuwagaba & Rule, 2015).

Moving Forward: Research Priorities

Consensus exists regarding the need to include people with intellectual disability in research, upholding ethical principles for their safe participation, as well as maintaining academic rigour in conducting research. The scientific community has been urged to take an increased role in providing creative solutions for the inclusion of people with intellectual disability, their families and communities, and effectively addressing decision-making, consent process and research design issues (Dalton & McVilly 2004; Iacono & Carling-Jenkins, 2012; Northway, 2014). To this end, the establishment of partnerships among researchers and people with intellectual disability, and creation of protocols are all laudable aims to have (Carlson, 2013). Importantly, however, advancing the work in this area also necessitates a continued focus on 'researching research processes' and the establishment of priorities within these. The current strengths and gaps observed in the current state of the field begets the formulation of research directions that translate available knowledge into practical outcomes that address the ethical dilemmas that have plagued the field, as well as methodologies that can progress our understanding and knowledge about the meaningful inclusion people with intellectual disability in research activities (Watchman, 2016).

Contemporary literature as a whole identifies various research themes and policy work around ethical conduct in researching with people with intellectual disability. They include: promoting understanding of the ethical issues of decisional capacity, informed consent, and actual participation among people with intellectual disability; qualities of the research process to maximize participation, including the researcher's role; characteristics of research methodologies; the role of key stakeholders in supporting safe and meaningful participation; and themes not deemed suitable to be researched. First, working with people with intellectual disability to support their understanding of key ethical matters related to the research process (i.e. decisional capacity, provision of consent, actual participation) through the development of validated assessment and intervention support models has been identified as an important research target (Bailey et al., 2014; McDonald et al. 2017a, 2017b). Bailey et al. (2014) suggests that while these issues have been addressed with other populations, limited work has been done with people with intellectual disability whose experiences may be largely different and varied relative to people without disabilities. Limited opportunities for decision-making given their life circumstances have precluded them from exercising competencies, typically involving problem-solving, evaluation of risks and benefits, and consent to research participation. The usefulness and ethical integrity of some models of support for decision-making to participate in research studies above others also need to be evaluated (McDonald et al., 2017a). Further looking beyond the disability type, and instead focussing on the analysis of the various components of decision-making to participate in research, is necessary. In this regard, testing the use of new technologies, visual media, augmentative communication devices and so forth can generate evidence about factors that can potentially influence the ethical aspects of participation, as well as their usefulness in supporting models that promote understanding of risk and benefits of research participation in actual practice (McDonald et al., 2017b; Synnot et al., 2014).

Second, the qualities of the research enterprise that can be adapted to ensure the participation of people with intellectual disability can be posed as another research priority. As the planner, designer, implementer, analyst and interpreter of findings, the role of the researcher is crucial to the establishment of a research context that is conducive to maximizing the contribution of people with intellectual disability. The shift towards modifying elements of the research process may mean the adoption of research models that may not necessarily be consistent with academic control and institutional demands, and which may also pose some challenges for researchers. Furthermore, Woelders et al. (2015) argues that researchers need to move beyond the notion that they are the only authority in adjudicating 'validity', as we know it, to the research process. Investigation of views about the role of the researcher as an advocate and educator with ethical academic boards, policy-makers and other community stakeholders can also provide further impetus for policy re-development regarding the safe participation of people with intellectual disability (McDonald, Schwartz, Gibbons, & Olick, 2015). At a more 'micro' level, conversational and interpersonal encounters between researchers, participants and key stakeholders, that are characterized as respectful, positive, empathetic, and over a period of time have support in the literature (Carey & Griffiths, 2017; Carlson, 2013; McDonald, 2012; McDonald et al., 2016). Eliciting their views in the form of values, preferences, and concerns is central to supporting significant contributions by people with intellectual disability to the research process. They want to contribute to research and indicate the particular ways they want to do so, including the accommodations that need to be made such as in how research aims are communicated, the clarity of verbalizations used, to the settings where research interviews are conducted, and compensation for participation if appropriate (McDonald, 2012). Bigby, Frawley, and Ramcharan (2014) argue that there is more than one way to do inclusive and participatory research with people with intellectual disability while cautioning us against making universal and rigid prescriptions about its meaning for different groups and stakeholders. Despite these understandings, the evidence base for a model of support for inclusive and ethical research that encompasses all these qualities is yet to be developed. Such a model can offer the foundations to set ethical standards for optimal research processes from the outset, ranging from the conceptualization of a study and formulation of research questions, to the selection and implementation of methodologies to the dissemination of findings.

Third, some research methodologies may be more conducive to the meaningful participation of people with intellectual disability beyond the ethical considerations involved. Qualitative research designs, including participatory research models (Beighton et al., 2017; McDonald, 2012) as well as those following a grounded theory approach (Carey & Griffiths, 2017) have been reported to promote their active research participation. Interviews using open-ended questioning and delivered on a one-to-one basis offer the added benefit of capturing any risks associated with research participation while strengthening the richness of findings, especially when it comes to working with children and young people (Lewis

& Porter, 2004). Participatory action research designs may convey more accurately the nuanced expressions of people with intellectual disability, their thoughts, feelings and behaviours (Inglis & Cook, 2011). Comparative studies of specific costs and benefits of quantitative and qualitative research designs in terms of funding, time, labour intensity, and stakeholder involvement would also add to this evidence-base. Research quality can also be improved with further investigation into emerging research models that actively promote the empowerment of people with intellectual disability such as the community-based participatory approach (Stack & McDonald, 2018). Notions of 'co-creation' and 'co-design' of knowledge using modern technology (e.g. online communities) can also be examined to determine their usefulness for further collaboration and inclusion in research of people with intellectual disability (Amann & Rubinell, 2017).

Fourth, the role of key stakeholders in supporting safe and meaningful participation is fundamental to the inclusion of people with intellectual disability in research undertakings. Particular consideration should be given to already documented findings about the differential perceptions of key stakeholders, and the 'gatekeeping' that can occur in the name of protection against harm (McDonald et al., 2016). In the case of children with intellectual disability, the evaluation of parental views about the risks they encountered and benefits that they and their children may gain from research participation is also critical in informing the design of more relevant and valued research studies. Bailey et al. (2014) adds that research into parental perceptions of children's participation throws further light into the specific role parents may wish to play as advocates in research conceptualization, design and dissemination. It would also be important to canvass the perspectives of service providers and other care staff as sources of support for research participation, and their specific role and contribution in the development and implementation of research studies. Apart from investigations by McDonald and colleagues, little attention has been devoted to this area. Last but not least is the role of ethical review boards as key stakeholders in safeguarding people with intellectual disability. Much has been written about the labelling of people with an intellectual disability as a homogeneous and vulnerable group in need of protection, a view commonly held by ethical review boards. Yet, evidence indicates that stringent ethical requirements have failed to safeguard them effectively, and in fact have added greater complexity for researchers and people with intellectual disability to fully participate in research activities (Iacono & Carling-Jenkins, 2012). The role that researchers can play in educating ethical review boards about intellectual disability, and the reported alternatives to meaningfully involve them in research deserves attention as a possible research and policy development focus.

Fifth, several research themes, generally in the realm of investigations of interventions with people with intellectual disability, are considered to be too dangerous to be researched. Illustrations of interventions in this category shown to be associated with possible harm include chelation therapy (Mitka, 2008; Sinha, Silove, & Williams, 2006), and facilitated communication (Committee on Children with Disabilities, 1998). Interventions (and the research behind it) that may lead to further discrimination and promote categorization, stigma and exclusion have long been judged as deleterious to improving the well-being and quality of life of people with intellectual disability, and considered ethically questionable. For example, should the use of comparative groups (e.g. people with intellectual and disability and their counterparts without a disability) be an acceptable research methodology given its potential to characterize one group 'better' than the other? Ultimately, in making decisions related to the topics to investigate, good research practice demands that not only attention be given to the evidence-base behind such interventions, but also the values that guide those practices (Stoneman, 2009). In this way, the development and evaluation of a system with thresholds along the continuum of acceptability-non acceptability in research practices with children and adults with intellectual disability can be argued to be a research priority. This can assist in preventing the conduct of research that is to this day, still unacceptable and unethical (see ProPublica Illinois, Cohen, 2018). This task may demand more than just the stipulation of intervention procedures that are acceptable/non acceptable because of the possibility of dangerous outcomes. It should also include the operationalization of all possible components of the research enterprise, possible risks, and concomitant adjustments that support the ethical integrity of the study, and hence the inclusion of people with intellectual disability.

In summary, several lines of research have been identified that address questions with the aim of achieving the ethical research inclusion and participation of people with intellectual disability. The range of possible issues to investigate is ample; some are associated with the effective implementation of protections in research practice, while others relate to factors that facilitate or hinder inclusion of people with intellectual disability while maintaining adequate ethical safeguards. The knowledge basis on which to increase evidence that supports this objective already exists (e.g. Dalton & McVilly, 2004; Inglis & Cook, 2011; McDonald et al., 2015) and is well-placed to begin the development of systematic models of interventions and supports that can inform researchers, policy makers, people with intellectual disability and their communities.

Conclusion

Research experiences of people with intellectual disability in the past were characterized by abuse and stigmatization. As a result, they have endured marginalization in terms of inclusion in research activities. It is recognized that a balance must be reached where ethical research integrity is upheld while at the same time academic rigour is maintained in the conduct of research. This state of affairs still continues to offer a number of dilemmas where various stakeholders have a role to play beyond the person with intellectual disability. Central to this predicament is the relationship between researchers and people with intellectual disability, whereby collaborative, strength-based interactions must prevail. Along these lines, researchers need to ensure that their studies are carefully planned, while allowing for time to recruit and collect data rigorously in a sensitive and respectful manner. However, this, in addition to the demands on researchers to meet peer-reviewed publication quotas and dissemination (which could be considered essentially academic concerns), may not necessarily be consistent with promising models, which espouse the empowering qualities of research studies that may ultimately facilitate the research participation of people with intellectual disability. Researchers have similarly an important role to play in advocating for the increased research inclusion of people with intellectual disability in such a way that notions of vulnerability and risk are debated as a function of growing evidence in favour of their rights and wishes. The research priorities earlier proposed constitute only some of several potential research avenues that can be pursued. It is argued that collaborating with, including and enabling people with intellectual disability with knowledge, resources, and adaptations during the implementation of research can ultimately minimise and even prevent the use of unethical procedures.

References

Advisory Committee. (1995). *Final report of the advisory committee on human radiation experiments*. Washington, DC: U.S. Government Printing Office.

Amann, J., & Rubinell, S. (2017). Views of community managers on knowledge co-creation in online communities for people with disabilities: Qualitative study. *Journal of Medical Internet Research, 19*(10), e320. https://doi.org/10.2196/jmir.7406

American Psychiatric Association. (2013). *Diagnostic and statistical manual of mental disorders* (5th ed.). Washington, DC: Author.

Angell, M. (1992). Editorial responsibility: Protecting human rights by restricting publication of unethical research. In G. Annas & M. Grodin (Eds.), *The Nazi doctors and the Nuremberg code* (pp. 276–285). New York: Oxford University Press.

Annas, G., & Grodin, M. (Eds.). (1992). *The Nazi doctors and the Nuremberg code* (pp. 276–285). New York: Oxford University Press.

Appelbaum, P. S. (2007). Assessment of Patients' Competence to Consent to Treatment. *The New England Journal of Medicine, 357*, 1834–1840.

Bailey, D., Raspa, M., Wheeler, A., Edwards, A., Bishop, E., Bann, C., … Appelbaum, P. (2014). Parent ratings of ability to consent for clinical trials in Fragile

X Syndrome. *Journal of Empirical Research on Human Research Ethics., 9*(3), 18–28. https://doi.org/10.1177/1556264614540591

Beecher, H. (1959). Experimentation in man. *Journal of the American Medical Association., 169*, 461–478.

Beecher, H. (1966). Ethics and clinical research. *New England Journal of Medicine., 274*, 1354–1360.

Beighton, C., Victor, C., Carey, I. M., Hosking, F., DeWilde, S., Cook, D. G., … Harris, T. (2017). I'm sure we made it a better study…': Experiences of adults with intellectual disabilities and parent carers of patient and public involvement in a health research study. *Journal of Intellectual Disabilities*, 1–19. https://doi.org/10.1177/1744629517723485

Belmont Report. (1978). *Ethical Principles and Guidelines for the Protection of Human Subjects of Research*. Retrieved from https://www.hhs.gov/ohrp/regulations-and-policy/belmont-report/index.html

Berg, J. (1996). Legal and ethical complexities of consent with cognitively impaired research subjects: Proposed guidelines. *The Journal of Law, Medicine & Ethics, 24*(1), 18–35.

Bigby, C., Frawley, P., & Ramcharan, P. (2014). Conceptualizing inclusive research with people with intellectual disability. *Journal of Applied Research in Intellectual Disabilities, 27*, 3–12.

Calveley, J. (2012). Including adults with intellectual disabilities who lack capacity to consent in research. *Nursing Ethics, 19*, 558–567. https://doi.org/10.1177/0969733011426818

Carey, E., & Griffiths, C. (2017). Recruitment and consent of adults with intellectual disabilities in a classic grounded theory research study: Ethical and methodological considerations. *Disability & Society, 32*(2), 193–212. https://doi.org/10.1080/09687599.2017.1281793

Carlson, L. (2013). Research ethics and intellectual disability: Broadening the debates. *The Yale Journal of Biology and Medicine, 86*(3), 303.

Cleaver, S., Ouellette-Kuntz, H., & Sakar, A. (2010). Participation in intellectual disability research: A review of 20 years of studies. *Journal of Intellectual Disability Research, 54*(3), 187–193. https://doi.org/10.1111/j.1365-2788.2010.01256.x

Clements, J., Rapley, M., & Cummins, R. A. (1999). On, to, for, with–vulnerable people and the practices of the research community. *Behavioural and Cognitive Psychotherapy, 27*(2), 103–115.

Cohen, J.S. (2018, April, 26). The $3 Million Research Breakdown. Retrieved from https://www.propublica.org/article/university-of-illinois-chicago-mani-pavuluri-3-million-research-breakdown

Committee on Children with Disabilities. (1998). Auditory integration training and facilitated communication for autism. *Pediatrics, 102*(2), 431–433.

Curran, W. (1982). Subject consent requirements in clinical research: An international perspective for industrial and developing countries. In Z. Bankowski & N. Howard-Jones (Eds.), *Human experimentation and medical ethics* (pp. 35–79). Geneva, Switzerland: Council for International Organizations of Medical Sciences.

Dalton, A., & McVilly, K. (2004). Ethics guidelines for international, multicenter research involving people with intellectual disabilities. *Journal of Policy and Practice in Intellectual Disabilities, 1*(2), 57–70.

Dawson, G. (2008). Ethics in medical research. In G. Dawson (Ed.), *Easy interpretation of biostatistics: The vital link to applying evidence in medical decisions* (pp. 128–132). Philadelphia, PA: Elsevier.

Declaration of Helsinki (1964). Recommendations guiding doctors in clinical research. Adopted by the 18th World Medical Assembly, Helsinki, Finland, June 1964. Retrieved form https://www.wma.net/wp-content/uploads/2016/11/DoH-Jun1964.pdf

Flory, J. H., & Emanuel, E. (2004). Interventions to improve research participants' understanding in informed consent for research: A systematic review. *The Journal of the American Medical Association, 292*(13), 1593–1601. https://doi.org/10.1001/jama.292.13.1593

Freedman, R. I. (2001). Ethical challenges in the conduct of research involving persons with mental retardation. *Mental Retardation, 39*(2), 130–141.

Glidden, L. (2008). *International Review of Research into Mental Retardation* (Vol. 35). Amsterdam: Elsevier.

Gridley, M., Jenkins, W., Adorno, T., Frenkel-Brunswik, E., Levinson, D. J., Sanford, R. N., … Bandura, A. (2017). Controversial psychological research methods and their influence on the development of formal ethical guidelines. In *An analysis of Stanley Milgram's obedience to authority: An experimental view* (Vol. 70, pp. 9), 9–9),13). New York: Institute of Group Relations, University of Oklahoma.

Grodin, M., & Glantz, L. (1994). *Children as research subjects: Science, ethics, and law*. New York: Oxford University Press.

Horner-Johnson, W., & Bailey, D. (2013). Assessing understanding and obtaining consent from adults with intellectual disabilities for a health promotion. *Journal of Policy and Practice in Intellectual Disabilities, 10*(3), 260–265.

Iacono, T. (2006). Ethical challenges and complexities of including people with intellectual disability as participants in research. *Journal of Intellectual & Developmental Disability, 31*(3), 173–179. https://doi.org/10.1080/13668250600876392

Iacono, T., & Carling-Jenkins, R. (2012). The human rights context for ethical requirements for involving people with intellectual disability in medical research. *Journal of Intellectual Disability Research, 56*, 1122–1132. https://doi.org/10.1111/j.1365-2788.2012.01617.x

Inglis, P., & Cook, T. (2011). Ten top tips for effectively involving people with a learning disability in research. *Journal of Learning Disabilities and Offending Behaviour, 2*(2), 98–104. https://doi.org/10.1108/20420921111152441

Jones, D., Grady, C., & Lederer, S. (2016). "Ethics and clinical research"—the 50th anniversary of Beecher's bombshell. *New England Journal of Medicine, 374*(24), 2393–2398.

Kalvemark, S., Hoglund, A. T., Hanssona, M. G., Westerholmb, P., & Arnetza, B. (2004). Living with conflicts-ethical dilemmas and moral distress in the health care system. *Social Science & Medicine, 58*, 1075–1084. https://doi.org/10.1016/S0277-9536(03)00279-X

Krugman, S., & Shapiro, S. (1971). Experiments at the Willowbrook State School. *The Lancet, 297*(7706), 966–967.

Krugman, S., Ward, R., Giles, J., Bodansky, O., & Jacobs, A. (1959). Infectious hepatitis: Detection of the virus during the incubation period and in clinically in apparent infection. *New England Journal of Medicine., 261*, 729–734.

Leake, D. B. (2001). Problem solving and reasoning: Case-based. *International Encyclopedia of the Social & Behavioral Sciences*, 12117–12120. https://doi.org/10.1016/B0-08-043076-7/00545-3

Lewis, A., & Porter, J. (2004). Interviewing children and young people with learning disabilities: guidelines for researchers and multi-professional practice. *British Journal of Learning Disabilities, 32*, 191–197.

McCarthy, M. (1998). Interviewing people with learning disabilities about sensitive topics: A discussion of ethical issues. *British Journal of Learning Disabilities, 26*(4), 140–145.

McDonald, K. (2012). "We want respect": Adults with intellectual and developmental disabilities address respect in research. *American Journal on Intellectual and Developmental Disabilities, 117*(4), 263–274. https://doi.org/10.1352/1944-7558-117.4.263

McDonald, K. E., Conroy, N. E., Kim, C. I., LoBraico, E. J., Prather, E. M., & Olick, R. S. (2016). Is safety in the eye of the beholder? Safeguards in research with adults with intellectual disability. *Journal of Empirical Research on Human Research Ethics, 11*(5), 424–438.

McDonald, K. E., Conroy, N. E., Olick, R. S., & The Project ETHICS Expert Panel. (2017a). A quantitative study of attitudes toward the research participation of adults with intellectual disability: Do stakeholders agree? *Disability and Health Journal, 11*(3), 345–350. https://doi.org/10.1016/j.dhjo.2017.12.004

McDonald, K. E., Conroy, N. E., Olick, R. S., & The Project ETHICS Expert Panel (Eds.). (2017b). What's the harm? Harms in research with adults with intellectual disability. *American Journal on Intellectual and Developmental Disabilities, 122*(1), 78–92. https://doi.org/10.1352/1944-7558-122.1.78

McDonald, K. E., Schwartz, N. M., Gibbons, C. M., & Olick, R. S. (2015). "You can't be cold and scientific" Community views on ethical issues in intellectual disability research. *Journal of Empirical Research on Human Research Ethics, 10*(2), 196–208.

Mitka, M. (2008). Chelation therapy trials halted. *Journal of the American Medical Association, 300*(19), 2236. https://doi.org/10.1001/jama.2008.607

Morgan, M. F., Cuskelly, M., & Moni, K. B. (2014). Unanticipated ethical issues in a participatory research project with individuals with intellectual disability. *Disability & Society, 29*(8), 1305–1318.

Musto, D. (1999). A historical perspective. In S. Bloch, P. Chodoff, & S. Green (Eds.), *Psychiatric ethics 3* (pp. 7–23). New York: Oxford University Press.

Northway, R. (2014). Editorial: To include or not to include. *Journal of Intellectual Disabilities., 18*(3), 209–210.

Nuremberg Code. (1949). *Trials of War Criminals before the Nuremberg Military Tribunals under Control Council Law No. 10* (Vol. 2, pp. 181–182). Washington, DC: U.S. Government Printing Office. Retrieved from https://history.nih.gov/research/downloads/nuremberg.pdf

Nuwagaba, E. L., & Rule, P. (2015). Navigating the ethical maze in disability research: Ethical contestations in an African context. *Disability & Society, 30*(2), 255–269.

Perley, S., Fluss, S., Bankowski, Z., & Simon, F. (1992). The Nuremberg Code: An international overview. In G. Annas & M. Grodin (Eds.), *The Nazi doctors and the Nuremberg Code* (pp. 157–158). New York: Oxford University Press.

Rios, D., Magasi, S., Novak, C. & Harniss, M. (2016). Conducting accessible research: Including people with disabilities in public health, epidemiological, and outcomes studies. *American Journal of Public Health, 106*(12), 2137–2144.

Shore, N. (2006). Re-conceptualizing the Belmont Report: A community-based participatory research perspective. *Journal of Community Practice., 14*(4), 5–26. https://doi.org/10.1300/J125v14n0402

Sinha, Y., Silove, N., & Williams, K. (2006). Chelation therapy and autism. *British Medical Journal, 333*(7571), 756. https://doi.org/10.1136/bmj.333.7571.756

Smith, D., & Mitchell, A. (2001). Sacrifices for the Miracle: The polio vaccine research and children with mental retardation. *Mental Retardation, 39*(5), 405–409.

Stack, E. E., & McDonald, K. (2018). We are "both in charge, the academics and self-advocates": Empowerment in community-based participatory research. *Journal of Policy and Practice in Intellectual Disabilities, 15*(1), 80–89. https://doi.org/10.1111/jppi.12236

Stalker, K. (1998). Some Ethical and Methodological Issues in Research with People with Learning Difficulties. *Disability & Society, 13*(1), 5–19. https://doi.org/10.1080/09687599826885Stack

Stoneman, Z. (2009). Disability research methodology: Current issues and future challenges. In S. L. Odom, R. H. Horner, M. E. Snell, & J. Blacher (Eds.), *Handbook of developmental disabilities* (pp. 35–44). New York: Guilford Press.

Synnot, A., Ryan, R., Prictor, M., Fetherstonhaugh, D., & Parker, B. (2014). Audio-visual presentation of information for informed consent for participation in clinical trials. *Cochrane Database of Systematic Reviews, 2014*(5), CD003717. https://doi.org/10.1002/14651858.CD003717.pub3

Watchman, K. (2016). Investigating the lived experience of people with Down syndrome with Dementia: Overcoming Methodological and Ethical Challenges. *Journal of Policy and Practice in Intellectual Disabilities, 13*(2), 190–198. https://doi.org/10.1111/jppi.12167

Weisstub, D., & Arboleda-Flórez, J. (1997). Ethical research with the developmentally disabled. *The Canadian Journal of Psychiatry, 42*(5), 492–496.

West, D. (1998). Radiation experiments on children at the Fernald and Wrentham schools: Lessons for protocols in human subject research. *Accountability in Research, 6*(1–2), 103–125.

Williams, A., & Moore, S. (2011). Universal design of research: Inclusion of persons with disabilities in mainstream biomedical studies. *Science Translational Medicine, 3*(82), 82cm12.

Woelders, S., Abma, T., Visser, T., & Schipper, K. (2015). The power of difference in inclusive research. *Disability & Society, 30*(4), 528–542. https://doi.org/10.1080/09687599.2015.1031880

World Health Organization. (2013). *How to use the ICF: A practical manual for using the International Classification of Functioning, Disability and Health (ICF). Exposure draft for comment. October 2013*. Geneva: WHO.

Yan, E., & Minur, K. (2004). Regulatory and ethical principles in research involving children and individuals with developmental disabilities. *Ethics Behavior, 14*, 31–49.

Restraint and Seclusion

16

Timothy R. Vollmer, Meghan A. Deshais, and Eliana M. Pizarro

Introduction

For the purposes of this discussion, *Restraint* is defined as physically holding or securing an individual, either for a brief time period to interrupt severe problem behavior or for an extended period of time using physical holds or mechanical devices to prevent dangerous behavior (Vollmer et al., 2011). *Seclusion* is defined as isolating an individual from others to interrupt problem behavior that places others at risk of harm (Vollmer et al.). It is recognized that not all professions or organizations use these terms in the same way, but some guiding and defining characteristics are necessary to present a fruitful discussion. Deviations from such usage will be pointed out as we discuss various position statements and regulations.

Historically, restraint and seclusion were used as methods to either protect individuals from harming themselves or others, or in some cases to punish harmful behavior (Metzner et al., 2007). In more recent times, restraint and seclusion are usually viewed primarily as protective measures designed to keep an individual from doing harm to self or others. It is recognized that restraint and seclusion can at times serve a punishing function (i.e., decrease the probability of the future occurrence of behavior), but that is not usually the primary purpose of contemporary usage. Though the ethical position of most professionals in autism and intellectual disabilities is that these procedures may be necessary at times, it is the role of the professional to protect individuals from misuse of restraint and seclusion. In this chapter we will provide some historical perspective on the use of restraint and seclusion, sample some position statements from various professional organizations that seem to confirm this ethical stance, and examine some state regulations. Finally, we will outline best practices as gleaned from the literature.

History

During the eighteenth and nineteenth centuries, acute and long-term psychiatric hospitals emerged and flourished in Europe and the United States. Throughout this period, institutionalization of individuals with mental illness, intellectual disabilities, or both was regarded as a humane practice that prevented those individuals from posing a danger to themselves or others. Institutionalization became so widespread that overcrowding became a significant concern (Colaizzi, 2005). Restraint and seclusion procedures were routinely used by staff to physically manage patients and as treatments for challenging

T. R. Vollmer (✉) · M. A. Deshais · E. M. Pizarro
University of Florida, Gainesville, FL, USA
e-mail: vollmera@ufl.edu

J. L. Matson (ed.), *Handbook of Intellectual Disabilities*, Autism and Child Psychopathology Series, https://doi.org/10.1007/978-3-030-20843-1_16

behavior (Steinert et al., 2010). Following some controversial debate over the use of these procedures, the American Psychiatric Association (formerly known as the Association of Medical Superintendents of American Institutions for the Insane; AMSAII) was developed in 1844. They asserted a collective decision to permit the use of restraint in American institutions (Colaizzi, 2005).

Although some doctors, such as French psychiatrist Philippe Pinel, advocated for the use of less intrusive strategies to treat and manage individuals with mental illness, the use of seclusion and physical, mechanical, or chemical restraint (i.e., use of medication that slows behavior in a general manner) was widespread and largely unregulated. In the mid-nineteenth century, John Connolly, a superintendent at a large asylum in Great Britain, developed and popularized the use of padded seclusion rooms as an alternative to restraint (Colaizzi, 2005). Connolly was a major proponent of the "non-restraint" movement, urging that psychiatric patients could receive therapeutic treatment in the absence of (mechanical) restraint.

Not much is known about the history of restraint practices in educational settings. We do know, however, that procedures that involve the removal of students from learning opportunities have been in practice for decades. Some examples include sending students home from school, in-school suspension, and various forms of time-out (including seclusion). Ryan, Peterson, Tetreault, and Van Der Hagan (2008) found that students have been secluded in empty rooms, specially designed time-out booths, closets, and refrigerator boxes.

Throughout the twentieth century, concerns about the conditions in psychiatric hospitals frequently arose in public discourse. Many of the debates about inhumane treatment focused largely around seclusion and restraint practices. For example, in 1998, the Hartford Courant published a series of articles revealing that over a 10-year span, 142 individuals died as a result of restraint or seclusion in psychiatric hospitals, facilities for individuals with disabilities, and group homes in the United States (Weiss, Altimari, Blint, & Megan, 1998). Children under the age of 17 accounted for over 26% of the deaths and the disturbing details in the articles sparked public outrage. Despite the fact that most professionals are generally in agreement that less restrictive procedures should be used primarily, investigations by the US Department of Justice revealed sustained inappropriate use of seclusion and restraint (U.S. Department of Education, 2012), and a period of great scrutiny and regulation of these procedures emerged (Weiss et al., 1998). Over the past 20 years, restraint and seclusion have remained controversial topics at the forefront of much debate. Numerous position statements have been disseminated, wherein the misuse of restrictive procedures and their negative effects have been well established. Sturmey (2009) urged researchers for a "call to action" for more research, acknowledging that while the literature has demonstrated that the excessive use of restrictive procedures is undesirable, we have not yet developed a strong body of literature exemplifying how we can effectively eliminate these procedures.

In response to the controversy and risk surrounding the use of restraint and seclusion, these procedures are among the most highly regulated in psychiatric treatment settings. Numerous organizations have developed policies to regulate the use of these procedures. Due to the potentially severe risk to the patient, some individuals even advocate for the total elimination of these procedures. However, failing to use restraint and seclusion in emergency situations could also result in adverse outcomes, either to the patient themselves or to others in their environment (Recupero, Price, Garvey, Daly, & Xavier, 2011). Below we discuss samples of the position statements of various organizations on the use of restraint and seclusion in clinical settings.

The Emergence of Position Statements

The Association of Professional Behavior Analysts (APBA) represents the interests of credentialed practitioners of applied behavior analysis.

It is the position of the APBA that advances in behavior analytic assessments and interventions have greatly suppressed the need for restraint and seclusion procedures. They acknowledge that in some cases, however, the severity of behavior may necessitate more restrictive intervention to minimize the risk of harm. Instances in which the use of restraint and seclusion are deemed essential, the APBA outlines a few critical stipulations. First, restraint and seclusion should only be used once less restrictive procedures have failed, or have been determined to be unsafe or insufficient. Second, the APBA states that restraint and seclusion procedures are not designed to be used in isolation. Rather, they should be used simply as a component of a comprehensive behavior plan. Finally, when restraint and seclusion are used, they should be carefully monitored by a supervising behavior analyst and only implemented by individuals specifically trained in implementation of the procedures (APBA, 2010).

The Association for Behavior Analysis International (ABAI) has been the primary organization of the field of behavior analysis since 1974 (representing not only practitioners, but behavioral scientists and theoreticians). ABAI's position on restraint and seclusion functions under three guiding principles: (1) the welfare of the individual served is the highest priority, (2) individuals have a right to choose, and (3) the principle of least restrictiveness. Given these principles, ABAI condemns the *unnecessary* or *inappropriate* use of intrusive measures such as restraint and seclusion. Similar to APBA, they acknowledge there may be instances in which the use of restraint or seclusion is unavoidable. In these cases, they stress the need for oversight and design by a licensed professional. Additionally, a Behavior Intervention Plan (BIP) that includes the use of restraint must be consistent with the current literature base, derived from a functional assessment, include reinforcement-based procedures, and be objectively monitored through data analysis. In regard to seclusion, ABAI states that when used within the context of a BIP, seclusion can function as a protective measure, as well as to facilitate the acquisition of appropriate behavior. While ABAI approves of a predetermined timeout or safety intervention procedure involving seclusion, it must ultimately serve as an evidence-based component of a comprehensive BIP (Vollmer et al., 2011).

The American Psychiatric Association (APA) supports intervention design that minimizes the use of restraint and seclusion. When restrictive procedures are deemed necessary, they should be implemented by trained individuals. In addition to providing guidelines for reactive procedures, APA also outlines proactive measures to prevent the need for seclusion and restraint. Following each instance of seclusion and restraint, a debriefing should be held during which research and clinical information is discussed to determine best practice procedures. Additionally, they support programs to educate practitioners on the minimal and safe use of these restrictive procedures as well as maintaining updated information on the use of pharmacological interventions that may prevent the need for seclusion and restraint (APA, 2003).

The American Academy of Pediatrics (AAP) acknowledges that "restraint" can be considered both chemical and physical. However, their position statement focused solely on the use of physical restraint, specifically for children or adolescents in the acute care setting. They also distinguish between restraint, and mechanisms typically used during transport or medical procedures. Devices used to protect the patient, to support the patient in a specific position, or to assist in the maintenance of normal body functions are not considered restraint interventions in the view of AAP. They note that beyond these customarily used procedures, restraint may sometimes be necessary for severe behavior to keep the individual from injuring themselves or others. When restraint is required under these circumstances, AAP notes specific procedures for safe application, documentation, reassessment, as well as criteria that such a restrictive procedure should only be used after consideration of alternative methods. AAP makes additional notice that verbal interventions and "therapeutic holding" have been used for children and adolescents in psychiatric facilities to avoid the use of restraint or seclusion. AAP defines therapeutic holding as the "physical restraint of a child by at least two people to assist the child who has lost control of behavior to regain control of strong emotions" (AAP, 1997).

The largest professional membership organization committed to psychiatric-mental health nursing is the American Psychiatric Nurses Association (APNA). In a statement on the topic of restraint and seclusion, they express a commitment to the reduction and ultimate elimination of these restrictive procedures. APNA advocates for further development of empirically based practices for the management and (ideally) complete prevention of behavioral emergencies. While complete elimination of such procedures may seem like a daunting task, APNA is committed to working toward that goal via collaboration with other disciplines and professionals. Until that time, however, they state a few critical specifications should restraint and seclusion be deemed necessary. First, these restrictive procedures should only be used (a) when less restrictive measures have failed and (b) for the minimal time necessary to ensure the safety of both the individual and others in their immediate environment. Additionally, they affirm that individuals who are restrained must be permitted to move as much as possible while still maintaining the safety of others. To ensure their safety, the individual must be monitored continuously throughout implementation of the procedure. The APNA also comments on intent, urging that restraint or seclusion should never be used as a means of convenience, punishment, or coercion (APNA, Updated 2014).

The Cochrane Library is a collection of independent, high-quality evidence to help inform healthcare decision-making. Of a search yielding 2155 citations, no controlled RCT studies evaluating the use of seclusion and restraint were identified. Given the lack of controlled research studies evaluating the effectiveness of these procedures, Cochrane Library states that no official recommendation can be made in regard to the effectiveness, benefit, or harmfulness of seclusion or restraint. However, they do note some qualitative data that suggest these procedures may have serious adverse effects (Fisher, 1994). Although the data are minimal, it is their view that until further analysis is completed, the clinical use of these procedures should be minimized. Until further empirical evidence is gathered, Cochrane Library expressed a need for identifying alternative techniques to replace these restrictive procedures (Sailas & Fenton, 2000).

Restraint and seclusion are of critical concern across a multitude of practitioners and their respective disciplines. The National Association of Psychiatric Healthy Systems (NAPHS) collaborated with the APA, the APNA, with support from the American Hospital Association to compile recommendations to the Board of Trustees of the APA. Among these recommendations, four priority areas were targeted for reform. One of these high-priority areas consisted of seclusion and restraint. The organizations acknowledge that, when used properly, restraint and seclusion can function as injury-sparing and even life-saving measures. Within this compilation, they provide detailed information on specific case studies in which restraint or seclusion may have resulted in unnecessary harm to the individual. They then provide a few regulations surrounding implementation of these procedures. Most notably, they specify that a debriefing should follow each episode of seclusion or restraint. The debriefing should include an assessment of the factors leading to the use of these techniques, steps to reduce their need in the future, and the clinical impact of the intervention on the patient (NAPHS, 2003).

While many groups described above have taken a more neutral position, offering guidelines and parameters for the use of such procedures, some organizations have taken a firm stance on one side or the other. For example, the Autism National Committee (AUTOCOM) has taken a position of complete opposition to the use of physical restraint or seclusion. They believe that these procedures are in direct violation of the civil and human rights of individuals with disabilities. AUTOCOM feels that physical restraint is simply evidence of treatment failure, and should not be used under any circumstance (1999). We will return to this point shortly, in an effort to point out that this position may be extreme and potentially impossible to adopt as policy (see section under "summary: best practices"). Along similar lines, The Autism Society supports legislation on the federal regulation of restraint, and that restraint only be used in situations of imminent danger and always implemented by trained individuals. Conversely, the American Association of School Administrators (AASA) released a statement in strong opposition of this sentiment. AASA argues that restraint and seclusion are sometimes necessary to maintain safety in public schools. Without the option of these procedures during emergency situations, some students would not be able to sustain placement in public school settings (Pudelski, 2012).

In a recently published article, Scheuermann, Peterson, Ryan, and Billingsley (2015) used nonfictional case examples to provoke discussion surrounding the professional use of seclusion and restraint. Although some previous reports described circumstances in which seclusion or restraint were misused, Pudelski (2012) argues that the acts committed by those wrongful individuals are not representative of the majority of personnel that utilize seclusion or restraint procedures in schools. However, Scheuermann et al. (2015) stress that those violations should not be so easily dismissed as isolated events, but rather, as an indication of a more severe foundational issue that warrants exploration. Six categories of professional and ethical concerns surrounding the use of restraint or seclusion were identified. These were: (a) potential for death or injury, (b) failure to use the least intrusive intervention and evidence-based practices, (c) inappropriate restrictions on liberty and access to education, (d) programming failures, (e) disproportionate use with certain minority groups, and (f) insufficient training, supervision, and monitoring. In conclusion, the authors acknowledge that although various professionals have differences of opinion, all seek to maximize the benefits of intervention while minimizing the risk to our clients (or students). With that said, they felt that a call-to-action is necessary both on the individual and group level to maximize the "beneficence" to clients. Legislation concerning the use of restraint and seclusion with individuals with disabilities ranges from nonexistent to comprehensive across various levels of the U.S. government. At federal, state, and local levels, there are laws, guidelines, and policies about the use of these procedures in schools, healthcare facilities, and correctional facilities. However, laws, guidelines, and policy documents do not carry the same weight with respect to adoption, implementation, and adherence by individual institutions (Butler, 2016). Laws, of course, are established via the legislative process and are enforced by the judicial system; thus, violations are subject to legal ramifications. Conversely, guidelines and policy documents are merely recommendations (Butler, 2016). There is no legal recourse if individual institutions (schools, healthcare facilities, correctional facilities) choose to develop policies and practices that do not align with federal or state guidelines or policy recommendations.

Guidelines in Educational Settings

According to the U.S. Department of Education's Office for Civil Rights (OCR), students with disabilities make up 12% of the public education student population but account for 67% of the students exposed to restraint or seclusion procedures (OCR, 2016). In 2016, the DOE published a "Dear Colleague Letter" suggesting that the differential rates of restraint and seclusion used on children with disabilities relative to children without disabilities in schools might constitute discrimination and a violation of Section 504 of the Rehabilitation Act of 1973 and Title II of the Americans with Disabilities Act of 1990 (ADA). Over the past decade, there have been a number of high profile allegations of abuse surrounding restraint and seclusion of children in public schools, including a government-issued report in 2009. Taken together, it is not surprising that there has been a recent push to establish standards for the use of restraint and seclusion in schools.

Federal

At the time of this publication, there is no federal legislation concerning the use of restraint or seclusion in public schools (U.S. Department of Education, 2017). From here forward, the term public schools will be used to refer to educational placements that receive federal, state, or local funding, or some combination (elementary, middle, and high schools, special education schools, charter schools, residential schools). Since 2010, several legislative bills have been introduced to the U.S. Congress. One bill, "Keeping All Students Safe Act" seeks to establish federal laws governing the use of restraint and seclusion in public schools. The bill has undergone a series of revisions and has been introduced to the U.S. House of Representatives and U.S. Senate multiple times, but no versions of the bill have been enacted.

In the most recent version, introduced to the 114th House of Representatives, the bill directs the U.S. Department of Education (DOE) to establish minimum standards about the use of restraint and seclusion in public schools (H.R. 927, 114th Cong., 2015a). Notable sections of the bill include: (a) prohibiting mechanical and chemical restraint, (b) banning restraint that restricts a student's breathing, (c) barring the use of restraint and seclusion under any circumstances except when a student poses an imminent danger of injury to themselves and others, (d) requiring Local Educational Agencies (LEAs) to ensure that school personnel undergo state-approved crisis intervention training, (e) prohibiting restraint and seclusion from being written into a student's education/safety/behavior plan, (f) mandating that schools notify parents in a timely manner if their child is restrained or secluded at school, and (g) directing the DOE to conduct a national assessment of the Act's effectiveness. The bill also authorizes the delivery of federal grant money to LEAs that demonstrate adherence to the items outlined in the bill, collect data on restraint and seclusion, and utilize school-wide positive behavior interventions. At the time of this publication, Keeping Students Safe has not been introduced to the current (115th) U.S. Congress.

In 2015, President Barack Obama passed the "Every Student Succeeds Act" (a reauthorization of the Elementary and Secondary Education Act). Every Student Succeeds calls for a reduction in "the use of aversive behavior interventions that compromise student health and safety" (S. 1177, 114th Cong., 2015b). However, the term "aversive behavior interventions" is not defined. Thus, although this terminology could be interpreted as including restraint and seclusion procedures, this act merely calls for a reduction in aversive behavioral procedures and does not explicitly regulate or prohibit the use of restraint or seclusion in public schools.

Perhaps in lieu of federal legislation, the DOE issued a Resource Document in 2012 outlining 15 guiding principles that LEAs should use when developing restraint and seclusion policies (DOE, 2012). The 15 guiding principles can be found in the bulleted listing below. Although the publication of the Resource Document appears to have influenced the laws and guidelines in some states (Freeman & Sugai, 2013), there is no legal recourse at the federal level if states fail to adhere to the guiding principles:

- Every effort should be made to prevent the need for the use of restraint and for the use of seclusion.
- Schools should never use mechanical restraints to restrict a child's freedom of movement, and schools should never use a drug or medication to control behavior or restrict freedom of movement (except as authorized by a licensed physician or other qualified health professional).
- Physical restraint or seclusion should not be used except in situations where the child's behavior poses imminent danger of serious physical harm to self or others and other interventions are ineffective; in cases where restraint and seclusion are used, they should be discontinued as soon as imminent danger of serious physical harm to self or others has dissipated.

- Policies restricting the use of restraint and seclusion should apply to all children, not just children with disabilities.
- Any behavioral intervention must be consistent with the child's rights to be treated with dignity and free from abuse.
- Restraint or seclusion should never be used as punishment or discipline (e.g., placing in seclusion for out-of-seat behavior), as a means of coercion or retaliation, or as a convenience.
- Restraint or seclusion should never be used in a manner that restricts a child's breathing or harms the child.
- The use of restraint or seclusion, particularly when there is repeated use for an individual child, multiple uses within the same classroom, or multiple uses by the same individual, should trigger a review and, if appropriate, revision of strategies currently in place to address dangerous behavior; if positive behavioral strategies are not in place, staff should consider developing them.
- Behavioral strategies to address dangerous behavior that results in the use of restraint or seclusion should address the underlying cause or purpose of the dangerous behavior.
- Teachers and other personnel should be trained regularly on the appropriate use of effective alternatives to physical restraint and seclusion, such as positive behavioral interventions and supports and, only for cases involving imminent danger of serious physical harm, on the safe use of physical restraint and seclusion.
- Every instance in which restraint or seclusion is used should be carefully and continuously visually monitored to ensure the appropriateness of its use and safety of the child, other children, teachers, and other personnel.
- Parents should be informed of the policies on restraint and seclusion at their child's school or other educational setting, as well as applicable Federal, State, or local laws.
- Parents should be notified as soon as possible following each instance in which restraint or seclusion is used with their child.
- Policies regarding the use of restraint and seclusion should be reviewed regularly and updated as appropriate.
- Policies regarding the use of restraint and seclusion should provide that each incident involving the use of restraint or seclusion be documented in writing and provided for the collection of specific data that would enable teachers, staff, and other personnel to understand and implement the preceding principles.

State

At the state level, the laws, guidelines, and policies related to restraint and seclusion practices vary. In 2009, the U.S. Government Accountability Office (GAO) published a report on restraint and seclusion in schools. The report contained a review of federal and state legislation concerning the use of restraint and seclusion in educational settings. In addition, the GAO report examined 10 cases of restraint and seclusion of school-aged children that resulted in a criminal conviction, a finding of liability, or a large financial settlement. Almost all of the children in these cases were diagnosed with an intellectual or developmental disability. The accounts of these cases contained within the GAO report contain graphic descriptions and disturbing allegations. For example, in 2002, a 14-year-old boy in a Texas public school died as a result of his trunk being compressed after being placed in a prone restraint by a special education teacher almost twice his size because the student refused to remain seated. At the time of the GAO report, 19 states had no legislation related to the use of seclusions or restraints in schools (GAO, 2009).

Since the publication of that report, 30 states have made changes to their laws or policy documents (Freeman & Sugai, 2013). By 2017, 38 states had legislation and 45 states had policy documents regarding restraint and seclusion (Marx & Baker, 2017). At present time, there are no state laws or policy documents that altogether prohibit the use of restraint with students with (or without) disabilities in schools. However, there are five states that explicitly prohibit the use of seclusion with students with disabilities (Butler, 2016). There are four states that do not have any laws or policy documents on restraint and seclusion in schools (Idaho, North Dakota, New Jersey, and South Dakota; Butler, 2016).

In their analysis of state laws and policy documents, Freeman and Sugai (2013) found four general trends in terms of content. One trend was the focus on preventive strategies. This included strategies such as de-escalation training, the use of functional behavior assessments, and schoolwide positive behavior intervention and supports. For example, in Rhode Island, all school staff must attend annual training on de-escalation strategies. Staff members who are identified as authorized to assist in restraint and crisis situations must also receive advanced training (Rhode Island Department of Elementary and Secondary Education, 2002).

Another trend identified by Freeman and Sugai was the establishment of procedural parameters for restraint and seclusion. In state laws and policy documents, procedural parameters are often preceded by definitions of what constitutes restraint and seclusion. Generally speaking, physical restraint is defined as restricting the physical movement of a student and seclusion is defined as placing a student in an area where they are physically prevented from leaving. Although many of the definitions are similar across states, differences in phrasing can have implications for practice and data collection in a given state. For example, in Colorado, restraint does not include instances in which a student is held for less than 4 min or brief holding of a student by one adult for the purposes of calming or comforting the student (Colorado State Board of Education, 2009). This definition leaves room for subjective interpretation. In other words, a student in Colorado could be physically held by an adult for periods of time longer than 5 min as long as the adult's intention is to calm the child. The definitions of seclusion in state laws and policy documents are often distinguished from time-out procedures. However, depending on the state, the distinction between seclusion and time-out is not always clear.

Two other trends that emerged during their analysis are parental and state notification regarding instances of restraint and seclusion and debriefing staff and students following each instance. Thirty-two states require parents to be notified when their child is restrained or secluded at school (Freeman & Sugai, 2013). Most states have established time frames for notification. For example, in Massachusetts, teachers must inform the student's parents within 1 day or send a written report within 3 days (Massachusetts Department of Elementary and Secondary Education, 2014). In terms of reporting rates of restraint and seclusion to the state, 25 states currently collect data at the state-level for students with disabilities (Butler, 2016).

Freeman and Sugai (2013) outlined three specific procedural parameters in their discussion of trends in state laws and policy documents. The first is the use of restraint or seclusion for punishment. According to Butler (2016), 23 states have laws or policy documents that indicate that restraint and seclusion procedures should only be used in emergency situations in which a student is at risk of physically harming themselves or others. Other states, such as Montana, permit the use of restraint if a student is threatening to damage school property (DOE, 2017). Many state laws and policy documents explicitly state that these procedures should be discontinued when the emergency ends and should not be used for the purposes of punishing students. The second procedural parameter is the use of prone restraints. Prone restraints are physical restraints in which an individual is immobilized on the floor in a face down position. Research has demonstrated that some prone restraint positions produce restriction of lung function (Parkes & Carson, 2008). Given this finding and the number of high profile cases in which students have died during or following prone restraints, the use of prone restraints in schools has been explicitly (by name) or implicitly (by definition/description) banned by 33 states (Butler, 2016). The third procedural parameter is the establishment of time limits for restraint and seclusion. Freeman and

Sugai (2013) found that the time limits for restraint or seclusion found in state laws and policy documents ranged across states but most states limited these procedures to 30 min to 1 h, with a stipulation that an administrator must authorize additional time.

In 2017, Marx and Baker reviewed the legislation and policy documents for each state and examined their alignment with the 15 guiding principles outlined in the Resource Document published by the DOE (DOE, 2012). For state laws, they found adherence or partial adherence to the principles ranged from 3 to 32 states, depending on the principle. For state policies, they found that adherence or partial adherence to the principles ranged from 4 to 38 states. Both state laws and policies were most likely to be aligned with Principle 3 (physical restraint must only be used when a student poses a danger to themselves or others) and least likely to be aligned with Principle 14 (policies are reviewed regularly and updated, data need to be collected and analyzed based on subgroups, settings, staff, etc.).

Noneducational Settings

Healthcare Facilities

The Code of Federal Regulations regarding restraint and seclusion are outlined in the Center for Medicare and Medicaid Services (CMS) State Operations Manual (Department of Health & Human Services, 2008). Any hospital, healthcare facility, residential, non-medical, or community-based facility that serves adults or children and that receives federal, state, or local funding must abide by these regulations. In other words, to continue receiving public funding, they must maintain the Conditions for Participation outlined in the manual. Facilities are routinely surveyed to ensure that conditions are being met. The CMS manual states, “All patients have the right to be free from restraint or seclusion, of any form, imposed as a means of coercion, discipline, convenience, or retaliation by staff. Restraint or seclusion may only be imposed to ensure the immediate physical safety of the patient, a staff member, or others and must be discontinued at the earliest possible time” (Department of Health & Human Services, 2008). Physical, mechanical, and chemical restraint are all included in the definition of restraint. The CMS manual also states that restraint and seclusion can only be ordered by a physician or other licensed independent practitioner who is authorized by the state. Standing orders and orders for restraint or seclusion on an as-needed basis are prohibited. Patients must be monitored continuously during periods of restraint or seclusion and patients must be examined face-to-face by an authorized physician or nurse within 1 h of the beginning of the procedure. An additional face-to-face assessment by the ordering physician must be conducted if a restraint lasts longer than 1 h for a patient under 10 years old, 2 h for a patient aged 10–18 years old, or 4 h for patients older than 18 (Masters, 2017). CMS mandates that staff be trained in the implementation of restraint and seclusion and requires all healthcare facilities to report deaths associated with the use of these procedures. State laws governing restraint and seclusion vary from state to state. In cases in which state laws are more stringent than federal laws, the state laws supersede federal laws.

The Joint Commission for hospital accreditation has also published regulations on restraint and seclusion. Technically, the Joint Commission is not a part of or affiliated with the federal government; it is an independent nonprofit organization. However, in the realm of healthcare, their accreditation is considered important. The 2017 Joint Commission standards are in the bulleted listing below.

- Uses restraint or seclusion only when it can be clinically justified or when warranted by patient behavior that threatens the physical safety of the patient, staff, or others.
- Uses restraint or seclusion safely.
- Initiates restraint or seclusion based on an individual order.
- Monitors patients who are restrained or secluded.
- Has written policies and procedures that guide the use of restraint or seclusion.
- Evaluates and reevaluates the patient who is restrained or secluded.

- Continually monitors patients who are simultaneously restrained and secluded.
- Documents the use of restraint or seclusion.
- Trains staff to safely implement the use of restraint or seclusion.

Correctional Facilities

According to the U.S. Department of Justice, approximately 2 in 10 prisoners and 3 in 10 jail inmates report having a cognitive disability (U.S. Department of Justice, 2015). Currently, there are no federal laws or guidelines regarding restraint or seclusion in public correctional facilities specific to inmates diagnosed with an intellectual disability. However, there are two laws that are potentially applicable in certain circumstances. First, the eighth Amendment stipulates that the government may not inflict cruel and unusual punishment on U.S. citizens (U.S. Const. amend. VIII). Second, the ADA prohibits discrimination against individuals with disabilities by public entities. In other words, individuals with intellectual disabilities who are sentenced to public correctional facilities are entitled to the protections outlined in the ADA, including "reasonable accommodation" for their needs and access to the services available to inmates without disabilities. A recent investigation by the United States Department of Justice Civil Rights Division reported that a state correctional facility in Pennsylvania exposed half of the prisoners diagnosed with an intellectual disability to three or more continuous months of solitary confinement. This practice was deemed a violation of the eighth amendment and the ADA.

In 2007, a Task Force commissioned by the APA issued a Resource Document on the use of restraint and seclusion in correctional mental health (Metzner et al., 2007). The Resource Document reported that correctional facilities use restraint (physical and mechanical) and seclusion as mental health interventions as well as for custody or disciplinary purposes. The Resource Document also noted that rules about restraint and seclusion outlined by the CMS manual do little to regulate the use of these procedures in correctional facilities because so few of them participate in Medicare or Medicaid. Correctional facilities usually seek accreditation from the American Correctional Association (ACA) and the Commission on Accreditation for Corrections (CAC). Both bodies require correctional facilities to have policies for the use of restraint but there are no specific guidelines regarding monitoring, time limits, and documentation (Champion, 2007). The Task Force concluded that correctional mental health facilities have not developed guidelines or procedures that are consistent with community practice. Additionally, the Resource Document suggested that the regulations outlined in the CMS manual should be used as a basis for developing national standards of care for the use of seclusion and restraint in correction facilities (Champion, 2007).

Summary and Best Practices

A simple "thought" problem will demonstrate that nearly all would agree that seclusion or restraint is sometimes necessary. Suppose that an able-bodied adult teacher is walking through a school hallway and a student shouts, "I have a gun and I am going to shoot!" The teacher sees that the student does in fact have a gun, and the student has begun to fire in the direction of other students by pulling the trigger. Probably most anyone would agree that the teacher should restrain the student in whatever way possible if the opportunity to do so should arise. Now suppose the student is not just any student, but is a student known to have an intellectual ability. Should the teacher refrain from restraining due to the disability? Now further suppose that there is no gun, but a student with a disability has just hurt a student and is running toward another student with a closed fist. The point is not to take a position that is pro-seclusion or pro-restraint, but rather to take the position that has been adopted by most entities concerned with abuses of seclusion and restraint: there are times when it is likely necessary or at least safest to use these approaches. Given this likelihood, most entities have chosen to identify best practices in the use of seclusion and restraint.

Seclusion and restraint should be reserved for cases when a person is posing immediate harm to themselves or others. The procedures should only be used if it is determined that other strategies, such as de-escalation, are ineffective. Staff in facilities and schools should be trained in the use of state or locally approved crisis prevention strategies. If seclusion or restraint is used, the procedures should be carefully monitored by professional staff, and the parents or guardians of the individual should be notified as immediately as possible. If use of these procedures is repeated (i.e., used more than once), future planning should occur and treatment strategies should be adopted and documented based on a functional assessment of the student's behavior. Such a treatment plan should be approved by relevant care providers and especially by parents or guardians. The treatment plan should include strategies for increasing alternative and safe behavior through positive reinforcement and other less intrusive strategies, and decisions about efficacy should be determined by careful monitoring of plan implementation and outcome data for behavior of concern.

It also seems important to further distinguish seclusion from timeout. Seclusion should never be considered a treatment strategy, it is reserved for protection of or from the individual. However, research on timeout shows that periods of restricted access to positive reinforcement can be effective at very short intervals (such as 2 min) and without isolating the individual (e.g., Donaldson, Vollmer, Yakich, & Van Camp, 2013). Thus, whereas timeout could be an acceptable component of a behavioral intervention plan, which would presumably also include reinforcement for alternative behavior, seclusion is not an acceptable treatment component. It is a protection strategy.

To conclude, seclusion and restraint will remain controversial because both procedures are at times misused. Such misuse occurs on a continuum from ignorance, to negligence, to abuse. It stands to reason that such misuses must stop, and that is the purpose of the many position statements, proposed regulations, and standards of use. However, to ignore that the procedures will be used in dangerous circumstances is to in fact set the occasion for terrible misuse. Haphazard use of seclusion and restraint should be avoided at all costs. Understanding implementation of seclusion and restraint should be a priority for staff and care provider training at all levels of care for individuals with intellectual disabilities.

References

American Academy of Pediatrics. (1997). *The use of physical restraint interventions for children and adolescents in the acute care setting*. Elk Grove Village, IL.

American Psychiatric Association, American Psychiatric Nurses Association, National Association of Psychiatric Health Systems. (2003). Learning from each other: Success stories and ideas for reducing restraint/seclusion in behavioral health.

American Psychiatric Nurses Association. (2000; Updated 2014). *APNA position statement on the use of seclusion and restraint.*

Americans with Disabilities Act of 1990. Pub. L. 101-336. 26 July 1990. 104 Stat. 328.

Association of Professional Behavior Analysts. (2010). Position statement on the use of restraint and seclusion as interventions for dangerous and destructive behaviors: Supporting research and practice guidelines.

Autism National Committee. (1999). *Position on restraints.*

Butler, J. (2016). How safe is the schoolhouse: An analysis of state seclusion and restraint laws and policies?

Champion, M. K. (2007). Commentary: Seclusion and restraint in corrections—A time for change. *Journal of the American Academy of Psychiatry and the Law Online, 35*(4), 426–430.

Civic Impulse. (2015a). H.R. 927 – 114th Congress: Keeping All Students Safe Act. Retrieved from https://www.govtrack.us/congress/bills/114/hr927

Civic Impulse. (2015b). S. 1177 – 114th Congress: Every Student Succeeds Act. Retrieved from https://www.govtrack.us/congress/bills/114/s1177

Colaizzi, J. (2005). Seclusion & restraint: A historical perspective. *Journal of Psychosocial Nursing and Mental Health Services, 43*(2), 31–37.

Colorado State Board of Education. (2009). *Code of Colorado Regulations.*

Department of Health & Human Services. (2008). *CMS Manual System.*

Donaldson, J. M., Vollmer, T. R., Yakich, T. M., & Van Camp, C. (2013). Effects of a reduced time-out interval on compliance with the time-out instruction. *Journal of Applied Behavior Analysis, 46*(2), 369–378.

Fisher, W. A. (1994). Restraint and seclusion: a review of the literature. *The American Journal of Psychology, 151*(11), 1584–1591.

Freeman, J., & Sugai, G. (2013). Recent changes in state policies and legislation regarding restraint or seclusion. *Exceptional Children, 79*(4), 427–438.

Marx, T. A., & Baker, J. N. (2017). Analysis of restraint and seclusion legislation and policy across states: Adherence to recommended principles. *Journal of Disability Policy Studies, 28*(1), 23–23.

Massachusetts Department of Elementary and Secondary Education. (2014). Code of Massachusetts Regulations.

Masters, K. J. (2017). Physical restraint: A Historical review and current practice. *Psychiatric Annals, 47*(1), 52–55.

Metzner, J. L., Tardiff, K., Lion, J., Reid, W. H., Recupero, P. R., Schetky, D. H., … Janofsky, J. S. (2007). Resource document on the use of restraint and seclusion in correctional mental health care. *Journal of the American Academy of Psychiatry and the Law Online, 35*(4), 417–425.

Parkes, J., & Carson, R. (2008). Sudden death during restraint: do some positions affect lung function? *Medicine, Science and the Law, 48*(2), 137–141.

Pudelski, S. (2012). *Keeping Schools Safe: How seclusion and restraint protects students and school personnel*. Retrieved from https://www.aasa.org/uploadedFiles/Policy_and_Advocacy/AASA-July-2012-Keeping-Schools-Safe.pdf

Recupero, P. R., Price, M., Garvey, K. A., Daly, B., & Xavier, S. L. (2011). Restraint and seclusion in psychiatric treatment settings: Regulation, case law, and risk management. *The Journal of the American Academy of Psychiatry and the Law, 39*, 465–476.

Rhode Island Department of Elementary and Secondary Education. (2002). *Board of regents for elementary and secondary education physical restraint regulations.*

Ryan, J. B., Peterson, R. L., Tetreault, G., & Van Der Hagan, E. (2008). Reducing the use of seclusion and restraint in a day school program. In M. A. Nunno, D. M. Day, & L. B. Bullard (Eds.), *For our own safety: Examining the safety of high- 141 risk interventions for children and young people* (pp. 201–215). Arlington, VA: Child Welfare League of America.

Sailas, E. E. S., & Fenton, M. (2000). Seclusion and restraint for people with serious mental illnesses. *Cochrane Database of Systematic Reviews*, 1–18.

Scheuermann, B., Peterson, R., Ryan, J. B., & Billingsley, G. (2015). Professional practice and ethical issues related to physical restraint and seclusion in schools. *Journal of Disability Policy Studies, 27*, 86–95.

Steinert, T., Lepping, P., Bernhardsgrütter, R., Conca, A., Hatling, T., Janssen, W., … Whittington, R. (2010). Incidence of seclusion and restraint in psychiatric hospitals: a literature review and survey of international trends. *Social Psychiatry and Psychiatric Epidemiology, 45*(9), 889–897.

Sturmey, P. (2009). Restraint, seclusion and PRN medication in english services for people with learning disabilities administered by the national health service: An analysis of the 2007 national audit survey. *Journal of Applied Research in Intellectual Disabilities, 22*(2), 140–144.

The Joint Commission. (2017). *Joint Commission Accreditation Standards.*

U.S. Department of Education. (2012). *Restraint and seclusion: Resource document.*

U.S. Department of Education. (2016). *Dear colleague letter.*

U.S. Department of Education. (2017). *Montana compilation school discipline laws and regulation.*

U.S. Department of Education's Office for Civil Rights. (2016). *2013–2014 Civil Rights Data Collection: A First Look.*

U.S. Department of Justice. (2015). Special report: Disabilities among prison and jail inmates, 2011–12.

U.S. Government Accountability Office. (2009). *Seclusions and restraints: Selected cases of death and abuse at public and private schools and treatment centers.*

Vollmer, T. R., Hagopian, L. P., Bailey, J. S., Dorsey, M. F., Hanley, G. P., Lennox, D., … Spreat, S. (2011). The Association for Behavior Analysis International Position Statement on Restraint and Seclusion. *The Behavior Analyst, 34*(1), 103–110.

Weiss, E.M., Altimari, D, Blint, D.F., & Megan, K. Deadly restraint: a nationwide pattern of death. a Hartford Courant investigative report. Hartford Courant, October 11, 1998.

Monitoring Drug Side-Effects

17

Gerda M. de Kuijper, Arne Risselada,
and Rianne van Dijken

Introduction

People with intellectual disabilities are one of the most heavily medicated population in society. This phenomenon is likely related to the frequently presence of comorbid ill-health conditions (Cooper et al., 2015). The prevalence of ill-health conditions in people with intellectual disability varies between populations and is highest in people living in institutional settings. Also, the nature of ill-health conditions may vary. In general, people with mild intellectual disability are more vulnerable for ill-health conditions associated with cardiovascular disorders like obesity and diabetes. People with severe intellectual disability more often suffer from ill-health conditions associated with brain dysfunction like epilepsy and cerebral palsy. Besides, a high proportion of people with intellectual disability suffer from gastrointestinal conditions like constipation and gastroesophageal reflux disorder. Mental ill-health conditions, including autism spectrum disorder and challenging behavior, are prevalent in approximately 40% of people with intellectual disability (Cooper, Smiley, Morrison, Williamson, & Allan, 2007; Smiley et al., 2007). The increased health risks in people with intellectual disability are probably related to life style factors, e.g., unhealthy diets, lack of exercises and sedentary life styles, and genetic conditions, e.g., increased risk for obesity and mood disorder in Down syndrome, for hearing loss and psychosis in Velo-cardio-fascial syndrome, and for sleep disorders and self-injurious behavior in Smith-Magenis syndrome.

In people with intellectual disability, the medication use is high and polypharmacy is common. Polypharmacy is defined as the use of more than five pharmaceutical agents simultaneously. Haider, Ansari, Vaughan, Matters, and Emerson (2014) found in a population-based study of people with intellectual disability in Victoria, Australia, that 76% of adults >18 year used one or more prescribed medicines and in 21% there was polypharmacy (Haider et al., 2014). O'Dwyer et al. (2016) studied a population of older adults (40–90 years) and found a percentage of 90% medication use and of 32% polypharmacy (O'Dwyer, Peklar, McCallion, McCarron, & Henman, 2016). In a population of children and adolescents aged 4–18 year Tobi et al. found that 22% used chronic medication of whom 47% used two or more drugs (Tobi et al., 2005).

G. M. de Kuijper (✉)
Centre for Intellectual Disabilities & Mental Health/
GGZ Drenthe Research, Assen, The Netherlands

A. Risselada
Department of Clinical Pharmacy, Wilhelmina
Hospital, Assen, The Netherlands

R. van Dijken
Mooi mijn lijf, Intellectual Disability Health Care &
Sexology Practice, Assen, The Netherlands

J. L. Matson (ed.), *Handbook of Intellectual Disabilities*, Autism and Child Psychopathology
Series, https://doi.org/10.1007/978-3-030-20843-1_17

The prevalence of drug use in these school-aged children increased with severity of intellectual disability. All studies showed that the most frequently prescribed classes of drugs were psychotropic drugs and antiepileptic drugs. Also, laxatives and drugs for peptic ulcer diseases were frequently prescribed as well as analgesics. Anticholinergic medication was prescribed in 16%, mainly in those with polypharmacy (O'Dwyer, Peklar, et al., 2016).

Polypharmacy may have negative consequences like increased health care costs and health risks (Maher, Hanlon, & Hajjar, 2014). Especially people with intellectual disability who live independently or who receive support to a small extend are prone to medication non-adherence or inappropriate storage of their medication. This may lead to lack of treatments effects or even life-threatening situations, e.g., in case of insulin storage outside a fridge resulting in a hyperglycemic coma. Also drug interactions may lead to decreased effects of pharmaceutical agents; however these interactions may also lead to increased effects. Drug interactions may occur when one or more pharmaceutical agents are administered concurrently resulting in reactions between the drugs. These reactions may influence the absorption or the excretion of the drug, and/or the availability of drug receptors. As a consequence these interactions may increase the risks for the occurrence of drug-induced adverse events. These may be serious and life threatening also, as was shown in studies of the occurrence of adverse events and reasons for health care visits and hospitalization (Maher et al., 2014; Warle-van Herwaarden et al., 2012). Moreover, adverse events may reduce the quality of life of people with intellectual disability (Brandt, Lahr, & May, 2015; Ramerman, Hoekstra, & de Kuijper, 2018a; Scheifes et al., 2016). People with intellectual disability are more vulnerable for the occurrence of drug-induced side-effects, especially for side-effects of psychotropic drugs, including antiepileptic drugs (de Kuijper et al., 2013; de Winter, Bastiaanse, Hilgenkamp, Evenhuis, & Echteld, 2012; Matson & Mahan, 2010; Scheifes, Walraven, et al., 2016; Sheehan et al., 2017), because of the frequent presence of comorbid disorders and the risk of drug interactions in case of multiple drug use. The recognition of drug-induced side-effects is often hampered, on the one hand by patient-related factors like communication problems and reduced capacity to adequately cooperate in assessments, on the other hand by staff-related factors like lack of knowledge on psychotropic drug side-effects (de Kuijper & van der Putten, 2017; Donley, Chan, & Webber, 2012).

In this chapter information on drug-induced side-effects of the most frequently prescribed classes of drugs in people with intellectual disability is provided, with an emphasis on side-effects of psychotropic drugs. Also, information on the monitoring of side-effects, including guideline recommendations on instruments and monitoring schedules which can facilitate the assessment of psychotropic drugs' side-effects, is presented. The role of the different disciplines involved with pharmaceutical care in people with intellectual disability, i.e., clinicians, pharmacists, and nurses regarding the monitoring and management of side-effects will be briefly outlined. Finally, a tool which is useful in evaluating effects of drug-use and management of side-effects will be discussed.

Side-Effects of Drugs: Definition and Precautions

Drug-induced adverse events, otherwise named side-effects of drugs, are defined as any harmful, undesirable effect of drugs. Three types of side-effects can be distinguished related to the origin of occurrence. There are two types of patient-related side-effects. Both are drug dose independent. The first type is thought to be caused by allergic or disturbed immunological responses. Examples are skin rashes and lowering of blood platelets or leucocytes. The other type of patient-related side-effect is an increased risk for specific comorbid disorders, e.g., increased risk for cardiovascular disorder in case of use of NSAID analgesics. The frequency of both types of side-effects is low and their existence mostly becomes yet clear when the drug in question is used on a large scale. The third type of side-effect is drug dependent. This type of side-effect is often dose dependent and related to the pharmaceutical effect of the drug in question. It is mostly discovered in clinical trials before the drug in question is licensed and registered by pharmaceutical and health care authorities. After a licensed registration all the available information on side-effects should be included in the medication leaflet.

Drugs are cleared from the body in different ways, i.e., metabolization by the liver and excretion in urine or feces. In case of compromised renal or liver function the dosage of the drug should be lowered to minimize the chance for potential dose dependent side-effects. Furthermore, for several metabolizing enzymes in the liver (e.g., CYP2D6 and CYP2C19) there are known genetic variances influencing their metabolic capacity that result in altered elimination half-lives of drugs and therefore different plasma levels. Based on their genetic makeup patients can be classified as poor, intermediate, extensive, or ultrarapid metabolizers (Berm, Risselada, Mulder, Hak, & Wilffert, 2013).

In case of rapid metabolizers higher doses than normal are needed to treat the patient, whereas in poor metabolizers much lower doses are needed to achieve the same result. Intermediate and extensive metabolizers can often be dosed as normal. Especially in case of psychotropic drugs (e.g., antidepressants and antipsychotics) and cytotoxic drugs there is a lot of evidence supporting the benefits of genotyping (Swen et al., 2011). However, given the relative lack of experience of most physicians with genotyping in daily clinical practice it is advisable to consult an expert beforehand, to maximize the potential benefits of genotyping.

Drug metabolizing and/or drug clearance problems of patients increase the risk of dose dependent side-effects and drug interactions. Therefore, in case of difficulties in history taking, which will often be the case in people with intellectual disability, a laboratory examination on potential liver and renal dysfunction and genotyping of metabolizing enzymes may be needed before prescribing the drug. Also, monitoring of drug plasma levels during drug use is warranted in case of drug clearance problems and altered metabolization.

Side-Effects by Drug Classes

As was described in the introduction section, psychotropic drugs, antiepileptic drugs, laxatives, proton pump inhibitors, antacids, analgesics, and anticholinergic medication are the most frequently prescribed drug classes in people with intellectual disability. Although in this chapter the focus is on side-effects of psychotropic drugs, some information on the other drug classes is also given.

In people with intellectual disability there is often comorbidity of mental disorders, epilepsy and gastrointestinal disorders, and therefore polypharmacy consisting of drugs used to treat these disorders is common. Moreover, agents from these drug classes are also prescribed to reduce the burden caused by side-effects of psychotropic drugs.

Antiepileptic Drugs

There are different types of antiepileptic drugs, and these agents are licensed for different indications. Main reasons for use are seizure disorders, neuropathic pain, migraine, and last but not least for bipolar and mood disorders.

Common side-effects of all types of antiepileptics are sleepiness, fatigue, and weight gain.

Laxatives

Laxatives, e.g., macrogol or lactulose are most often prescribed for long-term prevention of constipation in people with cerebral palsy or otherwise disturbed autonomic bowel function. Laxatives are also often prescribed as co-medication to prevent constipation caused by other drugs, e.g., psychotropic drugs and iron supplements.

Common side-effects are inherent to their function: abdominal cramps, flatulence, and diarrhea.

Proton Pump Inhibitors and Antacids

These agents are prescribed to reduce heartburn and the risk of gastroesophageal disorders. Especially people with severe intellectual disability are in need for this type of medication.

Furthermore, these agents are often prescribed as co-medication to reduce the risk of peptic ulcers when patients are treated with NSAID's (e.g., diclofenac, ibuprofen, naproxen). Side-effects of proton pump inhibitors and antacids are rare, but may include headache and nausea. Long-term use may cause anemia due to a decrease in absorption of iron. Furthermore, the decrease of acidity in the stomach may increase the risk for respiratory tract infections, especially in case of aspiration of stomach content which is relatively common in people who are more severely disabled.

Analgesics

Chronic use of analgesics is most common in people with multiple disabilities, who may also have mental or behavioral problems. NSAID's and paracetamol are commonly prescribed.

Use of NSAID's is associated with gastrointestinal complaints and with increased risks for peptic ulcer and stomach bleeding.

Anticholinergic Agents

These drugs are prescribed to reduce symptoms of Parkinson disease such as muscle stiffness and brady- and hypokinesia. Other indications are asthma and bladder dysfunction, and reduction of antipsychotic drug-induced parkinsonism.

Side effects of these agents and drug interactions are common. Side-effects are, among others, constipation, dizziness, sleepiness, headache, dry mouth and dry eyes, and bladder dysfunction/urinary retention. Especially older people and people with intellectual disability are vulnerable to these side effects and prescription of anticholinergic medication in these populations should preferably be avoided.

Side-Effects of Psychotropic Drugs

Psychotropic drugs are prescribed for a wide range of mental disorders and psychiatric and behavioral symptoms. Most are registered and licensed for specific indications, sometimes extended to specific symptoms. An example of the latter is the antipsychotic drug risperidone which is licensed for the treatment of psychosis and for the short-term treatment of aggressive or irritable behavior. Psychotropic drugs are also often prescribed off-label (outside licensed indications) where evidence for their effectiveness and safety may be scarce. In people with intellectual disability the off-label use of psychotropic drugs, especially of antipsychotics, is high (de Kuijper & Hoekstra, 2017; Haw & Stubbs, 2005; Sheehan et al., 2015).

Psychotropic drugs act by modulating the availability of neurotransmitters by stimulating or inhibiting their release, or enhancing or blocking the function of their receptors on neurological cell membranes. Neurotransmitters are biochemical agents, which are produced by neurons and bind on neurotransmitter receptors located at the cell membrane of other neurons. Brain functions are based on coordinated activities of neurons via neuronal pathways of neuronal axons and neurotransmitters. There are different types of neurotransmitters, e.g., dopamine, serotonin, histamine, (nor) adrenalin, glutamine, acetylcholine, and gamma-aminobutyric acid, each with its own receptor. Neurotransmitter receptors are located on neurons of neuronal pathways in the central nervous system (the brain), but also on neurons of peripheral neuronal pathways. Psychotropic drugs exert their actions by modulating specific neurons of neuronal pathways in the central nervous system. Apart from hopefully achieving the desired result like decrease of symptoms of mental disorders this may also lead to part of the side-effects caused by these medications. Examples are parkinsonism caused by antipsychotic drugs and tremors caused by antidepressants. Besides these side-effects caused by modulation of the central nervous system, psychotropic-induced side-effects also occur by modulation of the peripheral nervous system. Examples of the latter are for instance constipation and urinary retention caused by anticholinergic properties of various antipsychotic and antidepressant drugs.

Side-effects of psychotic drugs are very common and often harmful to health and may therefore reduce the quality of life (de Kuijper et al., 2013; Koch et al., 2015; Ramerman, Hoekstra, & de Kuijper, 2018b; Young, Taylor, & Lawrie, 2014). When prescribing psychotropic drugs clinicians ought to discuss the advantages and disadvantages of treatment with the client, caregivers, and representatives. In case of serious mental illness the potential benefits will be clear, but in case of off-label prescriptions pros and cons should be carefully balanced.

Side-effects of psychotropic drugs can be distinguished in metabolic, neurological, and hormonal side-effects. Furthermore, there are sexual side-effects, which probably are caused by neurological as well as by hormonal effects of psychotropic drugs, and a category miscellaneous, e.g., skin rashes. Side-effects of the latter category are mostly patient-related, occur infrequently and cannot be explained by the pharmaceutical drug-properties. This type of side-effects will not be discussed in this chapter.

- Metabolic side-effects are weight gain, increase in abdominal fat mass, unfavorable changes in plasma lipids, and disturbed glucose metabolism with increased risks for diabetes. Most of these side-effects are associated with the use of second generation antipsychotic drugs, also commonly described as atypical antipsychotic drugs, and increase the risks for the presence of the so-called metabolic syndrome, which in turn increases the risk for cardiovascular disorders.
- Neurological side-effects; an overview of the different kinds of these side-effects is presented in Table 17.1.

Motor side-effects of psychotropic drugs, so-called extrapyramidal symptoms, may occur shortly after the first administration of the drug, e.g., acute dystonia, or after long-term use, e.g., tardive dyskinesia and tardive akathisia. The occurrence of tardive dyskinesia is sometimes related to dose changes or discontinuation of the drug and may persist after the use of the drug has been stopped. Motor side-effects are associated with the use of antipsychotics, and especially in case of multiple psychotropic drug use, long-term use and older age a substantial part of patients will develop tardive dyskinesia and/or akathisia.

Table 17.1 Neurological side-effects of psychotropic drugs

Motor side-effects	Dystonia[a]	Dyskinesia[b]	Parkinsonism[c]	Akathisia[d]
Autonomic side-effects	Gastrointestinal side-effects; drooling, obstipation, abdominal pain	Cardiovascular side-effects; tachy/bradycardia, blood pressure dysregulation	Bladder function; incontinence, urine retention	Blurred vision, disturbed sexual function, temperature dysregulation
Sensory side-effects	Hypo/hyperpathy of the skin	Tingling of fingers, hands or feet	Abnormal taste or smell	Tinnitus
Cognitive/psychological side-effects	Sedation, sleepiness, dizziness	Disturbed mood and/or sleep	Affect and cognitive flattening	Loss of appetite/increased appetite

[a] Dystonia: painful, persistent muscle cramps
[b] Dyskinesia: involuntary movements of face, eyelids, mouth, and tongue, of hands and fingers, and of ankles and feet
[c] Parkinsonism: muscle stiffness, tremor, brady- and hypokinesia
[d] Akathisia: feelings of- or motor restlessness

- Hormonal side-effects are caused by drug-induced increased release of the lacto trophic hormone prolactin. In turn, this hormone may suppress the release of the sex hormones estrogen and testosterone. Effects are disturbed menstrual cycle, loss of libido, and reduction of muscles. Secondary effects are loss of bone mass and feelings of fatigue and loss of energy.
- Sexual side-effects of psychotropic drugs may be underestimated or ignored by clinicians, in particular in people with intellectual disability. Therefore, we will extend on this theme below.

It is well known that psychotropic drugs can contribute to sexual dysfunction or sexual problems (Bella & Shamloul, 2014). Sexual dysfunction can be defined as any reduction in desire or libido, diminished arousal, a decline in the frequency of intercourse, or an undesirable delay in or inability to achieve orgasm. In the general population sexual dysfunction is rarely reported spontaneously, leading to underestimation of its prevalence. In studies, using structured interviews or questionnaires, it has been shown that 16–60% of patients using oral antipsychotics experience sexual dysfunction (de Boer, Castelein, Wiersma, Schoevers, & Knegtering, 2015). However even though this is well known there are hardly any studies available when it comes to people with an intellectual disability. People with an intellectual disability have limited verbal communication abilities which results in an inability to report adequately sexual side-effects of medication. For example the inability to achieve orgasm can result in excessive masturbation. Possibly, this behavior may be interpreted as an increase in maladaptive behavior, which in turn may lead to dosage increases or additional prescriptions of psychotropic drugs.

The pharmacological mechanisms of antipsychotic-induced sexual dysfunction are not yet fully understood. Dopamine antagonism, α1-antagonism and prolactin elevation are likely to be involved. Most antipsychotics are potent postsynaptic dopamine antagonists and can cause elevation of prolactin. Dopamine is involved in experiencing motivation and rewards, including sexual motivation (desire) and probably also sexual reward. While dopamine antagonists decrease levels of dopaminergic output, prolactin levels are increased (de Boer et al., 2015). Antipsychotic-induced hyperprolactinemia has been associated with a number of side effects including galactorrhea, menstrual disturbances, amenorrhea, decreased libido, and erectile dysfunction (de Boer et al., 2015; Lambert, Farmer, & Brahm, 2013). Furthermore, histamine receptor antagonism and hormonal changes could also be involved (Bella & Shamloul, 2014).

Table 17.2 shows the main effects of antipsychotics on sexual functioning.

Treatment with antidepressant drugs is also associated with significant rates of sexual dysfunction. The pharmacological mechanisms are largely unknown. A hypothesis is that SSRI's reduce dopaminergic transmission via serotonin receptors in the mesolimbic area, thereby reduc-

Table 17.2 Effect of antipsychotics on sexual functioning[a]

	Effects on sexual response
D2 agonism	Increased sexual desire (anticipation of reward)
D2 antagonism	Decreased sexual desire and activity, erection and ejaculation
5-HT2 agonism	Delay of orgasm
5-HT1a agonism	Activation of sexual behavior, facilitation of orgasm
5-HT2a and 5-HT2c antagonism	Probable stimulation of sexual behavior
α1 antagonism	Central effect: Decrease of erection and ejaculation. Decrease of lubrication peripheral effect: May have a stimulating effect on erection
α2 antagonism	Stimulation of erection
H1 antagonism	Sedation causing indirect effect on sexual performance
M1 antagonism	Decreased erection and lubrication

[a] Adapted from De Boer, Castelein, Wiersma, Schoevers & Knegtering, The facts about sexual (Dys)function in schizophrenia: an overview of clinically relevant findings, Schizophrenia Bulletin 2015: 674–686

ing the dopaminergic stimulating effect on sexual desire and orgasm (Bella & Shamloul, 2014).

In section "Psychotropic drugs' side-effects by subclasses" information per specific subclass of psychotropic drugs will be provided.

Assessment of Psychotropic Drugs' Side-Effects

The assessment of the presence and severity of drug-induced side-effects in people with intellectual disability may be difficult. In this population there are often limitations in self-reporting of subjective symptoms, and in adequately cooperating in physical examinations and blood sampling because of motor or behavioral problems.

In people with intellectual disability, regular methods for physical examinations of blood pressure, body weight and length, and waist circumference will mostly suffice; however in people with multiple disabilities adapted methods may be needed. Also, blood and urine sampling for assessment of metabolic and hormonal side-effect will mostly be quite possible. In case of urine incontinence catheter sampling may be done. Measurements of bone density may be needed for assessments of osteoporosis caused by drug-induced bone loss. Here, also adapted methods may be needed.

Neurological side-effects are generally assessed by taking history and observation of the patient, and a brief physical examination, which is aimed to identify symptoms of parkinsonism, dystonia, dyskinesia, and akathisia. Also examination of heart rate and heart rhythm, and of blood pressure are needed to assess cardiovascular autonomic side-effects. Sometimes Electro Cardio Gram (ECG) laboratory is necessary in case of suspected heart rhythm disorder or use of certain types of psychotropic drugs, which are known to cause disturbed heart rhythms. Cognitive, psychological, and autonomic side-effects are generally assessed with help of questionnaires and self-report instruments.

In people with intellectual disability assessment of neurological side-effects may be hampered, because of underlying motor disabilities or dyspraxia, and patients' limitations in self-report of autonomic, psychological, and cognitive side-effects. Observations in daily life of clients by support professionals and interviews of clients along with help of caregivers, or in case of non-verbal clients, interviews with proxies will be needed for taking history of potential subjective symptoms of side-effects. There are a number of instruments which are specifically developed or adapted for use in patients with intellectual disability. One of these is the Matson Evaluation of Side-effects scale (MEDS) (Matson et al., 1998; Matson & Cervantes, 2013), a comprehensive scale covering all types of psychotropic drugs-induced side-effects. It is an informant-based scale, which should be completed by a high graduated nurse based on the medical record, own observations and by interviewing the client's caregiver. It consists of nine subscales on bodily functions or anatomic regions, including four scales for neurological side-effects, i.e., Central Nervous System (CNS)-General (G), CNS-Dystonia (D), CNS-Parkinsonism/Dyskinesia P/D, CNS-Behavioral/Akathisia (BA), and covers all types of side-effects. It is a widely used scale, but its use may be limited in countries where there is lack of high graduated nurses in health care systems for people with intellectual disability.

In general, in most countries there is lack of adapted scales to assess neurological side-effects in people with intellectual disability. Yet, there are some scales which may be suitable for people with intellectual disability. For the assessment of autonomic side-effects the Scale Outcomes Parkinson disease-Autonomic symptoms (SCOPA-AUT) (Visser, Marinus, Stiggelbout, & Van Hilten, 2004) may be a useful instrument when slightly adapted by adding questions on urine and fecal continence. It was used in research projects on discontinuation of antipsychotics (de Kuijper, Evenhuis, Minderaa, & Hoekstra, 2014). The Epworth Sleepiness Scale may be used for assessment of drug-induced sedation (Ramerman et al., 2018b) and the Antipsychotics and Sexual Functioning Questionnaire (ASFQ) for the assessment of sexual side-effects (de Boer et al., 2013). However, all these scales are self-report

Table 17.3 Reliable[a] instruments for assessment of the presence and severity of drug-induced extrapyramidal symptoms

	Dystonia	Dyskinesia	Parkinsonism	Akathisa
Single instrument General population		Abnormal involuntary movement scale (AIMS) Dyskinesia condensed user scale (DISCUS)	Simpson angus scale (SAS) Unified Parkinson disease rating scale (UPDRS)/motor items	Barnes Akathisia rating scale (BARS)
Multi-instrument General population	St. Hans rating scale (SHRS) Yale extrapyramidal symptom scale (YESS)	SHRS Drug-induced extrapyramidal symptoms scale (DIEPSS) Extrapyramidal symptoms rating scale (ESRS)	SHRS YESS	SHRS YESS DIEPSS ESRS
Single instrument Intellectual disability population	Matson[b] CNS-dystonia	DISCUS Matson[b] CNS-dyskinesia/Parkinsonism (DP)	UPDRS/motor items 20, 21, 22, 31 Matson[b] CNS-PD	Akathisia ratings of movements scale (ARMS)[c] Matson[b] CNS-behavioral/ Akathisia

[a] Van Strien, Keijsers et al. Rating Scales to measure side-effects of antipsychotic medication. J. Psychofarmacol., 2015: 857–866

[b] Informant-based scale. Matson & Cervantes. Current status of the MEDS. Res. Dev. Disabil., 2013:1849–1853

[c] Bodfish, Newell et al. Akathisia in adults with mental retardation: development of the ARMS. Am. J. Ment. Retard, 1997:413–423

instruments and therefore should preferably be completed by the main caregiver along with the client or client‘s family.

To assess drug-induced extrapyramidal symptoms there are a lot of instruments and scales available. There are also a number of instruments which are specifically developed or validated for use in patients with intellectual disability. Table 17.3 shows instruments for assessments of dystonia, dyskinesia, parkinsonism, and akathisia for use in the general population and for use in people with intellectual disability. There are single- and multi-instruments, developed for measurement of a single kind of extrapyramidal symptom or instruments covering all kind of extrapyramidal symptoms respectively.

In intellectual disability practice the AIMS, DISCUS, UPDRS, BARS, and ARMS are widely used. As a multi-instrument the SHRS may be an alternative, however, for assessments of dyskinesia addition of the DISCUS or AIMS is possibly needed.

The reliability of instruments for use in the general population as well as for use in the intellectually disabled population may be less well when assessments are based on judgments of less well trained health care professionals. The latter was shown in a recent study on agreements of main caregivers and physicians in the presence and severity of dyskinesia, akathisia, and parkinsonism in their patients. There were hardly any correlations between the informant-based three neurological subscales CNS-D, CNS-PD, and CNS-BA of the MEDS and assessments of physicians with help of the AIMS, DISCUS, and BARS (de Kuijper & Hoekstra, 2016).

It is very important that support professionals and other staff members, who are working with people with intellectual disability and mental or behavioral disorders, are well educated and trained in the effects of psychotropic drug use in people with intellectual disability. Staff ought to have some knowledge on storage and administration of medication, as well as on registered indications, desired effects and adverse events of psychotropic drugs. They should have knowledge on reasons for prescription and know of the precautions which are needed, in particular in case of off-label use. However, studies have shown support professionals lack sufficient knowledge and they indicated that they were in need for education and training on this issue (de Kuijper & van der Putten, 2017; Donley et al., 2012).

In people with intellectual disability a major problem is the identification and assessment of dyskinesia and akathisia, since these symptoms may be held for behavioral symptoms. This is especially so when psychotropic drugs are prescribed for challenging behavior, e.g., dyskinesia may be mistaken for hyperactivity and akathisia for restlessness. Here, training and education off support professionals, nurses and physicians in both effects of psychotropic drug use, as well as in management of challenging behavior and symptoms of mental disorders should be a priority.

Videotaping of observations and of physical examinations directed towards identification and assessment of extrapyramidal symptoms may be used to enhance the intra-rater and inter-rater reliability of these symptoms.

A promising future direction may be the use of Telemedicine. An example is videoconferencing of patients and clinicians, when the patient is in his own environment, which may aid to better diagnostics of extrapyramidal symptoms. Videotaping of observations and of the physical examinations directed towards extrapyramidal symptoms may aid in more reliable assessments of clinicians. Software programs may be used to compare clinical comparisons over time (Shore et al., 2015). Furthermore, the use of electronic devices for measurements of bradykinesia and akathisia may be promising (Mentzel et al., 2016; Mentzel et al., 2016; Poyurovsky et al., 2000). Actually, a research project is running, investigating the applicability in people with intellectual disability (Beumer & Maes-Festen, 2017).

Assessment of Sexual Side-Effects

Several studies have shown that sexual dysfunction is one of the most distressing side effects of psychotropic drugs and a major cause of poor quality of life (Bella & Shamloul, 2014). As we know this, what does this mean for people with an intellectual disability. What do we know and how can we recognize sexual dysfunction? There are no studies which describe the epidemiology, impact, and treatment of sexual dysfunction among persons with an intellectual disability. Yet, some recent studies have shown that sexuality is an important issue for people with intellectual disability (Brown & McCann, 2018; Gil-Llario, Morell-Mengual, Ballester-Arnal, & Diaz-Rodriguez, 2018). Inappropriate sexual behavior among persons with an intellectual disability is quite common with the incidence ranging from 15 to 33%. However, this may be more attributable to socially inappropriate behavior than to serious assaults (Thom, Grudzinskas Jr, & Saleh, 2017). The studies investigating sexuality issues in men and women with mild and moderate intellectual disability had shown that the majority of clients had some knowledge and had received education on sexuality, although this may be incomplete. Furthermore, a majority had sexual feelings and fantasies, and had sexual relationships with another person. The most frequent sexual behaviors and practices were masturbation, kissing and petting, vaginal intercourse and oral sex. Yet, just 41% reported they had sufficient sexual satisfaction (Brown & McCann, 2018; Gil-Llario et al., 2018). In clinical practice in institutional settings it is known that a considerable part of women experience pain during intercourse, are in need for education on sexual behavior and skills, and have no understanding of their own sexual needs and desires. Also, in men there is lack of adequate knowledge and skills and there is often report of excessive or inappropriate masturbation, inappropriate touching and/ or touching others and erection problems. Furthermore, men report that they are often dissatisfied with their partner or lack a partner. So, we may conclude that sexual side-effects of psychotropic drugs are likely to be an important issue in people with intellectual disability.

Adults with intellectual disability tend to have limited verbal communication abilities. This results in their inability to express a subjective experience of adverse drug reactions and side-effects of medication (Lambert et al., 2013). An important goal in clinically assessing sexual side effects of psychotropic drugs in people with an intellectual disability is therefore to recognize sexual dysfunction. Psychological, social, symptom, and medication-related aspects of sexual performance should be disentangled. Therefore, investing in the relationship between patient and clinician tends to be worthwhile. Furthermore, clinicians should strive to establish and maintain a good relationship with family members and/or staff and caregiver. Obviously, if applicable, clinicians must obtain the permission of clients prior to discussing their sexual problems with any other person than the client him/herself. It is important to explain the cause of their sexual problems to the client and also to understand the influence of sexuality on the individual's overall quality of life in order to try to find a solution and to assess the need for additional therapy. Treatment alternatives should be discussed and considered. Lowering the dose, switching to an antipsychotic with less detrimental effects on sexual functioning, or prescribing additional medication, e.g., a dopamine agonist, aripiprazole, or phosphodiesterase-5-inhibitor, are the main treatment options (de Boer et al., 2015). Clinicians should be aware that people with a mild or borderline intellectual disability might stop their medication use when they don't understand the possibilities to decrease the sexual side effects and the consequences of stopping their medication. Stories and explanation in simple language and pictures may enable people with an intellectual disability to understand the nature and causes of their sexual problems.

There are a number of instruments available for the assessments of sexual side-effects (de Boer, Castelein, Wiersma, Schoevers, & Knegtering, 2014). Unfortunately, these questionnaires are not specifically designed for people with intellectual disability and because most are self-report instruments their usefulness may be limited in people with intellectual disability. However, since the ASFQ has been designed for people with cognitive and concentration problems (de Boer et al., 2013), this questionnaire may be also suitable for people with intellectual disabilities. Furthermore, in a recent study in people with intellectual disability, a questionnaire, evaluating sexual behavior, preventive behavior and knowledge, and experience of sexual abuse, has been used (Gil-Llario et al., 2018). It may be worthwhile to investigate whether this questionnaire is useful in assessments of psychotropic drugs' sexual side-effects in people with intellectual disability.

In contrast to people with mild or borderline intellectual disability, recognizing sexual side effects in people with a severe or profound intellectual disability may be more difficult, due to their limited communication abilities. Inappropriate touching and/or approaching others may be a sign of sexual dysfunction as well as excessive masturbation. Clinicians may try to lower the dose or taper off the drugs which likely cause the sexual side-effects, in order to investigate whether the sexual problems are caused by the drug in question. In case of considerable risks that psychiatric symptoms might increase or re-emerge when the drug dose is lowered, other causes should be excluded first.

Psychotropic Drugs' Side-Effects by Subclasses

Antipsychotic Drugs

Antipsychotic drugs primarily achieve their therapeutic efficacy in psychotic disorders by blocking dopaminergic and serotonergic receptors. Apart from their antagonistic properties for dopamine and serotonergic receptors, these drugs may also have antihistaminergic, anticholinergic, and antiadrenergic effects. Antipsychotic drugs may be divided in three generations (Table 17.4), with the second and third generation also often being referred to as atypical due to their lower antidopaminergic activity compared to the first generation antipsychotic drugs and their antiserotonergic properties.

Although there are substantial differences in pharmacological mode of action, research has shown that there are only small differences in antipsychotic efficacy (Leucht et al., 2013). Only clozapine appears to be more effective than the

Table 17.4 Three generations of antipsychotic drugs and their properties

Generation	Predominant mode of action	Important antipsychotic drugs in this group
First generation	Antidopaminergic (D_2), except for clozapine Antiadrenergic Anticholinergic (some)	Haloperidol, pipamperone, pimozid, zuclopentixole, clozapine
Second generation	Antidopaminergic Antiserotonergic Antihistaminergic Antiadrenergic Anticholinergic	Olanzapine, risperidone, paliperidone, quetiapine
Third generation	Partial antagonism for D_2-receptor (ca. 80% inhibition, 20% activation) Partial antagonism for serotonergic 5HT1A/ Antiserotonergic (5HT2A) Antihistaminergic (higher doses) Antiadrenergic (higher doses)	Aripiprazole, brexpiprazole

other antipsychotic drugs, specifically in treatment resistant schizophrenia. When differences in efficacy are small, side-effects and comorbidities become important characteristics in choosing between antipsychotic drugs.

Based on experience and cost often first generation antipsychotics and risperidone are chosen first. These drugs primarily exert their action by strongly inhibiting dopaminergic receptors, and therefore also often cause side-effects associated with this: affective flattening, extrapyramidal symptoms, and hyperprolactinemia. When these side-effects occur a dosage reduction should be the first step, since these side-effects are dose dependent. If the side-effects persist, or dosage reduction is not possible without losing therapeutic benefit, a switch towards an antipsychotic drug with less antidopaminergic action is warranted, e.g., clozapine, quetiapine, olanzapine or aripiprazole. The second generation antipsychotic drugs are less antidopaminergic than the first generation antipsychotic drugs, apart from risperidone and paliperidone, but have strong antiserotonergic and antihistaminergic properties. Antipsychotic drugs with strong antihistaminergic properties (e.g., olanzapine, quetiapine, clozapine) are strongly sedative. This may be beneficial when a patient also suffers from a sleeping disorder, but increases the chance for falling incidents. Antipsychotic drugs with strong antiserotonergic properties (e.g., olanzapine, quetiapine, clozapine) decrease feelings of satiety during eating and lower the metabolic rate. Combined with the often present antihistaminergic action of these antipsychotic drugs this causes weight gain, and potentially diabetes and/or the metabolic syndrome. When a patient is underweight these side-effects may be beneficial, but more often weight gain is a harmful side-effect, since it increase the risk for diabetes, hypertension and several forms of cancer.

The third generation antipsychotic drugs currently consists of aripiprazole, but a new antipsychotic drug in this class is on its way, brexpiprazole. These antipsychotic drugs primarily have a partial antagonistic action on dopaminergic (D2) and serotonergic (5HT1A) receptors, and are therefore also sometimes called dopaminergic and serotonergic modulators. Antipsychotic drugs in this generation have a more favorable profile regarding side-effects, since they less frequently cause extrapyramidal and metabolic side-effects. They are considerably more expensive than the other antipsychotic drugs however and frequently cause akathisia.

Many antipsychotic drugs (e.g., olanzapine, quetiapine, and clozapine) have strong anticholinergic properties, and often give rise to anticholinergic side-effects like constipation, urinary retention, dry mouth (and associated periodontitis), and cognitive problems. These side-effects are almost always harmful to the patient.

All antipsychotic drugs also increase the risk of a stroke, especially in the first 3 months, and most give rise to QTc prolongation, which increases the risk of a ventricular arrhythmia and cardiac mortality (Chen et al., 2017; Hsu et al., 2017). Some antipsychotics, especially clozapine and to a lesser extent quetiapine and olanzapine, also increase the risk of an epileptic seizure (mostly myoclonic) (Alper, Schwartz, Kolts, & Khan, 2007).

Antidepressant Drugs

Most antidepressants exert their action by increasing serotonin and/or noradrenalin levels in the synaptic cleft, the small space between two adjacent neurons. Based on their mechanism of action or chemical structure antidepressant drugs are often divided into several groups (Table 17.5).

SSRI's

The drugs belonging to the SSRI's hardly differ in their mechanism of action, and therefore also cause the same side-effects: tremors, excessive sweating, nausea and vomiting, diarrhea, sleeping disorders, restless legs, and sexual dysfunction (mostly impairment of libido and ejaculation). Paroxetine is somewhat anticholinergic and may therefore cause constipation more often than other SSRI's.

SNRI's

The SNRI's duloxetine and venlafaxine inhibit both the reuptake of serotonin and noradrenalin. Since this includes the same pharmacologic action as SSRI's these drugs can also cause the same side-effects. Their noradrenergic action however adds some side-effects like agitation, hypertension, and tachycardia. This may be undesirable in most patients, but when a patient also experiences neuropathic pain, which benefits from a noradrenergic action, duloxetine may be a desirable drug.

TCA's

The various tricyclic antidepressant drugs differ more with regard to their mechanism of action. All TCA's inhibit the reuptake of serotonin and noradrenalin, but some are predominantly serotonergic (imipramine, clomipramine) whereas others are predominantly noradrenergic (nortriptyline). Amitriptyline inhibits both the reuptake of serotonin and noradrenalin equally. Since TCA's exert the same mechanism of action as the SSRI's and or SNRI's, they can also cause the same side-effects as those groups. However, TCA's also have strong antihistaminergic and anticholinergic properties and are more cardiotoxic due to an inhibition of electrical conduction in the atrioventricular node which can result in a cardiac arrest when an overdose is taken. Their antihistaminergic properties result in weight gain and sedation, whereas their anticholinergic properties result in constipation, urinary retention, dry mouth, and cognitive problems among others. Although harmful in many patients, some

Table 17.5 Overview of antidepressants and their mechanism of action

Group	Mechanism of action	Important antidepressants in this group
Selective serotonin reuptake inhibitors (SSRI's)	Inhibit reuptake of serotonin	Paroxetine, fluoxetine, sertraline, citalopram, escitalopram, fluvoxamine
Serotonin and noradrenalin reuptake inhibitors (SNRI's)	Inhibit reuptake of serotonin and noradrenalin	Venlafaxine, duloxetine
Tricyclic antidepressants (TCA's)	Inhibit reuptake of serotonin and/or noradrenalin Antihistaminergic Anticholinergic	Amitriptyline, nortriptyline, clomipramine, imipramine
MAO-inhibitors	Inhibit mono-amine oxidase (which breaks down dopamine, serotonin and noradrenalin)	Tranylcypromine, phenelzine, moclobemide
Miscellaneous	Divers. Most have an impact on serotonergic, dopaminergic or noradrenergic receptors	Bupropion, mirtazapine, trazodone, agomelatine.

patients may benefit from these side-effects. As an example, bed wetting can be treated with amitriptyline or clomipramine, and neuropathic pain or sleeping disorders can also benefit from amitriptyline. Nortriptyline is the least anticholinergic TCA and is therefore often the drug of choice when a TCA is indicated.

MAO-Inhibitors

MAO-inhibitors are sometimes used in treatment resistant depression, and consist of two irreversible MAO_A and MAO_B inhibitors (phenelzine and tranylcypromine) and one reversible MAO_A-inhibitor (moclobemid). Inhibition of MAO, which is an enzyme responsible for the degradation of serotonin, dopamine and noradrenaline can result in a variety of side-effects. The most-important side-effects of these drugs are orthostatic hypotension, nausea and vomiting, and sexual dysfunction. When an irreversible MAO-inhibitor is used a diet excluding tyramine, an amino acid that can be found in some wines and cheese among others, becomes very important because ingestion of tyramine can result in a hypertensive crisis in these patients. The same goes for use of inotropic drugs like isoprenalin, adrenalin, xylomethazoline, dopamine and noradrenalin. Tranylcypromine has a dexamphetamine like effect, which can cause hypertension shortly after a dose is ingested, resulting in headache.

Miscellaneous

Antidepressants belonging to the miscellaneous group differ greatly in their mechanism of action and therefore also in associated side-effects. The two most commonly used drugs in this group are bupropion and mirtazapine. Bupropion inhibits the reuptake of dopamine. Side-effects include nausea and vomiting, convulsions, and tremors, but the dopaminergic action of bupropion might be of benefit in patients with Attention Deficit Hyperactivity Disorder (ADHD). Mirtazapine has strong antihistaminergic and antiserotonergic properties resulting in sedation and weight gain. This mechanism of action might be of added value in people with depression who also suffer from sleeping disorders. Mirtazapine can sometimes be added (in low dose) to treatment with SSRI's or SNRI's to treat sexual dysfunction and sleeping disorders caused by the SSRI or SNRI.

Trazodone acts as a serotonin antagonist in low dosages, but inhibits reuptake of serotonin when higher dosages are used. In low dosages trazodone is used to prevent drug-induced sleeping disorders (e.g., MAO-inhibitors).

Agomelatin is an antidepressant that stimulates melatonin receptors and blocks a specific serotonergic receptor. It might have some benefit in treatment of depression with concomitant sleeping disorders, but is known to cause serious side-effects, specifically hepatic failure.

Psychostimulants

There are several psychostimulants that can be used to treat ADD and ADHD. The most commonly used stimulants are methylphenidate, dexamphetamine, atomoxetine, clonidine, and bupropion. Methylphenidate and dexamphetamine stimulate the central nervous system by inhibiting the reuptake of noradrenaline and dopamine and stimulating the release of these two neurotransmitters. Side-effects include loss of appetite, weight loss, hypertension, and nausea and vomiting.

Atomoxetine is a selective noradrenalin reuptake inhibitor causing associated side-effects like hypertension, tachycardia, loss of appetite, and weight loss. Bupropion inhibits the reuptake of dopamine, and also causes side-effects like nausea and vomiting, convulsions, and loss of appetite.

Growth retardation can occur during the use of stimulants as a result of the loss of appetite and weight loss.

Clonidine acts as an adrenergic-alpha2a-agonist and therefore has a very different mode of action. Its main side-effects include sedation and hypotension, and is not associated with growth retardation.

Mood Stabilizers

The most commonly used mood stabilizers are lithium, sodium valproate, carbamazepine, lamotrigine, and the antipsychotic drugs. These drugs are used to treat severe and ongoing periodic mood swings that are symptoms of bipolar disorder. Sodium valproate, carbamazepine, and lamotrigine are also used as antiepileptic drugs.

Lithium is the keystone in treating bipolar disorder, since lithium is an effective treatment for both the manic and depressive states and reduces the suicide risk. Lithium may however cause several serious side-effects, including weight gain, tremors, cognitive and concentration problems, polyuria, nephrogenic diabetes insipidus, and thyroid dysfunction. Long-term use of lithium may also cause a decrease in renal function.

Sodium valproate is also frequently used as a mood stabilizer and may cause side-effects including polycystic ovarium syndrome, liver toxicity, hematologic problems, elevated ammonia levels, and weight gain.

Carbamazepine is less frequently used as a mood stabilizer due to the multiple drug interactions carbamazepine causes and its anticholinergic properties. Its side-effects include sedation, liver toxicity, gamma-GT elevation, hematologic problems, Stephen Johnson syndrome, and hyponatremia.

Lamotrigine is used mainly to treat depressive episodes, since it lacks efficacy in treating mania. Lamotrigine is often well tolerated but frequently causes skin disorders, especially when dose accumulation is rapid. It may also rarely cause Stephen Johnson Syndrome and toxic epidermal necrosis which are life-threatening conditions that are comparable to third degree burns.

The antipsychotic drugs are most effective in treating mania. Their side-effects have been described in the previous section.

Anxiolytic Drugs

Drugs used to treat anxiety include the aforementioned SSRI's, benzodiazepines (e.g., oxazepam, diazepam, lorazepam), buspirone, and propranolol.

Benzodiazepines are drugs with strong sedative properties that may therefore have a negative impact on response time and cognition, cause affective flattening, and increase the risk of falling. In people with intellectual disability benzodiazepines may cause paradoxical reactions including agitation.

Buspirone is less sedative than benzodiazepines. Buspiron acts as a partial serotonin agonist and may cause side-effects including dizziness and headaches.

Propranolol is a non-selective beta antagonist which is mainly used as an anxiolytic drug when there is sympathetic over-activity and in aggressive outbreaks. Caution is warranted in patient suffering from asthma or COPD since propranolol may impair the efficacy of bronchodilators used in those patients. Side-effects include bradycardia, sleeping disorders, nightmares, fatigue, and cold hands and feet.

Hypnotics

Benzodiazepines used as a hypnotic drug include nitrazepam, temazepam, midazolam, zolpidem, and zopiclon. Their side-effects are the same as the other benzodiazepines.

Melatonin and promethazine may also be used as hypnotic drugs. Melatonin is frequently used as a hypnotic/chronobiotic when there are circadian problems and rarely causes side-effects. Promethazine may be used when benzodiazepines aren't effective and has strong antihistaminergic and anticholinergic properties. Side-effects include sedation, dizziness, confusion, constipation, urinary retention, and dry mouth.

Low dosages of the antidepressant mirtazapine (3–15 mg) or the antipsychotic quetiapine (12.5–50 mg) are also used to treat sleeping disorders. However, since the metabolic side-effects of these drugs are largely independent of the dosage monitoring for these potential side-effects is necessary.

Monitoring of Psychotropic Drugs' Side-Effects

There are a number of guidelines on the prescribing of psychotropic drugs in people with intellectual disability. Examples are the NICE guideline "Psychotropic medicines in people with learning disabilities whose behaviour challenges," which includes an algorithm for the review, reduction or stopping of psychotropic drugs (nice.org.uk/guidance/ktt19, January 2017), the guideline of The Royal College of Psychiatrists (UK, April 2016) "Psychotropic drug prescribing for people with intellectual disability, mental health problems and or behaviours that challenge: practice guidelines "(http://www.rcpsych.ac.uk/pdf/FR_ID_09_for_website.pdf) and a paper in the journal "Australian Prescriber" (Trollor, Salomon, & Franklin, 2016). Also, guidelines on this issue exist in other countries, adapted to own health care systems and language. All guidelines recommend evaluating on reasons for (ongoing prescription) effects and safety of the prescribed drugs.

Before a pharmaceutical treatment is started an assessment of baseline health condition, including use of prescribed and of "over the counter" medication is needed and symptoms of potential side-effects should be examined. Baseline blood pressure, heart rate, body weight, body mass index, and laboratory parameters should be noted. In people with intellectual disability the presence and severity of minor neurological deficits, e.g., developmental coordination disorder, mild forms of dyspraxia or cerebral palsy and symptoms of Parkinson's disease should be noted, since these symptoms may be mistaken for extrapyramidal symptoms.

The monitoring of psychotropic drugs differs between psychotropic drug subclasses.

Below you will find monitoring schedules of the two most frequently prescribed psychotropic drugs subclasses in people with intellectual disability, i.e., antipsychotic drugs and antidepressants. For the monitoring of mood stabilizers we refer to national professionals guidelines of psychiatrists; a guideline on the prescribing of psychotropic drugs, including monitoring schedules, of the Dutch Association of Intellectual Disability Physicians (NVAVG) is also available (nvavg.nl/voorschrijven-van-psychofarmaca/; bijlage 9.5d-9.5 g, December 2016).

It is important to realize that all of these monitoring schedules are based on expert opinions and are generated in multidisciplinary groups. There are no studies comparing multiple monitoring schedules with regard to harms and benefits. Therefore it is important to weigh the potential benefits of wanting to know if certain side-effects occur against the harm it may cause for each individual patient. Since collection of blood can be stressful for the patient monitoring for side-effects should be noninvasive whenever possible. When the potential benefits of blood monitoring outweigh the potential harm it is important to also have a baseline measurement before the drug is started. A baseline measurement may help in determining which drug is best suited (e.g., when sodium is already low carbamazepine may not be the best choice) and also helps in identifying whether the recently started drug is to blame for a certain change in blood value.

Antipsychotics

The monitoring schedule for antipsychotics, as shown in Table 17.6, was specifically designed for patients with intellectual disability (de Leon, Greenlee, Barber, Sabaawi, & Singh, 2009). Monitoring is largely noninvasive, apart from measurements focused on metabolic side-effects (glucose and lipid levels) and prolactin. Baseline measurements are more elaborate.

Antidepressants

The monitoring schedule for antidepressants, as shown in Table 17.7, was not specifically designed for patients with intellectual disability. Monitoring is largely noninvasive, apart from measurements of glucose levels, renal function, and electrolyte levels. The latter is mainly relevant for diagnosing a drug-induced hyponatremia otherwise known as Syndrome for Inappropriate ADH release (SIADH), which mostly occurs in the first few weeks of treatment. An ECG may be necessary in case of relevant cardiac comorbidities or risk factors for QTc prolongation when TCA's (e.g., nortriptyline and amitriptyline) are used (Table 17.8).

Management of Side-Effects of Psychotropic Drugs

When side-effects occur, regardless of what type of drug causes it, the first step towards preventing or treating the side-effect should be to determine

Table 17.6 Monitoring for side-effects when using antipsychotic drugs (excl. clozapine)[a]

	Baseline	*t* = 1 month	*t* = 2 months	*t* = 3 months	*t* = 6 months	*t* = 12 months	Half-yearly	Yearly
Somatic case history, metabolic	X			X				
Family anamnesis, metabolic	X							
MMSE								
Tardive dyskinesia score	X			X	X	X	X	X
RR	X			X				
Heart frequency	X			X				
Waist circumference	X			X		X		X
Weight	X	X	X	X				X
Length	X							
BMI	X	X	X	X				
Side-effects [A]		X	X	X	X	X		X
Liver function								
AP	X							
AST	X							
ALT	X							
gammaGT	X							
Lipids								
Triglycerides	X				X	X	X	X
Cholesterol	X				X	X	X	X
LDL	X				X	X	X	X
HDL	X				X	X	X	X

Electrolytes								
Sodium	X							
Potassium	X							
Calcium								
Renal function								
Creatinine	X							
Urea	X							
Blood sugar								
Glucose (fasting)	X	X		X	X	X	X	X
HbA1c	X			X				
Cardiology								
ECG	X					X		X
Hormones								
Prolactin	X					X		X
Physical examination on prolactin-induced side-effects						X		X
Plasma level antipsychotic								
Plasma levels only on indication								

[a] Adapted from: De Leon J, Greenlee B, Barber J, Sabaawi M, Singh NN. Practical guidelines for the use of new generation antipsychotic drugs (except clozapine) in adult individuals with intellectual disabilities. Research in Developmental Disabilities 2009;30:613–669

Table 17.7 Monitoring for side-effects when using antidepressants: TCA's (X), SSRI's (Y), SNRI's (Z), and mirtazapine (as Z)[a]

	Baseline	*T* = 3 weeks	*T* = 3 months	*T* = 6 months	*T* = 12 months	Yearly
Metabolic syndrome						
Length	X, Y, Z					
BMI	X, Y, Z	X, Y, Z	X, Y, Z	X, Y, Z	X, Y, Z	X, Y, Z
Waist circumference	X, Y, Z	X, Y, Z	X, Y, Z	X, Y, Z	X, Y, Z	X, Y, Z
HbA_{1C} and (sober) glucose	X, Y, Z			X, Y, Z	X, Y, Z	X, Y, Z
Lipids	X, Y, Z			X, Y, Z	X, Y, Z	X, Y, Z
Cardiovascular						
ECG	X[b]	X[b]		X[b]	X[b]	X[b]
Blood pressure (standing up/lying)	X, Z	X, Z	X, Z	X, Z	X, Z	X, Z
Heart frequency	X, Z	X, Z	X, Z	X, Z	X, Z	X, Z
Therapeutic drug monitoring						
Drug levels	X: When starting, and after dosage alterations					
Hematologic						
Complete blood count	Only when clinically indicated					
Liver functions						
AP, ALT, AST, GammaGT, bilirubin	Only when clinically indicated					
Renal function and electrolytes						
eGFR	X, Y, Z	X, Y, Z		X, Y, Z	X, Y, Z	X, Y, Z
Sodium	X, Y, Z	X, Y, Z		X, Y, Z	X, Y, Z	X, Y, Z
Potassium	X, Y, Z	X, Y, Z		X, Y, Z	X, Y, Z	X, Y, Z

[a] Adapted from: Simoons M, Pot HJ, van Roon EN, Ruhé HG, Mulder H. Monitoring van bijwerkingen bij gebruik van antidepressiva. Psyfar 2014, 2; 10–18

[b] Only at the start of treatment and after significant dosage alterations in patients with risk factors for cardiac problems including QTc prolongation (table x), otherwise on indication

whether or not that specific side effect is dose dependent. If the side effect is dose dependent, and most of them are, or when there is uncertainty whether or not it is dose dependent the first action should be to lower the dose of that drug. If lowering the dose does not solve the problem or is not possible without losing efficacy, then the second step should be to see whether an alternative treatment is possible with a different psychotropic drug that may be equally effective, but is less likely to cause the same side effect. For example, if side-effects like parkinsonism are caused by an antipsychotic drug with strong antidopaminergic activity (e.g., risperidone) a switch towards an antipsychotic drug with less antidopaminergic activity (e.g., aripiprazole or quetiapine) may solve the problem. If none of these actions are possible, then addition of other drugs to treat the side-effects is warranted. Otherwise the risk for polypharmacy increases, with more and more drugs being added to treat the side-effects of other drugs. The treatment of four side-effects with a high prevalence in people with intellectual disability caused by psychotropic drugs will be discussed; constipation, extrapyramidal symptoms, sexual dysfunction and weight gain.

Table 17.8 Risk factors for QTc prolongation[a]

Alcoholism
Change in nutrition (anorexia nervosa, starvation, malnutrition)
Bradycardia (<50 strokes/min)
Cerebrovascular problems (intracranial and subarachnoïdal bleeding, stroke, intracranial trauma)
Congenital long QT-syndrome
Diabetes mellitus
High age
Electrolyte problems (hypokalemia, hypomagnesaemia, hypocalcaemia)
Female sex
Heart failure (cardiomyopathy)
Hypertension
Hypoglycemia
Hypothermia
Thyroid insufficiency
Myocardial infarction or ischemia
Obesity

[a] Adapted from: Moleman P. Psychofarmaca en het QT-interval. Psyfar 2012;7:10–17

Constipation

Constipation is often caused by drugs with anticholinergic properties, like most antipsychotic drugs, tricyclic antidepressants, and promethazine. A recent study used the Anticholinergic Burden Scale (ABS) to compute a cumulative score of drugs to determine an individual's anticholinergic burden in older people with intellectual disability (O'Dwyer, Maidment, et al., 2016). Drugs with no anticholinergic effects score 0, drugs with possible anticholinergic effects score 1 and drugs with definite cognitive anticholinergic effects score 2 or 3. In these older patients 42% had an ACB score of 1–4, 29% had an ACB score ≥ 5, and 29% had an ACB score of 0. So overall, around 71% of the older population with intellectual disability had at least one drug with anticholinergic properties, of which antipsychotics were the dominant group. Antipsychotics comprised 35% of the total cumulative ACB score, followed by anticholinergics (16%, e.g., biperidene) most of which were probably added to treat extrapyramidal symptoms caused by the same antipsychotic drug. Antidepressant drugs with anticholinergic properties (e.g., TCA's) were also used regularly (9.4% of the ACB-3 drugs used in this population).

Apart from daytime drowsiness which was significantly associated with a higher ACB score, a greater proportion of those with a higher anticholinergic burden were chronically constipated (27% ACB ≥ 5 vs. 8% ACB 0) and frequently used two or more concurrent laxatives (29% ACB ≥ 5 vs. 5% ACB 0).

Regarding the use of psychotropic drugs with anticholinergic properties, the first step should be to see if treatment with that drug is still needed or if a switch is possible towards an alternative with less or no anticholinergic properties like some first and third generation antipsychotic drugs, risperidone, SNRI's, and SSRI's.

If switching is not possible or constipation persists treatment towards treating constipation should be started. The basic treatment of every form of constipation consists of non-medical interventions focused on adequate intake of fluids and fiber and increasing physical activity. Laxatives are used if those interventions are insufficient to resolve the constipation.

Laxatives are generally divided into four groups, depending on the way they work; bulk-forming laxatives (also known as fiber supplements, e.g., psyllium fibers), osmotic laxatives (e.g., lactulose, macrogols, magnesiumhydroxide, and phosphate enemas), stimulant laxatives (e.g., bisacodyl, senna), and fecal softeners (e.g., paraffin).

The stimulant laxatives and paraffin are less suited for chronic use, because the constant irritation of the colonic mucosa by the stimulant laxatives may result in a megacolon, and paraffin may cause anal irritation. The other types of laxatives can be used chronically, but there are some practical issues to consider. Lactulose may cause flatulence which may cause stomach and intestinal discomfort. Bulk-forming laxatives have to be combined with adequate intake of fluids to prevent bezoar formation. Therefore in most practices macrogols are used as laxatives for chronic use, since these less frequently cause flatulence and require less fluid intake.

To monitor a patients fecal pattern and determine constipation in an early stage a Bristol stool chart (https://commons.wikimedia.org/wiki/

File:BristolStoolChart.png) with type 1 and type 2 being indicative for constipation.

Extrapyramidal Symptoms

Extrapyramidal symptoms (EPS) are drug-induced movement disorders that include acute and tardive symptoms. The symptoms include dystonia, akathisia (including restless legs syndrome and bruxism), parkinsonism, bradykinesia, tremors, and tardive dyskinesia. EPS are frequently caused by antipsychotic drugs, but can also be caused by serotonin reuptake inhibitors and sodium valproate.

EPS are a dose-dependent side effect, so again, reduction of the dose should be the first potential step in preventing EPS. If a dose reduction is not possible without losing efficacy a switch towards a more favorable psychotropic drug may be warranted. Regarding antipsychotic drugs, a choice can be made towards less antidopaminergic antipsychotic drugs like clozapine, quetiapine, olanzapine, and aripiprazole. When EPS occur during use of SSRI's a switch towards an antidepressant that does not inhibit the reuptake of serotonin is advisable, like nortriptyline, mirtazapine, or bupropion. EPS during use of sodium valproate may be less easy to switch away from, since a switch to lithium may also result in tremors as a side-effect of lithium, and a switch to carbamazepine often results in unwanted drug interactions. Mood stabilizers that may be a suitable alternative regarding EPS are lamotrigine, olanzapine, aripiprazole, or quetiapine.

EPS can be treated by starting anticholinergic drugs like biperidene, but these drugs cause anticholinergic side-effects that are troublesome in this population. Moreover, use of biperidene may increase the severity of dyskinesia. If this option is chosen extra attention towards anticholinergic side-effects is warranted.

When a patients suffers from a sleeping disorder caused by restless legs syndrome or bruxism (frequently caused by SSRI's) dopaminergic agents (e.g., pramipexole) can be used to treat these problems. In patients who are prone to psychosis adding a dopaminergic agent is not absolutely contraindicated, but caution is advised and a lower starting dose of the dopaminergic agent should be considered.

Sexual Dysfunction

Many psychotropic drugs can cause sexual dysfunction. This may sometimes be a desirable side effect since 15–33% of the population with ID show inappropriate sexual behavior (Thom et al., 2017), but often is not. Sexual dysfunction may consist of a diminished libido, erectile dysfunction and premature or delayed ejaculation and is often caused by antipsychotic and antidepressant drugs.

Antipsychotic drugs with antidopaminergic properties and antidepressants that inhibit serotonin reuptake (e.g., SNRI's, SSRI's, TCA's) increase prolactin levels, which in turn causes a reduction in estrogen and testosterone levels leading to a diminished libido. Switching to antipsychotic drugs with less antidopaminergic activity (e.g., quetiapine, clozapine, aripiprazole) or antidepressants that don't inhibit the reuptake of serotonin (e.g., nortriptyline, mirtazapine, bupropion) may solve the problem. Addition of a low dose of bupropion, when antidepressants are the cause of the sexual dysfunction, is also possible since this reduces elevated prolactin levels (Pereira, Arias-Carrion, Machado, Nardi, & Silva, 2014).

Although erectile dysfunction may also be caused by a diminished libido it is often associated with anticholinergic drugs. Switching to a comparative drug with less or no anticholinergic action may improve the erectile dysfunction. Addition of a phosphodiesterase-5-inhibitor (e.g., sildenafil, tadalafin) may also improve the erectile dysfunction.

Ejaculation problems are often associated with noradrenergic actions of psychotropic drugs. Drugs with a noradrenergic stimulating action (e.g., the noradrenalin reuptake inhibitors atomoxetine and nortriptyline) may cause a premature or even a spontaneous ejaculation, whereas drugs with anti-noradrenergic properties (e.g., antipsychotics, TCA's) may cause a delayed ejaculation. Dose reduction when possible or switching to a psychotropic drug with a different noradrenergic profile may solve the problem.

Table 17.9 Antipsychotic-induced weight gain (mean) in first 10 weeks of treatment

Antipsychotic	Weight gain (kg)
Clozapine	4.45
Olanzapine	4.15
Thioridazine	3.19
Sertindole	2.92
Chloorpromazine	2.58
Risperidone	2.10
Quetiapine	2.18
Sulpiride	1.90
Haloperidol	1.08
Aripiprazol	0.71
Bromperidol	0.50
Fluphenazine	0.43
Ziprasidone	0.04
Placebo	−0.74

Adapted from: Allison DB, Mentore JL, Heo M, et al. Antipsychotic-induced weight gain: a comprehensive research synthesis. Am. J. Psychiatry 1999;156:1686–1696

Weight Gain

Weight gain can be caused by some antidepressant drugs (e.g., TCA's and mirtazapine), but is most frequently caused by antipsychotic drugs. Weight gain can occur during use of any antipsychotic drug, but is more severe with second generation antipsychotics, especially clozapine and olanzapine (Table 17.9), since they have a higher affinity for receptors associated with weight gain (e.g., muscarin, histamine, serotonin) .

Weight gain is most clearly visible during the first 3 months of treatment with antipsychotics, after which a new plateau is reached. However, weight gain can endure for years during treatment with olanzapine or clozapine. Apart from the increased risk of comorbidities associated with weight gain, patients also consider weight gain as one of the most disturbing adverse events and an important reason for non-adherence to therapy (Hugenholtz et al., 2005).

Increased awareness for weight gain, and other metabolic adverse effects caused by antipsychotic drugs, lead in 2008 to the development of the first Dutch (Cahn, Ramlal & Bruggeman, 2008) protocol for routine somatic screening and treatment procedures in patients with schizophrenia in order to prevent long-term complications like cardiovascular disease and diabetes.

The cornerstone in treatment of drug-induced weight gain are life style interventions, including diets and promoting physical exercise. If those options are not possible or effective, then switching to a comparable drug with a more favorable metabolic profile is advisable (e.g., SNRI's/SSRI's instead of mirtazapine or TCA's, or aripiprazole instead of other antipsychotic drugs).

If switching to a more favorable drug is also not possible, then additional pharmacotherapy with a low dose of the antidiabetic drug metformin or the antiepileptic drug topiramate may be effective in achieving weight loss (Chen et al., 2013; Mahmood, Booker, Huang, & Coleman, 2013).

Multidisciplinary Care in Monitoring and Management of Psychotropic Drugs' Side-Effects

As outlined above the monitoring and management of side- effects of psychotropic drugs on physical health and mental functioning is a multifaceted, sometimes complicated process. For good quality pharmaceutical care a multidisciplinary approach is required, wherein the role and responsibilities of each discipline involved is clear. Where physicians and pharmacists are responsible for the prescription, delivery and quality of drugs, nurses are responsible for the administration. Furthermore, main caregivers, nurses, and/or support professionals are responsible for daily care and observation of behavioral and physical symptoms, possibly due to side-effects of drugs. In mental health care for people with intellectual disability the psychologist and/or behavioral scientist will mostly be responsible for the care plan of the client, including the assessment and management of challenging behavior and symptoms of mental disorders. It will be clear that when it comes to the monitoring and management of side-effects of psychotropic drugs, cooperation of all disciplines is required.

The past decade the role of specialized intellectual disability nurses, although not clearly defined, has become more prominent. Nurses are the main contacts of families and representatives, and they feel they have responsibilities towards clients with regard to advocacy and health promotion, assessment and case management, behavioral interventions, safety and risk management, communication and medication administration, and to take a leading role in the multidisciplinary team (Doody, Slevin, & Taggart, 2018; Taua, Hepworth, & Neville, 2012). In case nurses are educated and trained in specialized intellectual disability care, they may also take a role and share responsibilities with physicians in the monitoring of side-effects of psychotropic drugs provided that they have sufficient knowledge of effects of psychotropic drug and of alternative treatments of mental and behavioral disorders (Jenkins, 2000). However, studies have shown that support professionals' and nurses' knowledge was insufficient and that they were in need for education on this issue (de Kuijper & van der Putten, 2017; Donley et al., 2012).

The psychologist and behavioral scientist are very important disciplines with regard to the monitoring of effects of psychotropic drugs on clients' behavior and the management of side-effects. Especially in case of discontinuation of off-label use they have to look for alternative treatments and to support the team with regard to the management of clients' challenging behaviors. Also, when life style interventions are needed in case of overweight or risk of cardiovascular disorders due to metabolic side-effects, they have important roles regarding changes in the care plan and supporting the client and team to change behavioral patterns concerning food intake and exercises. Furthermore, their help will be needed in the management of sexual side-effects, when clients may be frustrated and display irritable or aggressive behavior.

In most guidelines on psychotropic drug use regular monitoring and a multidisciplinary approach is advised. However, the adherence to guidelines may be poor due to insufficient implementation (Ramerman, de Kuijper & Hoekstra, 2017; Ramerman et al., 2018a). Among others, changes in organizational policies are needed to facilitate professionals in offering the required quality of care.

A Tool for Customized Pharmaceutical Care

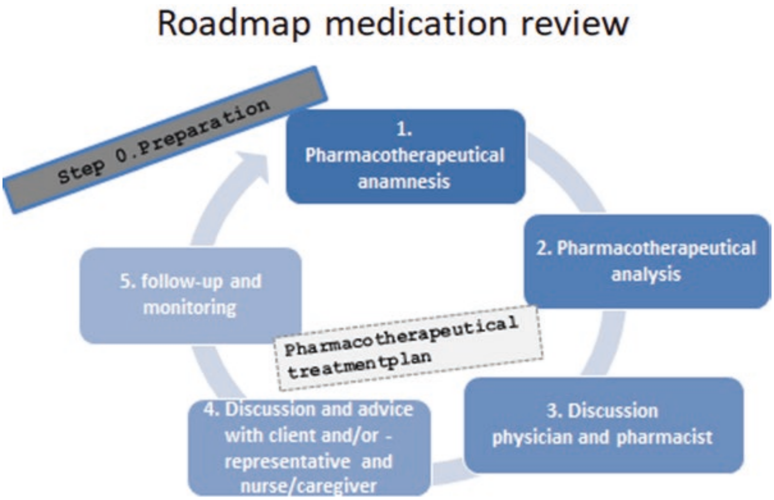
Although reduction of unnecessary use of medication should always been sought, many people with intellectual disability will have need for chronic medication use, including psychotropic drugs. Here, a good quality multidisciplinary evaluation of the medication use on indication, effects and side-effects of all prescribed drugs is needed. Disciplines who take part in such a medication review are the pharmacist, physician, nurse, and the support professional of the person with intellectual disability who is on chronic medication use. The review includes an interview with the patient and/or the patient-representative or main caregiver by the nurse, physician, and/or pharmacist. Preferably the medication review is in addition to an annual health check of the patient or is at least preceded by an evaluation and monitoring of the psychotropic drug use of the patient.

STRIP, a Systematic Tool to Reduce Inappropriate Prescribing, is a systematic method for reviewing medication prescribing, which may be very useful for customized pharmaceutical care in people with intellectual disability. The method includes a cycle of five steps, preceded by an assessment of the patient's health condition by studying the medical and pharmaceutical record, taking history and/or a physical examination (Fig. 17.1).

Step 1 can be carried out by the pharmacist, physician or specialized high graduated nurse. Step 2 is carried out by the pharmacist. Besides the pharmacist and physician, other disciplines involved in reviewing patients medication use (Step 3) may be the psychologist, support professional and nurse of the client. In step 4, discussion with the client and/or his representative and main caregiver, a psychologist should also be involved in case of psychotropic drug use. Step 5 is part of the regular monitoring of the medication use. After 1, 2, or 3 years, the cycle of five steps should be repeated.

The STRIP has been developed for use in elderly health care, but it is also a promising tool for application in intellectual disability health

Fig. 17.1 STRIP method



care (Scheifes, Egberts, Stolker, Nijman, & Heerdink, 2016; Zaal et al., 2016). The tool is targeted towards the identification of prescription errors and other drug-related problems. These are defined as:

1. Undertreatment, i.e., the missing of necessary medication for treatment or reduction of risk for side-effects caused by use of certain kinds of medication. An example is the need for prescription of an antacid agent to reduce the risk of stomach bleeding when the patient also uses an NSAID for treatment of pain.
2. Lack of effectiveness,
3. Overtreatment, e.g., there is no clear reasons for prescription (anymore).
4. The presence or risk of side-effects.
5. Clinically relevant contra-indications for medication, e.g., antipsychotic drugs in case of severe and untreatable seizures, or interactions between two or more agents.
6. Insufficient dosage (related to the indication for prescription).
7. Administration problems, e.g., the patients cannot swallow tablets or through probe.

When the prescription errors and drug-related problems are identified, the pharmacist will talk with the patient and the physician to discuss the errors and problems and will give advices for interventions to overcome the drug-related errors and problems. In intellectual disability medical practice, these interventions are not always implemented. This may be related to objections by patients, patient-representatives, caregivers, or other staff members, but also to organizational factors. In the study of Zaal et al. (2016), which took place in living facilities of service providers, after 6 months just 16% of advised interventions were implemented. In the study of Scheifes, Walraven, et al. (2016), which took place in a treatment facility for people with mild intellectual disabilities, almost 60% of advised interventions were implemented.

In many countries health care systems may be limited to allow for application of the STRIP method or even to apply for regular evaluations of (psychotropic) drug use in people with intellectual disability.

Yet, the quality of medication evaluations can be improved by cooperation of health care professionals affiliated to different health care systems within countries. Policy makers, health care insurances, intellectual disability service providers, and mental health care systems should facilitate the deployment of high graduated and specialized nurses, knowledge exchange and cooperation between pharmacists, physicians and nurses. In this way, good quality pharmaceutical care may be possible, at least to some degree, for people with intellectual disability in middle and lower income countries too.

Conclusions

To guarantee good quality pharmaceutical care in people with intellectual disability who use psychotropic drugs, a multidisciplinary approach is required: physicians, pharmacists, and main caregivers cooperate in medication reviews; physicians, pharmacists, main caregivers, and psychologists cooperate in observation and management of side-effects; physicians, nurses, and main caregivers cooperate in the monitoring of side-effects. However, nurses as well as support professionals should have sufficient skills and competence to reliably observe and monitor the side-effects. For this they should be educated in their vocational and specialist training. Also, there is need for validation and development of reliable instruments for assessments of neurological side-effects including sexual side-effects. The implementation of guidelines in the monitoring of psychotropic drug use should be enhanced by training of physicians and clinicians and changes in organizational policies.

References

Alper, K., Schwartz, K. A., Kolts, R. L., & Khan, A. (2007). Seizure incidence in psychopharmacological clinical trials: An analysis of food and drug administration (FDA) summary basis of approval reports. *Biological Psychiatry, 62*(4), 345–354.

Bella, A. J., & Shamloul, R. (2014). Psychotropics and sexual dysfunction. *Central European Journal of Urology, 66*(4), 466–471. https://doi.org/10.5173/ceju.2013.04.art22

Berm, E. J., Risselada, A. J., Mulder, H., Hak, E., & Wilffert, B. (2013). Phenoconversion of cytochrome P450 2D6: The need for identifying the intermediate metabolizer genotype. *The Journal of Clinical Psychiatry, 74*(10), 1025. https://doi.org/10.4088/JCP.13lr08555

Beumer, S., & Maes-Festen, D.A.M. (2017). Reduction or discontinuation of antipsychotics for challenging behaviour in adults with intellectual disability: Why does it fail? *The Lancet Psychiatry*, 4(3), e2 https://doi.org/10.1016/S2215-0366(17)30041-X

Brandt, C., Lahr, D., & May, T. W. (2015). Cognitive adverse events of topiramate in patients with epilepsy and intellectual disability. *Epilepsy & Behavior : E&B, 45*, 261–264. https://doi.org/10.1016/j.yebeh.2014.12.043

Brown, M., & McCann, E. (2018). Sexuality issues and the voices of adults with intellectual disabilities: A systematic review of the literature. *Research in Developmental Disabilities, 74*, 124–138. https://doi.org/10.1016/j.ridd.2018.01.009

Cahn, W., Ramlal, D., Bruggeman, R., et al. (2008). Preventie en behandeling van somatische complicaties bij antipsychoticagebruik. *Tijdschrift voor psychiatrie, 50*, 579–591.

Chen, C. H., Huang, M. C., Kao, C. F., Lin, S. K., Kuo, P. H., Chiu, C. C., & Lu, M. L. (2013). Effects of adjunctive metformin on metabolic traits in nondiabetic clozapine-treated patients with schizophrenia and the effect of metformin discontinuation on body weight: A 24-week, randomized, double-blind, placebo-controlled study. *The Journal of Clinical Psychiatry, 74*(5), e424–e430. https://doi.org/10.4088/JCP.12m08186

Chen, W. Y., Chen, L. Y., Liu, H. C., Wu, C. S., Yang, S. Y., Pan, C. H., … Kuo, C. J. (2017). Antipsychotic medications and stroke in schizophrenia: A case-crossover study. *PLoS One, 12*(6), e0179424. https://doi.org/10.1371/journal.pone.0179424

Cooper, S. A., McLean, G., Guthrie, B., McConnachie, A., Mercer, S., Sullivan, F., & Morrison, J. (2015). Multiple physical and mental health comorbidity in adults with intellectual disabilities: Population-based cross-sectional analysis. *BMC Family Practice, 16*, 110. https://doi.org/10.1186/s12875-015-0329-3

Cooper, S. A., Smiley, E., Morrison, J., Williamson, A., & Allan, L. (2007). Mental ill-health in adults with intellectual disabilities: Prevalence and associated factors. *The British Journal of Psychiatry : The Journal of Mental Science, 190*, 27–35. https://doi.org/10.1192/bjp.bp.106.022483

de Boer, M. K., Castelein, S., Bous, J., van den Heuvel, E. R., Wiersma, D., Schoevers, R. A., & Knegtering, H. (2013). The antipsychotics and sexual functioning questionnaire (ASFQ): Preliminary evidence for reliability and validity. *Schizophrenia Research, 150*(2–3), 410–415. https://doi.org/10.1016/j.schres.2013.08.037

de Boer, M. K., Castelein, S., Wiersma, D., Schoevers, R. A., & Knegtering, H. (2014). A systematic review of instruments to measure sexual functioning in patients using antipsychotics. *Journal of Sex Research, 51*(4), 383–389. https://doi.org/10.1080/00224499.2013.865111

de Boer, M. K., Castelein, S., Wiersma, D., Schoevers, R. A., & Knegtering, H. (2015). The facts about sexual (dys)function in schizophrenia: An overview of clinically relevant findings. *Schizophrenia Bulletin, 41*(3), 674–686. https://doi.org/10.1093/schbul/sbv001

de Kuijper, G., Evenhuis, H., Minderaa, R., & Hoekstra, P. (2014). Effects of controlled discontinuation of long-term used antipsychotics for behavioural symptoms in individuals with intellectual disability. *Journal Intellectual Disability Research, 58*(1), 71–83. https://doi.org/10.1111/j.1365-2788.2012.01631.x de Kuijper, G., Mulder, H., Evenhuis, H., Scholte, F., Visser, F., & Hoekstra, P. J. (2013). Determinants of physical health parameters in individuals with intellectual disability who use long-term antipsychotics. *Research in Developmental Disabilities, 34*(9), 2799–2809. https://doi.org/10.1016/j.ridd.2013.05.016

de Kuijper, G., & van der Putten, A. A. J. (2017). Knowledge and expectations of direct support professionals towards effects of psychotropic drug use in people with intellectual disabilities. *Journal of Applied Research in Intellectual Disabilities, 30*(Suppl 1), 1–9. https://doi.org/10.1111/jar.12357

de Kuijper, G. M., & Hoekstra, P. J. (2016). Assessment of drug-associated extrapyramidal symptoms in people with intellectual disability: A comparison of an informant-based scale with clinical rating scales. *Journal of Clinical Psychopharmacology, 36*(5), 508–512. https://doi.org/10.1097/JCP.0000000000000558

de Kuijper, G. M., & Hoekstra, P. J. (2017). Physicians‘ reasons not to discontinue long-term used off-label antipsychotic drugs in people with intellectual disability. *Journal of Intellectual Disability Research, 61*(10), 899–908. https://doi.org/10.1111/jir.12385

de Leon, J., Greenlee, B., Barber, J., Sabaawi, M., & Singh, N. N. (2009). Practical guidelines for the use of new generation antipsychotic drugs (except clozapine) in adult individuals with intellectual disabilities. *Research in Developmental Disabilities, 30*(4), 613–669. https://doi.org/10.1016/j.ridd.2008.10.010

de Winter, C. F., Bastiaanse, L. P., Hilgenkamp, T. I., Evenhuis, H. M., & Echteld, M. A. (2012). Overweight and obesity in older people with intellectual disability. *Research in Developmental Disabilities, 33*(2), 398–405. https://doi.org/10.1016/j.ridd.2011.09.022

Donley, M., Chan, J., & Webber, L. (2012). Disability support workers' knowledge and education needs about psychotropic medication. *British Journal of Learning Disabilities, 40*(4), 286–291. https://doi.org/10.1111/j.1468-3156.2011.00707.x

Doody, O., Slevin, E., & Taggart, L. (2018). Families‘ perceptions of the contribution of intellectual disability clinical nurse specialists in Ireland. *Journal of Clinical Nursing, 27*(1–2), e80–e90. https://doi.org/10.1111/jocn.13873

Gil-Llario, M. D., Morell-Mengual, V., Ballester-Arnal, R., & Diaz-Rodriguez, I. (2018). The experience of sexuality in adults with intellectual disability. *Journal of Intellectual Disability Research, 62*(1), 72–80. https://doi.org/10.1111/jir.12455

Haider, S. I., Ansari, Z., Vaughan, L., Matters, H., & Emerson, E. (2014). Prevalence and factors associated with polypharmacy in victorian adults with intellectual disability. *Research in Developmental Disabilities, 35*(11), 3071–3080. https://doi.org/10.1016/j.ridd.2014.07.060

Haw, C., & Stubbs, J. (2005). A survey of off-label prescribing for inpatients with mild intellectual disability and mental illness. *Journal of Intellectual Disability Research, 49*(Pt 11), 858–864. https://doi.org/10.1111/j.1365-2788.2005.00723.x

Hsu, W. T., Esmaily-Fard, A., Lai, C. C., Zala, D., Lee, S. H., Chang, S. S., & Lee, C. C. (2017). Antipsychotics and the risk of cerebrovascular accident: A systematic review and meta-analysis of observational studies. *Journal of the American Medical Directors Association, 18*(8), 692–699. https://doi.org/10.1016/j.jamda.2017.02.020

Hugenholtz, G. W., Heerdink, E. R., Meijer, W. E., Stolker, J. J., Egberts, A. C., & Nolen, W. A. (2005). Reasons for switching between antipsychotics in daily clinical practice. *Pharmacopsychiatry, 38*(3), 122–124. https://doi.org/10.1055/s-2005-864122

Jenkins, R. (2000). Use of psychotropic medication in people with a learning disability. *British Journal of Nursing (Mark Allen Publishing), 9*(13), 844–850. https://doi.org/10.12968/bjon.2000.9.13.5512

Koch, A. D., Vogel, A., Becker, T., Salize, H. J., Voss, E., Werner, A., … Schutzwohl, M. (2015). Proxy and self-reported quality of life in adults with intellectual disabilities: Impact of psychiatric symptoms, problem behaviour, psychotropic medication and unmet needs. *Research in Developmental Disabilities, 45–46*, 136–146. https://doi.org/10.1016/j.ridd.2015.07.022

Lambert, T. L., Farmer, K. C., & Brahm, N. C. (2013). Evaluation of serum prolactin levels in intellectually disabled patients using antipsychotic medications. *International Journal of Endocrinology and Metabolism, 11*(1), 57–61. https://doi.org/10.5812/ijem.4366

Leucht, S., Cipriani, A., Spineli, L., Mavridis, D., Orey, D., Richter, F., … Davis, J. M. (2013). Comparative efficacy and tolerability of 15 antipsychotic drugs in schizophrenia: A multiple-treatments meta-analysis. *Lancet (London, England), 382*(9896), 951–962. https://doi.org/10.1016/S0140-6736(13)60733-3

Maher, R., Hanlon, J., & Hajjar, E. (2014). Clinical consequences of polypharmacy in the elderly. *Expert Opinion Drug Safe, 13*(1), 57–65. https://doi.org/10.1517/14740338.2013.827660

Mahmood, S., Booker, I., Huang, J., & Coleman, C. I. (2013). Effect of topiramate on weight gain in patients receiving atypical antipsychotic agents. *Journal of Clinical Psychopharmacology, 33*(1), 90–94. https://doi.org/10.1097/JCP.0b013e31827cb2b7

Matson, J. L., & Cervantes, P. E. (2013). Current status of the matson evaluation of drug side effects (MEDS). *Research in Developmental Disabilities, 34*(5), 1849–1853. https://doi.org/10.1016/j.ridd.2013.02.030

Matson, J. L., & Mahan, S. (2010). Antipsychotic drug side effects for persons with intellectual disability. *Research in Developmental Disabilities, 31*(6), 1570–1576. https://doi.org/10.1016/j.ridd.2010.05.005

Matson, J. L., Mayville, E. A., Bielecki, J., Barnes, W. H., Bamburg, J. W., & Baglio, C. S. (1998). Reliability of the matson evaluation of drug side effects scale (MEDS). *Research in Developmental Disabilities, 19*(6), 501–506.

Mentzel, T. Q., Lieverse, R., Levens, A., Mentzel, C. L., Tenback, D. E., Bakker, P. R., … van Harten,

P. N. (2016). Reliability and validity of an instrument for the assessment of bradykinesia. *Psychiatry Research, 238*, 189–195. https://doi.org/10.1016/j.psychres.2016.02.011

Mentzel, T. Q., Mentzel, C. L., Mentzel, S. V., Lieverse, R., Daanen, H. A., & van Harten, P. N. (2016). Instrumental assessment of bradykinesia: A comparison between motor tasks. *IEEE Journal of Biomedical and Health Informatics, 20*(2), 521–526. https://doi.org/10.1109/JBHI.2015.2412656

O'Dwyer, M., Maidment, I. D., Bennett, K., Peklar, J., Mulryan, N., McCallion, P., … Henman, M. C. (2016). Association of anticholinergic burden with adverse effects in older people with intellectual disabilities: An observational cross-sectional study. *The British Journal of Psychiatry: The Journal of Mental Science, 209*(6), 504–510. https://doi.org/10.1192/bjp.bp.115.173971

O'Dwyer, M., Peklar, J., McCallion, P., McCarron, M., & Henman, M. C. (2016). Factors associated with polypharmacy and excessive polypharmacy in older people with intellectual disability differ from the general population: A cross-sectional observational nationwide study. *BMJ Open, 6*(4), e010505. https://doi.org/10.1136/bmjopen-2015-010505

Pereira, V. M., Arias-Carrion, O., Machado, S., Nardi, A. E., & Silva, A. C. (2014). Bupropion in the depression-related sexual dysfunction: A systematic review. *CNS & Neurological Disorders Drug Targets, 13*(6), 1079–1088.

Poyurovsky, M., Nave, R. F., Epstein, R. F., Tzischinsky, O. F., Schneidman, M., Barnes, T. R., … Lavie, P. (2000). Actigraphic monitoring (actigraphy) of circadian locomotor activity in schizophrenic patients with acute neuroleptic-induced akathisia. *European Neuropsychopharmacology, 10*(3), 171–176.

Ramerman, L., de Kuijper, G., & Hoekstra, P. J. (2017). Adherence of clinicians to guidelines for the prescription of antipsychotic drugs to people with intellectual disabilities. *Advances in Mental Health and Intellectual Disabilities, 11*(3), 110–125. https://doi.org/10.1108/AMHID-02-2017-0005

Ramerman, L., Hoekstra, P. J., & de Kuijper, G. (2018b). Exploring barriers and facilitators in the implementation and use of guideline recommendations on antipsychotic drug prescriptions in people with intellectual disabilities. *Journal of Applied Research in Intellectual Disabilities, 31*(6), 1062–1070.

Ramerman, L., Hoekstra, P. J., & de Kuijper, G. (2018a). Health-related quality of life in people with intellectual disability who use long-term antipsychotic drugs for challenging behaviour. *Research in Developmental Disabilities, 75*, 49–58. https://doi.org/10.1016/j.ridd.2018.02.011

Scheifes, A., Egberts, T. C., Stolker, J. J., Nijman, H. L., & Heerdink, E. R. (2016). Structured medication review to improve pharmacotherapy in people with intellectual disability and behavioural problems. *Journal of Applied Research in Intellectual Disabilities, 29*(4), 346–355. https://doi.org/10.1111/jar.12183

Scheifes, A., Walraven, S., Stolker, J. J., Nijman, H. L., Egberts, T. C., & Heerdink, E. R. (2016). Adverse events and the relation with quality of life in adults with intellectual disability and challenging behaviour using psychotropic drugs. *Research in Developmental Disabilities, 49–50*, 13–21. https://doi.org/10.1016/j.ridd.2015.11.017

Sheehan, R., Hassiotis, A., Walters, K., Osborn, D., Strydom, A., & Horsfall, L. (2015). Mental illness, challenging behaviour, and psychotropic drug prescribing in people with intellectual disability: UK population based cohort study. *BMJ (Clinical Research Ed.), 351*, h4326. https://doi.org/10.1136/bmj.h4326

Sheehan, R., Horsfall, L., Strydom, A., Osborn, D., Walters, K., & Hassiotis, A. (2017). Movement side effects of antipsychotic drugs in adults with and without intellectual disability: UK population-based cohort study. *BMJ Open, 7*(8), e017406–2017-017406. https://doi.org/10.1136/bmjopen-2017-017406

Shore, J., Vo, A., Yellowlees, P., Waugh, M., Schneck, C., Nagamoto, H., & Thomas, M. (2015). Antipsychotic-induced movement disorder: Screening via telemental health. *Telemedicine Journal and e-Health: The Official Journal of the American Telemedicine Association, 21*(12), 1027–1029. https://doi.org/10.1089/tmj.2014.0242

Smiley, E., Cooper, S. A., Finlayson, J., Jackson, A., Allan, L., Mantry, D., … Morrison, J. (2007). Incidence and predictors of mental ill-health in adults with intellectual disabilities: Prospective study. *The British Journal of Psychiatry : The Journal of Mental Science, 191*, 313–319. https://doi.org/10.1192/bjp.bp.106.031104

Swen, J. J., Nijenhuis, M., de Boer, A., Grandia, L., Maitland-van der Zee, A. H., Mulder, H., … Guchelaar, H. J. (2011). Pharmacogenetics: From bench to byte—An update of guidelines. *Clinical Pharmacology and Therapeutics, 89*(5), 662–673. https://doi.org/10.1038/clpt.2011.34

Taua, C., Hepworth, J., & Neville, C. (2012). Nurses' role in caring for people with a comorbidity of mental illness and intellectual disability: A literature review. *International Journal of Mental Health Nursing, 21*(2), 163–174. https://doi.org/10.1111/j.1447-0349.2011.00779.x

Thom, R. P., Grudzinskas, A. J., Jr., & Saleh, F. M. (2017). Sexual behavior among persons with cognitive impairments. *Current Psychiatry Reports, 19*(5). https://doi.org/10.1007/s11920-017-0777-7

Tobi, H., Scheers, T., Netjes, K. A., Mulder, E. J., de, B. A., & Minderaa, R. B. (2005). Drug utilisation by children and adolescents with mental retardation: A population study. *European Journal of Clinical Pharmacology, 61*(4), 297–302.

Trollor, J. N., Salomon, C., & Franklin, C. (2016). Prescribing psychotropic drugs to adults with an intellectual disability. *Australian Prescriber, 39*(4), 126–130. https://doi.org/10.18773/austprescr.2016.048

Visser, M., Marinus, J., Stiggelbout, A. M., & Van Hilten, J. J. (2004). Assessment of autonomic dysfunction in parkinson‘s disease: The SCOPA-AUT. *Movement Disorders : Official Journal of the Movement Disorder Society, 19*(11), 1306–1312. https://doi.org/10.1002/mds.20153

Warle-van Herwaarden, M. F., Kramers, C., Sturkenboom, M. C., van den Bemt, P. M., De Smet, P. A., & Dutch HARM-Wrestling Task Force. (2012). Targeting outpatient drug safety: Recommendations of the dutch HARM-wrestling task force. *Drug Safety, 35*(3), 245–259. https://doi.org/10.2165/11596000-000000000-00000

Young, S. L., Taylor, M., & Lawrie, S. M. (2014). “First do no harm”. A systematic review of the prevalence and management of antipsychotic adverse effects. *Journal of Psychopharmacology (Oxford, England), 29*(4), 353–362. https://doi.org/10.1177/0269881114562090

Zaal, R. J., Ebbers, S., Borms, M., Koning, B., Mombarg, E., Ooms, P., … Evenhuis, H. M. (2016). Medication review using a systematic tool to reduce inappropriate prescribing (STRIP) in adults with an intellectual disability: A pilot study. *Research in Developmental Disabilities, 55*, 132–142. https://doi.org/10.1016/j.ridd.2016.03.014

Part V

Causes and Risk Factors of Intellectual Disabilities

The Effects of Genetic Disorders on Language

18

Natalia Freitas Rossi and Célia Maria Giacheti

Analyzing the effects of genetic disorders on language is like putting together the pieces of a big puzzle or like moving—most of the time—in uncharted waters. This analogy that represents the challenges that researchers have to face while approaching the issue is due to a set of factors that can influence language development; genetic influence is one of the factors in this context.

It represents the past and present views of the nature vs. nurture debate. Fortunately, due to advances in interdisciplinary studies using molecular genetics and brain imaging techniques, this issue is currently better understood.

Language From a Neurobiological Perspective and Its Evolution

The evolution of communication and language systems among different species can be approached from a number of different perspectives, ranging from a biological to an anthropological point of view. The fundamental question of how genetic and environment factors work together is still open, but a little better understood now. The open-ended complex questions regarding the gene-brain-language interaction have stimulated the nature vs. nurture debate.

Biological and environment factors have received different weights among theoretical models to explain language acquisition and its developmental trajectory. However, both factors are recognized as part of this process, even if the theoretical models seem to indicate either one or the other as the driving factor. On the one hand, we have those who defend the theory that language evolution results from interplay between genetic and brain modifications; this theory reinforces Chomsky's assumption that language is fundamentally a biological structure. On the other hand, this assumption is too simple to explain human language acquisition, and it has been suggested that human language acquisition is the result of a long process of cultural evolution (Chater & Christiansen, 2010; Christiansen & Kirby, 2003).

In fact, the evolution of the complex communication system is the result of a long biological and cultural process that makes humans unique; this evolution was a turning point that made *Homo sapiens* a symbolic species. The evolution of symbolic language (gestures, spoken, and written symbols) over thousands of years provided substantial evolutionary advantage from monkeys to other primitive hominids and finally to humans, in the Darwinian process of natural selection. Due to its complexity, coevolution of the brain and human language seems to be a plausible hypothesis (Deacon, 1998).

N. F. Rossi (✉) · C. M. Giacheti
Speech, Language and Hearing Sciences, Sao Paulo State University, (UNESP), Marilia, Sao Paulo, Brazil

J. L. Matson (ed.), *Handbook of Intellectual Disabilities*, Autism and Child Psychopathology Series, https://doi.org/10.1007/978-3-030-20843-1_18

The evolutionary approach toward language development takes into consideration multiple factors involved in communication, from vocal anatomical structural modifications to the evolution of social cognition. So far, the following questions regarding the emergence and evolution of speech and language in the human species remain unanswered: did the brain evolve to accommodate the evolution of language and the complexity of social relations, or were language and social relations more complex changes as a result of genetic influences and brain structural modifications, or is this all a reflection of coevolution?

The coevolution hypothesis argues that the need to communicate may have led humans to change their communication strategy from simple to more complex forms. Human communication requires sophisticated capabilities that involve more than nonlinguistic communication skills (e.g., facial expression, gestures); it requires a grammar system domain and the ability to deal with the idiosyncrasies of language and its discontinuities (Deacon, 1998).

It is known that genetic changes with respect to the evolution of the brain occurred with no major genome-wide changes; this may seem paradoxical with regard to language evolution. In fact, what is seen in the primate-specific lineage is that new genes are recruited during the early development of the brain, which indicates that new genes have an important function during the development of the human fetal brain (Zhang, Landback, Vibranovski, & Long, 2011).

Social behavior is shaped by a combination of motivation, emotion, and communication skills, which leads a species to move from a rigid eusocial behavior to constructing representations of the relations between oneself and others, and to use those representations flexibly to guide social behavior (Adolphs, 2001).

Prosocial motivation is a well-known, remarkable aspect of the human communication system. It is based on fundamentally cooperative intentions, and this makes human communication skills different from those of other primates (Tomasello, 2010). Prosocial motivation and caregiving behaviors (inhibiting aggression, and facilitating affective communication and cooperation between members of a similar social group) is driven by empathy, which is not exclusive to humans, but certainly a hallmark of humans. Empathy is supported by the neurobiological architecture, including the brainstem, amygdala, anterior cingulate cortex, hypothalamus, striatum, insula, and orbitofrontal cortex, as well as the autonomic nervous system and neuroendocrine processes (Decety, Bartal, Uzefovsky, & Knafo-Noam, 2016).

Despite the evolution in communication systems among humans from primitive modalities to highly sophisticated social interactions in current times, including communication via internet modalities (Yeger, 2015), the ability to match the emotional state of another (empathy) and to understand the other person's mental state (theory of mind) still remains crucial in everyday interactions (Goldstein & Winner, 2012); together these two social cognitive domains with language probably formed an evolutionary cycle, wherein, advances in one fed advances in the other. It is unclear what human cognition (social or otherwise) would be like without the powerful cultural augmentation that language provides.

Evolution of the temporal and frontal regions of the brain is of significance because these brain areas support the evolution of the mechanisms of comprehension and production of spoken language, as well as social cognition that brought to our species, an important adaption for living in a society (Poeppel, 2014). In terms of maturation, it is well known that the frontal cortex is a late-developing region of the neocortex (Fuster, 2002). The frontal lobe is responsible for sophisticated spoken language organization in the form of a narrative discourse (Friend & Bates, 2014), executive functions, and social cognition (Miller & Cummings, 2017; Yeh, Tsai, Tsai, Lo, & Wang, 2017). It is highly unlikely that the sophisticated cognitive and communication system of humans with complex language capacities is only due to the large brain size in our species, in part, because the size of the human brain is not so different from that of chimpanzees. Nevertheless, in spite of similarities, there are important differences in behavior and social skills between humans and chimpanzees. Changes at the protein level with evolution of regulatory networks (Zhang et al., 2011), a more complex organization and distribution pattern of neurons and glial cells, and changes in gene expression seem to be reasonable explanations for the more complex brain function in humans (Verendeev & Sherwood, 2017).

This is in agreement with the perspective of an evolutionary advancement that favors brain connectivity rather than size itself. For example, the neural circuit involving the inferior frontal lobe and the posterior superior temporal cortex via a dorsally located fiber tract was a crucial evolutionary advancement toward the development of the uniquely human capability of language (Friederici, 2017).

While it is known that communication involves more than linguistic skills (phonology, semantics, syntax, and pragmatics), for many years, the ontogeny of the brain as well as the cortical language network have been constructed based on different models not only to comprehend the evolution of language in the primate brain, but also to better understand the course of maturation of speech and language areas in the brain and their developmental trajectories (Skeide & Friederici, 2016).

Language, cognition, and motor control are lateralized to the left hemisphere of the brain, possibly due to a genetic predisposition for hemispheric specialization during cognitive, linguistic, and brain development (Karmiloff-Smith, 1992). Localization of neural processing of language has been shown in functional magnetic resonance imaging and electroencephalography studies, wherein more diffuse and bilateral activation patterns with regard to language processing are seen in children, but at the same areas of the brain as those of adults (Holland et al., 2007).

Understanding the changes in brain functioning in the typical and deviant language developmental context has significant implications on developmental theories and in understanding how language is represented in the brain along the maturational trajectory (Rossi, 2016). It is well known that pathological conditions contribute significantly not only in understanding brain evolution, but also in comprehending brain functioning, and supporting significant advances in neuroscience (de Sousa & Cunha, 2012). However, adult models of high-order cognitive function correlations (e.g., language) cannot explain deviations in language development in children; thus, models that can clarify the neurobiological mechanisms from a developmental perspective are lacking (Friederici, 2006; Karmiloff-Smith, 1997; Karmiloff-Smith, 2018).

In this context, speech and language in neurodevelopmental disorders with a known genetic cause, as well as others with complex genetic etiology, are not understood enough. It would be useful to identify the critical gene for brain development with regard to language, understand the gene-brain-language relationship, and to define the neurodevelopmental phenotype, including the language phenotype, and its association with neurocognitive and behavioral patterns. Furthermore, by studying genetic disorders and their impact on language development, we are contributing to the nature-nurture debate and theories concerning the development of language.

Defining the Language Phenotype in Genetic Disorders

A genetic disorder is defined as a disease caused by alterations in one or more genes. Genetic disorders in humans are classified as chromosomal disorders, monogenic conditions (typical Mendelian heritance), multifactorial or polygenic disorders, and mitochondrial disorders. Chromosomal disorders could be due to missing chromosomes (deletion) or gaining extra segments or an entire chromosome (duplication). Single-gene diseases (monogenic) have a typical Mendelian inheritance pattern (dominant or recessive) in the autosomal or sex chromosome. On the other hand, multifactorial disorders have a complex interaction between genes (polygenic) and the environment, and finally, mitochondrial disorders are a small part of genetic disorders (Baty, Carey, & McMahon, 2010).

Online Mendelian Inheritance in Man (OMIM) is a database of all known Mendelian disorders and over 15,000 currently identified genes; it is a genuine representation of the advances in genetic disorders. OMIM was created in 1985 by a collaboration between the National Library of Medicine and the William H. Welch Medical Library at Johns Hopkins University and made publicly available on the internet since 1987. In 1995, OMIM was developed for the World Wide Web by the National Center for Biotechnology Information (NCBI). OMIM is authored and edited at the McKusick-Nathans Institute of Genetic Medicine, Johns Hopkins University School of Medicine, under the direction of Dr. Ada Hamosh.

One of the important challenges faced by OMIM is how best to catalog multifactorial disorders and phenotypes, especially with epigenetic mechanisms bringing complex phenotypes and genotypes into focus, in addition to the need for a large cohort of subjects. The cataloging system was suggested by McKusick in McKusick, 2007; however, it is still valid in recent times and can be extended to the context of the language phenotype, especially since a growing body of genetic studies has emerged over the last few years providing a more comprehensive view of the genetic map and the genes involved in the language evolution process, as well as its disorders.

Studying the role of genetic influence in the context of language disorders requires a comprehensive approach taking into account a set of factors; low scores on language performance (standardized tests and naturalistic language sampling) is one of these factors. Research needs to be focused on whether the language disorder is accompanied by other developmental difficulties, so that a distinction can be made between children whose language difficulties can be attributed to a known cause and those with unexpected, unexplained language problems, and also whether there are any associated significant environmental factors (low socioeconomic status or low educational level in the family).

Therefore, for the purposes of diagnosis and intervention in patients with language impairment, speech-language pathologists and other professionals have to take into account the family history as a fundamental part of the medical history investigation and to identify any history of speech and language difficulties and comorbidities across the generations. In the course of the clinical and academic routine, we teach speech-language students to construct the family pedigree using a standardized nomenclature (Bennett, French, Resta, & Doyle, 2008). A practical application of pedigree elaboration in our laboratory routine has been its benefit in identifying the genetic risks for language disorders and formulating an intervention plan in the early stages of language development. In this context, a 2-year-old late-talking child with a positive family history of speech or language impairments and with no obvious cause that could explain the language acquisition problems might require language follow-up to distinguish between a persistent language difficulty (disorder) and a transient language difficulty (normal course, but delayed).

Basically, experimental designs in studies evaluating spoken language development in genetic disorders have to consider five language domains (phonology, morphology, syntax, semantics, and pragmatics) across two processes (receptive and expressive) with different levels of organization and complexity (words, sentences, and narratives).

A didactic description of each component was provided by Hoff (2013). The phonology domain constitutes the sound system of language, while the system of combining units of meaning is morphology and the system of combining words into sentences is syntax. The content domain of words and associated knowledge constitutes the lexicon, and finally, how to use language to communicate is part of the pragmatic and sociolinguistic development.

It is useful for researchers and students of language development, to make distinctions among the sub components of language (Hoff, 2013). The vast majority of studies regarding language development and related disorders have focused on the receptive or expressive language process. Most of the time, these two language processes and the language components (phonology, semantic, syntactic, and pragmatic) have been described separately, depending on the major objective of the research.

Language assessment usually includes a set of standardized measures to allow a relatively objective and reliable assessment of the language component, and more qualitative observations with naturalistic language sampling (e.g., conversation and narration) to capture the functional aspects of communication that are not always picked up on formal assessment. Understanding the development of different language sub components with regard to the receptive and expressive processes and its interaction with the cognitive developmental profile (neurocognition and social cognition), as well as with behavioral and personality traits is the accurate way to understand the nature of language problems and describe the language phenotype of genetic disorders.

Language phenotype delineation is challenging since it is influenced by critical genes involved in brain development and by environmental factors. Therefore, the association between genetic disorders and language development necessarily refers us to a research context where a focus on interdisciplinary mechanisms is necessary to understand the different mechanisms involved in communication, as well as the characterization of these manifestations (Giacheti & Rossi, 2012).

It is well known that understanding the genetic influences of a complex genetic trait affecting behavior, cognition, and language at the molecular level is not possible without a very well characterized phenotype (Giacheti & Rossi, 2012; Mervis & Robinson, 2005). The key role of genetics in language development is complex and surrounded by controversies; therefore, a clear specification of the target phenotype is necessary (Hauser et al., 2014), which in turn requires consensus on the terminology used to describe it. The only way to understand the nature of the impairment and, in turn, depict a language phenotype is to have differentiating terminologies with clear meanings (Bishop, 2014).

First, let us make a distinction among the concepts with regard to speech, language, and communication. According to Bishop, Snowling, Thompson, and Greenhalgh (2016), "speech," "language," and "communication" are not interchangeable and the distinction among these terms and their meanings is the starting point of this research topic. Language is the comprehension and use of words and sentences to convey ideas and information. The different modalities of language are spoken, written, and sign; speech is part of the spoken language modality. Speech refers to the production of vocal sounds, a process that involves both motor (articulatory) and linguistic skills. Language and speech are both facets of communication, which encompasses the broader set of nonverbal and verbal means of conveying information and emotions (Bishop, 2017).

The terminological debate over language impairment in children is a current and necessary movement, which has important implications on genetic advances in the study of child language problems (Bishop, 2014; Bishop et al., 2016; Reilly, Bishop, & Tomblin, 2014). The debate related to the pros and cons of developmental language disorder as a primary manifestation or as part of a medical condition that may co-occur with other deficits in other developmental domains, such motor or intellectual impairment, is not a new phenomenon; however, there is a growing consensus among researchers regarding the use of a neurobiological approach to language and its disorders. Therefore, if the aim is to identify genetic specificities and neural correlates in typical and atypical developmental trajectories, it is necessary—not to say mandatory—to know the distinctions between language disorders (conditions or symptoms).

The key point of this problematic question has been explored in the studies by Dorothy Bishop and depicted in two studies published by an international group of experts, the CATALISE panel (Bishop et al., 2016; Bishop et al., 2017), created to discuss consensus on diagnostic terminology for language disorders in children. Based on the results of the project, the group has recommended the following terms: "Developmental Language Disorders (DLD)" for children with language difficulties not associated with a biomedical condition, and "Language Disorder," when associated with a biomedical condition (e.g., genetic syndromes, hearing loss, intellectual impairment, or autism spectrum disorders [ASD]).

Regarding terminology, the trend in literature seems to favor the use of DLD to refer to primary language disorders or specific language impairment, and will be used in this chapter as well. However, this is not the position in the 5th edition of the Diagnostic and Statistical Manual of Mental Disorders (DSM-5) (APA, 2013). In effect, in DSM-5, the term "Language Disorder," which is part of the Communication Disorder diagnostic category, is supported by four criteria, including: (1) persistent difficulties across modalities (spoken, written, sign language) with deficits in both language process comprehension and production; (2) functional language deficits due to language abilities below the chronological age; (3) manifestation of symptoms in the first years of life; and (4) difficulties not explained by intellectual impairment or medical condition. This fourth criterion drives us to DLD, but the position is not clear. There is no terminology distinction in DSM-5, which has motivated the continuing debate on this issue (Bishop, 2014).

ASD might be a good analogy with regard to the DLD debate, from the perspective of describing the phenotype. ASD is one of the neurodevelopmental disorders in DSM-5 (APA, 2013), and the diagnostic criteria include persistent deficits in social communication and social interaction in multiple contexts, and restricted and repetitive patterns of behavior. Symptoms are present early, although they may not be fully apparent. The identified symptoms cause clinically significant impairment in social, occupational, or other important areas of functioning. The specifier "associated with any known medical or genetic condition or environmental factor" should be used when the person has a known genetic disease (e.g., Rett syndrome, Fragile X syndrome, Down syndrome) or a history of environmental exposure (e.g., valproic acid, fetal alcohol syndrome, very low birth weight).

Therefore, researchers need to answer important questions while describing a phenotype, for example: is studying ASD manifestations as part of a genetic disorder with a well-known genetic etiology like Fragile X syndrome commensurate to studying the ASD phenotype? Dilemmas like this have to be considered in the study design so that researchers can identify the susceptibility genes for a developmental phenotype, and describe the natural history of the condition.

This issue is beyond just an academic question, as the researcher observes the actual phenomena, which puts it in a different perspective. Moreover, different genetic mechanisms need to be considered, even if the language manifestation is similar (Bishop et al., 2016).

Insights from Language Phenotypes in Down Syndrome, Fragile X Syndrome, and Williams Syndrome

Because language development is a complex process that depends on both neurobiological and social factors, the acquisition and development of language is an important developmental parameter that is susceptible to intervening factors (internal and external), which drive the developmental path of language and may manifest as deviant language development that could persist in some children throughout their lives (Bishop, 2014).

Individuals who exhibit persistent and early-onset developmental impairment are part of the diagnostic category of the DSM-5 (APA, 2013) "Neurodevelopmental Disorders." This diagnosis is attributed to individuals who present impairments in one or more domains of development (e.g., motor, intellectual, social cognition, behavioral, and language) when compared to their peers of the same age and gender in the same linguistic and social culture. This group includes cases with intellectual disability, communication disorders (language, speech, fluency, and communication disorders), autism spectrum disorder, attention deficit hyperactivity disorder, specific learning disorders, motor disorders, and other neurodevelopmental disorders (DSM-5; APA, 2013).

With regard to neurodevelopmental disorders, it is estimated that approximately 10 to 15% of the children born alive have a deviant neurodevelopmental trajectory, and 30 to 40% of these cases are associated with a known genetic etiology, such as chromosomal and genetic alterations, which result in deviant cerebral development, right from conception. The remaining cases are due to an interaction between genetic predisposition and environmental factors, such as biological events or chemicals with neurotoxic effects (e.g., alcohol consumption during pregnancy or maternal viral infections) (Andersen, 2003), which impact brain development in different ways depending on the developmental stage at which the event occurred and the epigenetic mechanisms (Grandjean & Landrigan, 2014; Rice & Barone Jr, 2000; Rodenhiser & Mann, 2006).

Certainly, a well-defined genetic condition in terms of genetic etiology might be a good neurobiological model to explore other complex conditions. Research into phenotype-genotype correlations in neurodevelopmental disorders has contributed to our knowledge regarding the contribution of genetic and neurobiological factors to variations in typical and atypical development (Järvinen-Pasley et al., 2008).

However, the results reported over the years have shown that impairments in different developmental domains manifest differently in each clinical group. A possible explanation for this is the existence of different etiopathological mechanisms that determine the neurofunctional differences that contribute to the expression of different neurodevelopmental phenotypes, including language (Knowland & Thomas, 2011; Luyster, Seery, Talbott, & Tager-Flusberg, 2011). Differences with regard to neurodevelopmental phenotypes are important considering their implications in the intervention methods used in these individuals (Bishop & Rutter, 2008; Mervis & Robinson, 2005; Rossi & Giacheti, 2017).

Studies that have focused on the delineation of neurodevelopmental phenotypes and their specificities taking into account the main developmental domains (e.g., motor, intellectual, social, behavioral, and cognitive) have gained importance over the years. In this regard, some neurodevelopmental disorders have been more systematically investigated, which was justified by Bishop (2010) based on some factors: (1) population frequency, which explains the large number of studies on individuals with Down syndrome; (2) a biological model to discuss and sustain theoretical models that contributes to the understanding of the neurobiological processes of language and related functions, as in the case of Williams syndrome; (3) conditions with a suggested genetic basis that have not yet been confirmed, such as ASD and primary language developmental disorders, and finally (4) the impact on public health, which justifies the interest in the study of the fetal alcohol spectrum disorders.

Cross-syndrome comparison of developmental domains is a very useful methodology to delineate neurodevelopmental phenotypes, and to identify overlaps or specifications of a deviant neurodevelopmental phenotype (Camp, Karmiloff-Smith, Thomas, & Farran, 2016). However, there are few studies exploring cross-language comparisons between genetic disorders in comparison with those describing language disorders in a specific clinical group.

The most frequent cross-syndrome studies of language and its disorders with well-known genetic etiologies have been conducted with Down syndrome (OMIM # 190685) (Johns Hopkins University, 2018a), Williams syndrome (OMIM # 194050) (Johns Hopkins University, 2018b), and Fragile X syndrome (OMIM #300624) (Johns Hopkins University, 2018c). Down syndrome and Fragile X syndrome are the most common genetic causes of intellectual disability. Down syndrome occurs in approximately 1 in 700 live births and Fragile X syndrome in approximately 1 in 4000 males and 1 in 8000 females (Baty et al., 2010). Williams syndrome is considered a rare genetic disorder occurring in approximately 1 in 7500 live births (Strømme, Bjømstad, & Ramstad, 2002).

The genetic bases of these conditions are quite different. Down syndrome is a classic chromosomal disorder caused by complete or partial triplication of chromosome 21, while Fragile X syndrome is a single-gene disorder caused by an expansion mutation of the trinucleotide (CGG) in the promoter region of the FMR1 gene, and Williams syndrome is a chromosomal microdeletion at 7q11.23 (Goldstein & Reynolds, 2010).

In terms of general cognitive functioning, individuals with Down syndrome, Fragile X syndrome (Abbeduto et al., 2001), and Williams syndrome show similar performance in standardized tests, ranging from mild to moderate intellectual impairment (Martens, Wilson, & Reutens, 2008). Age-associated decline is a particular characteristic of Down syndrome, which makes this genetic condition an optimum model for studying the genetic basis of Alzheimer-type dementia (Grieco, Pulsifer, Seligsohn, Skotko, & Schwartz, 2015; Potter, 2016).

The presence of intellectual disability in all these three genetic conditions invariably results in language disorder. In the face of intellectual disability, mixed receptive and expressive deficits in language are expected, although overall, it is seen that there is greater difficulty with expressive than with receptive language abilities (Rice, Warren, & Betz, 2005). Individuals with these three genetic disorders have problems in comprehension, which affect their capabilities to perceive and correct breakdowns in communication during spoken language production (Martin et al., 2017).

The expressive language abilities of individuals with Williams syndrome have been highlighted as fluent and well-articulated speech with an effective phonological working memory, which is responsible for facilitating the retention and manipulation of verbal information (Bellugi, Wang, & Jernigan, 1994; Vicari, Bellucci, & Carlesimo, 2006), and also, for favoring the syntactic domain of language (Jarrold, Baddeley, Hewes, & Phillips, 2001; Volterra, Caselli, Capirci, Tonucci, & Vicari, 2003); this is opposite to the trait seen in Down syndrome and fragile X syndrome.

Compared to individuals with Williams syndrome, morphosyntax and phonology are weak domains in individuals with Down syndrome and fragile X syndrome. However, this is a more prominent characteristic in Down syndrome compared to fragile X syndrome. As suggested by Abbeduto, Brady, and Kover (2007), the expressive language of individuals with fragile X syndrome shows more variation with regard to these language domains.

While children with Williams syndrome exhibit the most intelligible speech at the beginning of spoken language development using spoken language as the main communicative resource, children with Down syndrome show significant speech intelligibility problems, leading to prolonged use of gestures as a communicative resource. The use of representational gestures in children with Down syndrome and fragile X syndrome is controversial among researchers. Interventions promoting parental input into representational gestures have been suggested as an additional opportunity for language development in children with Down syndrome, and have been considered to have a predictive value with regard to vocabulary development Dimitrova, Özçalışkan, & Adamson, 2016; Lorang, Sterling, & Schroeder, 2018). On the other hand, the use of representational gestures in early developmental stages in children with Fragile X syndrome seems to be not effective due to the impact of autistic symptoms (Flenthrope & Brady, 2010; Rague, Caravella, Tonnsen, Klusek, & Roberts, 2018). The regulatory function of language seems to have greater positive predictive value in language development in Fragile X syndrome than gestures (Hahn, 2016).

One of the hypotheses explaining expressive language difficulties due to speech intelligibility problems in Down syndrome is weakness in the phonological working memory (Dodd & Thompson, 2001) and impairment of motor speech skills, with typical manifestations of childhood apraxia of speech (CAS), which include impairments in precision and consistency of speech movements (Kumin, 2006). The motor speech disorder associated with Down syndrome seems to be complex, involving single speech movements to sequential movements, and causes diadochokinetic deficits (Rupela, Velleman, & Andrianopoulos, 2016). Similarly, persistent speech production deficits and motor speech disorders are very common in children with Fragile X syndrome (Shriberg, Strand, & Mabie, 2016; Spinelli, Rocha, Giacheti, & Richieri-Costa, 1995; Zingerevich et al., 2009). Regarding Williams syndrome, these deficits seen in Down and Fragile X syndrome are not a main characteristic; however, Krishnan, Bergström, Alcock, Dick, and Karmiloff-Smith (2015) described a surprising oromotor praxis deficit in a study of individuals with Williams syndrome.

The other common evidence that supports spoken language deficits in Down syndrome is the higher prevalence of disfluencies in these patients (Eggers & Van Eerdenbrugh, 2018). In a comparative study conducted by our research team (Giacheti, Medina, & Rossi, 2010), individuals with Down syndrome showed a higher percentage of stuttering disfluencies (blocking, repetition of sound), whereas, individuals with Williams syndrome showed typical disfluencies related to linguistic formulation difficulties (hesitation and a silent pause between sentences), which suggests that this is an important aspect that differentiates the phenotypic expression related to fluency of speech between these two genetic syndromes. In fact, in a subsequent study we showed that the type of disfluencies presented in a language sample of individuals with Williams syndrome were important indicators of difficulties in elaboration and organization of the oral narrative, and therefore were possibly associated with language processing rather than speech processing (Rossi, Sampaio, Gonçalves, & Giacheti, 2011).

Regarding comorbidities and their implications in the language phenotype of genetic disorders, behavioral problems (hyperactivity and attention deficit symptoms, anxiety, and social interaction issues among others) and psychopathological manifestations (autistic symptoms) are commonly described in genetic syndromes. There is strong evidence that autism-like symptoms are associated with lower cognitive functioning in all three genetic disorders, namely, Down syndrome (Channell et al., 2015), Fragile X syndrome (McDuffie, Thurman, Hagerman, & Abbeduto, 2015), and Williams syndrome (Rossi & Giacheti, 2017; Royston et al., 2018).

Since individuals with Williams syndrome are hypersociable and not reserved with strangers (Gosch & Pankau, 1997; Mervis et al., 2000), this syndrome is considered an exciting model for social neuroscience, and is considered a unique phenotype that reflects the dimensions of prosocial behavior (Järvinen, Korenberg, & Bellugi, 2013). The social cognition phenotype of Williams syndrome has been described to be the opposite of that of autism (Couture et al., 2010) and to that of Fragile X syndrome. Fragile X syndrome is considered the most studied genetic model for ASD, as many of the typical autistic manifestations are part of the fragile X syndrome phenotype (Budimirovic, Protic, & Toma, 2017).

Repetitive and stereotyped behaviors are heterogeneous, and are usually described in many genetic disorders such as Fragile X syndrome, Prader-Willi syndrome, Smith-Magenis syndrome, and Algelman's syndrome (Riby & Hancock, 2008; Whitehouse & Lewis, 2015). The neurobiology of repetitive behavior is still under investigation, but there is strong evidence that it may result from corticostriatal circuit dysfunction (Langen, Durston, Kas, van Engeland, & Staal, 2011).

Repetitive and restrictive behaviors occur differently between and within genetic conditions; however, there is some level of deficit in social interaction and social communication in all these syndromes (Klein-Tasman, van der Fluit, & Mervis, 2018; Riby & Hancock, 2008). This is because many of these repetitive characteristics affect spoken language (e.g., echolalia, repetitive questions, conversational topic perseveration), which, in turn, has an impact on the functional use of language.

A set of studies conducted in our laboratory (Rossi, Moretti-Ferreira, & Giacheti; Rossi & Giacheti, 2017) are examples of how repetitive and restrictive behaviors occur in different ways within a genetic condition, and even more, how the simultaneous occurrence of these behaviors with other speech-language characteristics and behavioral problems might be important for describing sub-phenotypes in genetic disorders. However, we observed that both echolalia and perseverative speech were restrictive and repetitive behaviors that were observed in the speech of individuals with Williams syndrome; these behaviors had different impacts on communication performance. Echolalia is more restrictive than being just repetitive in a topic of conversation (perseverating on a topic) or using clichés.

Using clichés with exaggerated prosody and talkative behavior contributes to an apparent superior socio-communicative domain in Williams syndrome, contrary to echolalia. Furthermore, curiously, our study on individuals with Williams syndrome also showed opposite speech–language and behavioral traits, including monotone intonation and shyness, which represent the exact opposite of the Williams syndrome phenotype. Shy participants show lower speech rates in narration and have monotone intonation, which are similar to the fragile X syndrome phenotype.

Taking into account the overall advances with regard to our understanding of the three syndromes, Williams syndrome is without doubt the most fascinating genetic condition that has genuinely changed our understanding of language over the years. Though it is a rare disease, Williams syndrome has been investigated extensively as a neurobiological model of modularity with regard to the hypothesis of dissociation between verbal and nonverbal skills; this has been a milestone in relation to cognitive and language studies (Brock, 2007).

The hypothesis of superiority of auditory and verbal performance over visual-spatial deficits with relatively good language in spite of general cognitive deficits (Udwin & Yule, 1990) is currently discussed with caution.

For years, the language performance of individuals with Williams syndrome was supported by the static adult neuropsychological model that explained the language development in these individuals as parts intact and parts impaired (Karmiloff-Smith, 1997; Karmiloff-Smith, Brown, Grice, & Paterson, 2003). Certainly, the developmental perspective brought about a new approach with regard to language and cognition in Williams syndrome and suggested that it was a different developmental trajectory from that seen in typical development (Karmiloff-Smith et al., 2003; Laing et al., 2002; Mervis & Becerra, 2007). The intact language paradigm has been broken in view of the concept of different developmental trajectories with strengths and weaknesses in language abilities (Karmiloff-Smith et al., 2003). In these individuals, the interests in social-communication interactions and skills in using prosody to capture the attention of the audience (Gonçalves et al., 2010; Losh, Bellugi, & Reilly, 2000; Reilly, Losh, Bellugi, & Wulfeck, 2004) contrast with the pragmatic language impairment, inappropriate turn-taking and initiation of the turn, difficulties in maintaining the topic of conversation (Laws & Bishop, 2004; Philofsky, Fidler, & Hepburn, 2007; Rossi, Moretti-Ferreira, & Giacheti, 2007), and macrostructural deficits in narrative, with significant coherence deficits (Diez-Itza, Martínez, Pérez, & Fernández-Urquiza, 2018; Gonçalves et al., 2010; Reilly et al., 2004).

A cross-cultural study conducted in our laboratory with international collaboration investigated the oral narrative performance of individuals with Williams syndrome from Brazil, Portugal, and Spain. Narration was analyzed taking into account macrostructural elements (scenario, theme, plot, challenges, and resolution) and its organization. The results confirmed that these individuals have difficulty in organizing the global structure of a story, which leads to important pragmatic narrative deficits, regardless of their cultural background (Rossi, Garayzábal-Heinze, Sampaio, Gonçalves, & Giacheti, 2015). It appears that the major narrative problem in Williams syndrome occurs at the macrostructural level of narration (Diez-Itza et al., 2018; Gonçalves et al., 2010; Reilly et al., 2004; Rossi et al., 2015); this is in contrast to the impairment seen in Down syndrome and fragile X syndrome, which manifest mainly as significant impairments in phonology and in the syntactic structures of sentences (Finestack, Palmer, & Abbeduto, 2012).

However, our experience with Williams syndrome has shown that problems related to microstructures are also to be expected, since there are difficulties with the use of cohesive elements (Garayzábal Heinze, Prieto, Sampaio, & Gonçalves, 2007), and lowering of lexical diversity and sentence structure complexity in individuals with lower general cognitive functioning, and mainly with comorbidities such as autism-like symptoms.

Narrative intervention studies in genetic disorders are rare. A study conducted by Diez-Itza et al. (2018) in individuals with Williams syndrome reported both microstructural and macrostructural improvements. Therefore, considering that pragmatic narrative skills are weak overall in neurodevelopmental disorders, narrative assessment may be helpful in understanding the language phenotypes in genetic disorders, and in planning the necessary interventions.

The Role of Genetics in Spoken Language and its Disorders

Over the years, the critical neural elements and genes that are involved in spoken language have been described by studying some communication disorders such as DLDs, speech sound disorders, and fluency disorders.

DLD is a neurodevelopmental disorder that is not accompanied by intellectual disabilities, global developmental delays, hearing or other sensory impairment, motor dysfunction, or other mental disorders or medical conditions that can justify the child's language development as below that expected for the chronological age (Bishop & Hayiou-Thomas, 2008).

Children with DLD have significant difficulties in acquiring and developing language skills despite normal general cognitive abilities; these children have a gap between their verbal and nonverbal IQs (lower verbal IQ). Difficulties in lexical acquisition with speech delays are common manifestations reported by parents in the two first years of the children's lives (Bishop, 2006). These difficulties, including morphosyntactic deficits (which is considered a hallmark of the DLD phenotype), persist until the later years (Conti-Ramsden, Ullman, & Lum, 2015).

Theories of language organization and development have been proposed to explain the nature of the language problems in DLD. Among the different theoretical hypotheses to explain DLD, one is based on morphosyntactic deficits, with impairments in grammatical inflections and poor comprehension of sentences due to the complexity of the syntactic structure. Another perspective is based on weak phonological short-term memory with difficulty repeating nonwords (Leonard, 2014); this has been considered by some researchers as a strong diagnostic marker of this condition. Furthermore, performance with regard to nonword repetition is a useful measure of the heritable phenotype in DLD (Girbau, 2016; Newbury, Bishop, & Monaco, 2005).

The heterogeneity of the DLD phenotype might be represented by the multiple loci associated with this disorder (Bishop & Snowling, 2004). Currently, there are five loci associated with DLD documented in the OMIM database: 16q (OMIM # 606711, SLI1) (Johns Hopkins University, 2018d), 19q (OMIM # 606712, SLI2) (Johns Hopkins University, 2018e), 13q21 (OMIM # 607134, SLI3) (Johns Hopkins University, 2018f), 7q35-36 (OMIM # 612514, SLI4) (Johns Hopkins University, 2018g), and SLI5 (OMIM # 615432) (Johns Hopkins University, 2018h) caused by a mutation in the TM4SF20 gene on chromosome 2q36.

It is interesting to note that the forkhead box protein P2 (FOXP2) region is not listed among the DLD loci. The FOXP2 frenzy and media attention have overestimated FOXP2 as the "language gene." However, since the time when the FOXP2 gene was first described as the "language gene," there has been an increasing body of evidence arguing that this gene has low contribution to susceptibility to DLD (Newbury et al., 2002).

Fisher, Vargha-Khadem, Watkins, Monaco, and Pembrey (1998) described a three-generation pedigree with a severe speech and language disorder (the KE family) with an autosomal dominant monogenic trait and, indicated the 7q31 chromosome region to be responsible for the speech and language disorder. This was followed by a study by Lai, Fisher, Hurst, Vargha-Khadem, and Monaco (2001), which reported a point mutation in the FOXP2 gene in the KE family members. Many studies have been conducted since then to analyze and comprehend the role of FOXP2 in speech and language and its functions in the brain, since the primary neurobiological deficit was an open question.

The evolutionary history of the human FOXP2 gene indicates that it is biologically important. The FOXP2 gene located on chromosome 7q31 (OMIM # 605317) (Johns Hopkins

University, 2018i) is highly conserved in humans; the human gene differs at three amino-acid positions from its orthologue in the mouse and at two amino-acid positions in chimpanzees. Fixation of the allele in the human species is supposed to have occurred during the last 200,000 years, which gave humans a huge adaptive advantage (Enard et al., 2002). The forkhead box (Fox) family of transcription factors are genes that have implications in the developmental process, from organogenesis to speech acquisition (Hannenhalli & Kaestner, 2009).

In fact, mutation in the highly conserved genes has a huge impact on basic human functions, as they are involved in human developmental processes, such as spoken language. However, it would be naive to think that a complex symbolic system, such as the language system, would be controlled by only one gene. Moreover, there was a more important question related to the discovery of the FOXP2 gene mutation in the KE family: What was the nature of communication deficits in the KE family from the perspective of language development and diagnosis? Thus, this leads us to other important questions with regard to the debate on terminologies and taxonomies: What was, in fact, the KE family's speech-language diagnosis? Would it be a language disorder or a speech sound disorder?

Certainly, receptive language deficits in relation to the significant difficulties faced by KE family members to use syntactic rules must be the main reason that instigated the debate as to whether KE family members represented a language disorder or a speech sound disorder. It was not clear if the linguistic impairments in the KE family members were secondary manifestations of motor planning and sequencing deficits or if they arose simultaneously (Lai, Gerrelli, Monaco, Fisher, & Copp, 2003).

FOXP2 gene expression has been linked to neuronal circuits underlying the regulation of motor control and sensory-motor integration, which are considered critical to the development of the speech and language neural system, and therefore, FOXP2 is considered a central, but not unique, gene related to impairments in the movement sequence involved in producing speech and procedural learning (Lai et al., 2003).

Nowadays, there seems to be a consensus that the most overt feature of the KE family's phenotype is oromotor praxis deficit, which, in turn, is named "childhood apraxia of speech (CAS). CAS is a common term used by the American Speech-Language-Hearing Association (ASHA), which defines CAS as a speech sound disorder with deficits in the precision and consistency of speech movements in the absence of neuromuscular deficits. The main characteristics include errors in speech sound production and prosody due to impairment in planning and/or programming spatiotemporal parameters of movement sequences (ASHA, 2007).

Finally, taking into account DSM-5 (APA, 2013), the KE family diagnosis could be classified into the class of "speech sound disorder," which is defined as persistent difficulty in producing speech that interferes with speech intelligibility or prevents verbal communication of messages. The condition causes limitations in effective communication, which interferes with social participation, academic success, or professional performance, individually or in combination. The onset of symptoms occurs early in the developmental period and the speech difficulties are not attributable to congenital or acquired conditions, such as cerebral palsy, palatine cleft, deafness or hearing loss, traumatic brain injury, or any other cause. According to DSM-5 (APA, 2013), it is called "verbal dyspraxia" if motor coordination impairments are a particular characteristic.

The term "CAS" is also used as an alternative to "Speech-Language Disorder 1" in the OMIM database (OMIM # 602081, SPCH1) (Johns Hopkins University, 2018j), associated with FOXP2 disruption in the 7q31.1 chromosomal region. Further, the term "Speech Sound Disorder" in the OMIM database (Johns Hopkins University, 2018k) has the strongest linkage with the 3p12-q13 chromosomal region, which is also a locus linked to reading disability (dyslexia).

Quite apart from the research on the KE family and the FOXP2 gene, literature on CAS suggests concomitant language deficits as possible manifestations, which lead to difficulties in making an early diagnosis or including it in the differential diagnosis.

Differentiating CAS from other types of speech sound disorders or from DLD is challenging in children less than 3 years old. This is because children with severe speech sound disorders (with or without motor impairment) or with language disorders coexisting with articulation or phonology deficits frequently have a restricted expressive language or even more frequently, unintelligible speech, which most of the time is a limitation with regard to characterization, and consequently, identification of the nature of the deficit.

Frequently, the CAS phenotype with language impairments is characterized by superior performance in receptive language tasks and low performance in expressive language tasks, which is also a common pattern observed in early DLD (Lewis, Freebairn, Hansen, Iyengar, & Taylor, 2004). However, the receptive language scores of children with DLD are farthest from normative samples compared to scores of children with CAS. Moreover, speech characterized by inconsistency, slow speech rate, disturbed prosody, poorer performance during imitation (Dodd, 2014), and difficulties with diadochokinetic tasks in children with CAS (Fiori et al., 2016) are important parameters that can be help differentiate this condition from other speech sound disorders and DLD.

It is not surprising that there is evidence of linguistic and motor planning difficulties in CAS. There is a large overlapping network involving the temporal, frontal, and parietal areas that are involved in both language and praxis skills (Roby-Brami, Hermsdörfer, Roy, & Jacobs, 2012).

Speech sound disorder is a challenging diagnostic category due to the heterogeneous symptomatology (perceptual deficits, working memory impairments, speech motor control deficits) and the different subgroups of the condition are characterized by different manifestations (Dodd, 2014). According to Carrigg, Parry, Baker, Shriberg, and Ballard (2016), one of the major challenges with regard to this diagnostic category is that cognitive and other linguistic components are frequently neglected in speech assessment; therefore, coexisting language impairment may be underestimated, thereby reducing the precision of phenotype and sub-phenotype identification, as well as genetic and neural correlates.

The co-occurrence of speech and language impairment associated with FOXP2 disruption could be explained by the fact that FOXP2 is involved in the regulation of other genes, including the CNTNAP2 gene, which is located on chromosome 7.

Similar to the findings with FOXP2, variation within the CNTNAP2 gene was found among participants with CAS, but not with DLD (Centanni et al., 2015). However, its variants have also been associated with ASD (Rodenas-Cuadrado, Ho, & Vernes, 2014; Ross et al., 2017). Disruption of the CNTNAP2 gene is also associated with fluency disorder (stuttering), as described by our research team (Petrin et al., 2010). In that study, we described the case of a Brazilian boy with a complex neurodevelopmental phenotype with a 10 Mb deletion at 7q33-q35 involving several genes. The boy presented with a set of speech and language difficulties, including stuttering. Our study showed that one or more genes in the 7q33-q35 region may have a causal role in speech motor problems such as stuttering; specifically, partial deletion of CNTNAP2, including a deletion of the binding site for the transcription factor FOXP2, is involved. This case described by our team was a case of persistent stuttering, with poor prognosis even after speech-language intervention.

Stuttering is a childhood fluency disorder characterized by speech breakdown leading to an interruption in the flow of speaking. Typical stuttering manifestations include repetitions (sounds, syllables, words, and phrases), sound prolongations, blocks, interjections, and revisions. These breakdowns in speech also affect the rate and rhythm of speech. Physical tension, negative reactions, secondary behaviors, and avoidance of sounds, words, or speaking situations can co-occur with speech disfluencies (ASHA, 1993).

In DSM-5 (APA, 2013), stuttering is classified into the diagnostic class of neurodevelopmental disorders and the subgroup of communication disorder. The essential characteristics of developmental stuttering include a disturbance in normal fluency with an inappropriate temporal pattern of speech. This disturbance is characterized by frequent repetitions or prolongations of sounds or syllables, and other types of speech disfluencies, including interrupted words, audible or silent blocking, circumlocutions, words produced with excessive physical tension, and repetitions of monosyllabic words. Disturbance in fluency interferes with academic and professional success and social communication. The risk of stuttering among first-degree biological relatives of individuals with a fluency disorder is more than three times greater than the risk in the general population.

The OMIM database lists four loci for familial persistent stuttering: 15q21.2 (OMIM # 184450, STUT1) (Johns Hopkins University, 2018l) with a heterozygous mutation in the AP4E1 gene, 12q24.1 (OMIM # 609261, STUT2) (Johns Hopkins University, 2018m), 3q13.2-q13.33 (OMIM # 614655, STUT3) (Johns Hopkins University, 2018n), and 16q12.1-q23.1 (OMIM # 614668, STUT4) (Johns Hopkins University, 2018o).

This complex and multifaceted developmental fluency disorder can result from genetic and environmental factors, and there is an unfavorable effect on speech fluency if both factors act together (Nogueira, Oliveira, Giacheti, & Moretti-Ferreira, 2015). Regardless of whether there is a familial history of stuttering, factors such as masculine gender, persistent pattern of the fluency disorder, duration over 12 months, stutter disfluency, and negative familial attitudes occurring concomitantly increase the risk of stuttering (Oliveira, Domingues, Juste, Andrade, & Moretti-Ferreira, 2012). Familial stuttering has been classified as persistent and recovered and these subtypes have been put in perspective in genetic studies.

Understanding the developmental trajectory of stuttering and the course to persistence or recovery is necessary to take into account the interactions of stuttering symptoms with other developmental aspects such as temperament, language abilities, and speech motor skills (Smith & Weber, 2016). Beyond genetic influences, a growing body of research has suggested the interplay between cognitive function deficits (working memory), linguistic skills, and poor speech motor coordination as factors underlying persistent developmental stuttering (Yang, Jia, Fox, Siok, & Tan, 2019).

Similar to other communication disorders, the genetic basis of stuttering is heterogeneous and other chromosomal regions seem to be involved in this fluency disorder (2, 3p, 3q, 10, 14, and 16) (Frigerio-Domingues & Drayna, 2017). The exonic regions of FOXP2, CNTNAP2, GNPTAB, GNPTG, and NAGPA genes were sequenced in a study of individuals from North America and Brazil, including a group with familial developmental stuttering and a similar sized-group of neurologically normal controls, and the results showed that FOXP2 and CNTNAP2 genes were not involved in the genesis of familial persistent stuttering. However, mutations were found in the GNPTAB, GNPTG, and NAGPA genes, suggesting that the genetic basis of stuttering differs from that of CAS and DLD (Han et al., 2014). The role of GNPTAB, GNPTG, and NAGPA genes in stuttering have been demonstrated in others studies as well (Kazemi, Estiar, Fazilaty, & Sakhinia, 2018).

Conclusion

The scientific relevance of the language phenotype of neurodevelopmental disorders is supported by the fact that the professional has to deal with the probability that a genetic condition is presenting a certain phenotypic characteristic in the course of its developmental trajectory. It is relevant not only to explore the role of chromosomes and genes in the developmental domains but also to help the speech-language pathologist and other professionals formulate a well-defined intervention plan because of the clinical implications.

Thus, identifying the most prominent deficits in each condition (as exemplified by the descriptions of Down syndrome, fragile X syndrome, and Williams syndrome in this chapter), taking into account the subcomponents of language (phonology, syntax, morphology, semantics, and pragmatics) and its relationship with other specific developmental domains (motor, cognition, and behavioral) has enabled the description of a peculiar phenotype for each one, for example, the intelligible and well-articulated speech with rich prosody resources and clichés associated with a hypersociable personality in individuals with Williams syndrome, which contrasts with the unintelligible speech, agrammatic syntax, and stuttering disfluencies in individuals with Down syndrome and with the shy behavior, increased speech rate, and imprecise articulation of individuals with Fragile X syndrome.

The role of genes in the etiology of speech and language developmental disorders has not been completely determined. A deeper understanding of complex phenomena, such as the gene-brain-language relationship, has emerged from the study of different disciplines. In cases where DLDs and speech language disorders are the primary manifestations, in other words, the phenotype itself, the heterogeneity of manifestations is a huge challenge to molecular studies, in addition to the need for a large number of participants.

The emerging challenges facing the genetic basis of DLDs range from consensus regarding terminology and diagnostic criteria to the need to aggregate powerful technologies (molecular genetics and brain imaging) in order to map not only the susceptible genes, but also the neural circuits that might contribute to the phenotype of complex genetic disorders.

Finally, it is important to make it clear that when we highlight the genetic disorder and its effects on language, it does not mean that the sociocultural context and its influences are being ignored, as language development is a complex phenomenon. However, this chapter did not attempt to encompass the whole width of this issue, or explore the genetics of learning disabilities.

References

Abbeduto, L., Brady, N., & Kover, S. T. (2007). Language development and fragile X syndrome: Profiles, syndrome-specificity, and within-syndrome differences. *Mental Retardation And Developmental Disabilities Research Reviews, 13*(1), 36–46.

Abbeduto, L., Pavetto, M., Kesin, E., Weissman, M., Karadottir, S., O'Brien, A., & Cawthon, S. (2001). The linguistic and cognitive profile of Down syndrome: Evidence from a comparison with fragile X syndrome. *Down Syndrome Research and Practice, 7*(1), 9–15.

Adolphs, R. (2001). The neurobiology of social cognition. *Current Opinion in Neurobiology, 11*(2), 231–239.

American Psychiatric Association. (2013). Diagnostic and statistical manual of mental disorders (DSM-5®). American Psychiatric Pub.

American Speech-Language-Hearing Association. (1993). Definitions of communication disorders and variations.

American Speech-Language-Hearing Association. (2007). Childhood apraxia of speech.

Andersen, S. L. (2003). Trajectories of brain development: point of vulnerability or window of opportunity? *Neuroscience & Biobehavioral Reviews, 27*(1), 3–18.

Baty, B. F., Carey, J. C., & McMahon, W. M. (2010). Neurodevelopmental disorders and medical genetics: an overview. In S. Goldstein & C. R. Reynolds (Eds.), *Handbook of neurodevelopmental and genetic disorders in children* (2nd ed.). Guilford Press.

Bellugi, U., Wang, P. P., & Jernigan, T. L. (1994). Williams syndrome: An unusual neuropsychological profile. *Atypical Cognitive Deficits In Developmental Disorders: Implications For Brain Function, 23*, 23–56.

Bennett, R. L., French, K. S., Resta, R. G., & Doyle, D. L. (2008). Standardized human pedigree nomenclature: update and assessment of the recommendations of the National Society of Genetic Counselors. *Journal Of Genetic Counseling, 17*(5), 424–433.

Bishop, D., & Rutter, M. (2008). Neurodevelopmental disorders: Conceptual issues. In *Rutter's child and adolescent psychiatry* (pp. 32–41). Oxford: Blackwell.

Bishop, D. V. (2006). What causes specific language impairment in children? *Current Directions in Psychological Science, 15*(5), 217–221.

Bishop, D. V. (2010). Which neurodevelopmental disorders get researched and why? *PLoS One, 5*(11), e15112.

Bishop, D. V. (2014). Ten questions about terminology for children with unexplained language problems. *International Journal of Language & Communication Disorders, 49*(4), 381–415.

Bishop, D. V. (2017). Why is it so hard to reach agreement on terminology? The case of developmental language disorder (DLD). *International Journal of Language & Communication Disorders, 52*(6), 671–680.

Bishop, D. V., & Snowling, M. J. (2004). Developmental dyslexia and specific language impairment: Same or different? *Psychological Bulletin, 130*(6), 858.

Bishop, D. V., Snowling, M. J., Thompson, P. A., & Greenhalgh, T. (2016). CATALISE: A multinational and multidisciplinary Delphi consensus study. Identifying language impairments in children. *PLoS One, 11*(7), e0158753.

Bishop, D. V., Snowling, M. J., Thompson, P. A., Greenhalgh, T., Catalise-2 Consortium, Adams, C., ... & Boyle, C. (2017). Phase 2 of CATALISE: A multinational and multidisciplinary Delphi consensus study of problems with language development: Terminology. *Journal of Child Psychology and Psychiatry*, *58*(10), 1068-1080.

Bishop, D. V. M., & Hayiou-Thomas, M. E. (2008). Heritability of specific language impairment depends on diagnostic criteria. *Genes, Brain and Behavior, 7*(3), 365–372.

Brock, J. (2007). Language abilities in Williams syndrome: A critical review. *Development and Psychopathology, 19*(1), 97–127.

Budimirovic, D. B., Protic, D., & Toma, A. E. (2017). Fragile X syndrome: leading the way as the most common monogenic form of autism spectrum disorder and the most translated among neurodevelopmental disorders in clinical trials. *Journal of Clinical Genetics and Genomics*, (*1*), 1.

Camp, J. S., Karmiloff-Smith, A., Thomas, M. S., & Farran, E. K. (2016). Cross-syndrome comparison of real-world executive functioning and problem solving using a new problem-solving questionnaire. *Research in Developmental Disabilities, 59*, 80–92.

Carrigg, B., Parry, L., Baker, E., Shriberg, L. D., & Ballard, K. J. (2016). Cognitive, Linguistic, and Motor Abilities in a Multigenerational Family with Childhood Apraxia of Speech. *Archives of Clinical Neuropsychology, 31*(8), 1006–1025.

Centanni, T. M., Sanmann, J. N., Green, J. R., Iuzzini-Seigel, J., Bartlett, C., Sanger, W. G., & Hogan, T. P. (2015). The role of candidate-gene CNTNAP2 in childhood apraxia of speech and specific language impairment. *American Journal of Medical Genetics Part B: Neuropsychiatric Genetics, 168*(7), 536–543.

Channell, M. M., Phillips, B. A., Loveall, S. J., Conners, F. A., Bussanich, P. M., & Klinger, L. G. (2015). Patterns of autism spectrum symptomatology in individuals with Down syndrome without comorbid autism spectrum disorder. *Journal of Neurodevelopmental Disorders, 7*(1), 5.

Chater, N., & Christiansen, M. H. (2010). Language evolution as cultural evolution: how language is shaped by the brain. *Wiley Interdisciplinary Reviews: Cognitive Science, 1*(5), 623–628.

Christiansen, M. H., & Kirby, S. (2003). Language evolution: Consensus and controversies. *Trends in Cognitive Sciences, 7*(7), 300–307.

Conti-Ramsden, G., Ullman, M. T., & Lum, J. A. (2015). The relation between receptive grammar and procedural, declarative, and working memory in specific language impairment. *Frontiers in Psychology, 6*, 1090.

Couture, S. M., Penn, D. L., Losh, M., Adolphs, R., Hurley, R., & Piven, J. (2010). Comparison of social cognitive functioning in schizophrenia and high functioning autism: more convergence than divergence. *Psychological Medicine, 40*(4), 569–579.

de Sousa, A., & Cunha, E. (2012). Hominins and the emergence of the modern human brain. In *Progress in brain research* (Vol. 195, pp. 293–322). Amsterdam: Elsevier.

Deacon, T. W. (1998). *The symbolic species: The co-evolution of language and the brain*. New York: WW Norton & Company.

Decety, J., Bartal, I. B. A., Uzefovsky, F., & Knafo-Noam, A. (2016). Empathy as a driver of prosocial behaviour: Highly conserved neurobehavioural mechanisms across species. *Philosophical Transactions of the Royal Society B: Biological Sciences, 371*(1686), 20150077.

Diez-Itza, E., Martínez, V., Pérez, V., & Fernández-Urquiza, M. (2018). Explicit oral narrative intervention for students with Williams syndrome. *Frontiers in Psychology, 8*, 2337.

Dimitrova, N., Özçalışkan, Ş., & Adamson, L. B. (2016). Parents' translations of child gesture facilitate word learning in children with autism, Down syndrome and typical development. *Journal of Autism and Developmental Disorders, 46*(1), 221–231.

Dodd, B. (2014). Differential diagnosis of pediatric speech sound disorder. *Current Developmental Disorders Reports, 1*(3), 189–196.

Dodd, B., & Thompson, L. (2001). Speech disorder in children with Down's syndrome. *Journal of Intellectual Disability Research, 45*(4), 308–316.

Eggers, K., & Van Eerdenbrugh, S. (2018). Speech disfluencies in children with Down Syndrome. *Journal of Communication Disorders, 71*, 72–84.

Enard, W., Przeworski, M., Fisher, S. E., Lai, C. S., Wiebe, V., Kitano, T., … Pääbo, S. (2002). Molecular evolution of FOXP2, a gene involved in speech and language. *Nature, 418*(6900), 869.

Finestack, L. H., Palmer, M., & Abbeduto, L. (2012). Macrostructural narrative language of adolescents and young adults with Down syndrome or fragile X syndrome. *American Journal of Speech-Language Pathology, 21*(1), 29–46.

Fiori, S., Guzzetta, A., Mitra, J., Pannek, K., Pasquariello, R., Cipriani, P., … Chilosi, A. (2016). Neuroanatomical correlates of childhood apraxia of speech: A connectomic approach. *NeuroImage: Clinical, 12*, 894–901.

Fisher, S. E., Vargha-Khadem, F., Watkins, K. E., Monaco, A. P., & Pembrey, M. E. (1998). Localisation of a gene implicated in a severe speech and language disorder. *Nature Genetics, 18*(2), 168–170.

Flenthrope, J. L., & Brady, N. C. (2010). Relationships between early gestures and later language in children with fragile X syndrome. *American Journal of Speech-Language Pathology, 19*(2), 135–142.

Friederici, A. D. (2006). The neural basis of language development and its impairment. *Neuron, 52*(6), 941–952.

Friederici, A. D. (2017). Evolution of the neural language network. *Psychonomic Bulletin & Review, 24*, 41–47.

Friend, M., & Bates, R. P. (2014). The union of narrative and executive function: different but complementary. *Frontiers in Psychology, 5*, 469.

Frigerio-Domingues, C., & Drayna, D. (2017). Genetic contributions to stuttering: the current evidence. *Molecular Genetics & Genomic Medicine, 5*(2), 95–102.

Fuster, J. M. (2002). Frontal lobe and cognitive development. *Journal of Neurocytology, 31*(3-5), 373–385.

Garayzábal Heinze, E., Prieto, M. F., Sampaio, A., & Gonçalves, Ó. F. (2007). Valoración interlingüística de la producción verbal a partir de una tarea narrativa en el síndrome de Williams. *Psicothema, 19*(3), 428–434.

Giacheti, C.M., Medina, F.D.D., Rossi, N.F. (2010). Análise comparativa do perfil da fluência da fala de indivíduos com síndrome de Down e com a síndrome de Williams-Beuren. Anais 17 Congresso Brasileiro de Fonoaudiologia, Salvador-Ba. Disponível em http://www.sbfa.org.br/portal/anais2009/resumos/R1531-1.pdf

Giacheti, C. M., & Rossi, N. F. R. (2012). Chapter 4: Síndromes genéticas. In A. S. Lopes-Herrera & L. P. Maximino (Eds.), *Fonoaudiologia: Intervenções e Alterações da Linguagem Oral Infantil* (pp. 61–76). Ribeirão Preto: Novo Conceito.

Girbau, D. (2016). The Non-word Repetition Task as a clinical marker of Specific Language Impairment in Spanish-speaking children. *First Language, 36*(1), 30–49.

Goldstein, S., & Reynolds, C. R. (2010). *Handbook of neurodevelopmental and genetic disorders in children* (2nd ed.). New York: Guilford Press.

Goldstein, T. R., & Winner, E. (2012). Enhancing empathy and theory of mind. *Journal of Cognition and Development, 13*(1), 19–37.

Gonçalves, Ó. F., Pinheiro, A. P., Sampaio, A., Sousa, N., Férnandez, M., & Henriques, M. (2010). The narrative profile in Williams Syndrome: There is more to storytelling than just telling a story. *The British Journal of Development Disabilities, 56*(111), 89–109.

Gosch, A., & Pankau, R. (1997). Personality characteristics and behaviour problems in individuals of different ages with Williams syndrome. *Developmental Medicine & Child Neurology, 39*(8), 527–533.

Grandjean, P., & Landrigan, P. J. (2014). Neurobehavioural effects of developmental toxicity. *The Lancet Neurology, 13*(3), 330–338.

Grieco, J., Pulsifer, M., Seligsohn, K., Skotko, B., & Schwartz, A. (2015). Down syndrome: Cognitive and behavioral functioning across the lifespan. *American Journal of Medical Genetics Part C: Seminars in Medical Genetics, 169*(2), 135–149.

Hahn, L. J. (2016). Joint attention and early social developmental cascades in neurogenetic disorders. In *International review of research in developmental disabilities* (Vol. 51, pp. 123–152). Cambridge, MA: Academic Press.

Han, T. U., Park, J., Domingues, C. F., Moretti-Ferreira, D., Paris, E., Sainz, E., … Drayna, D. (2014). A study of the role of the FOXP2 and CNTNAP2 genes in persistent developmental stuttering. *Neurobiology of Disease, 69*, 23–31.

Hannenhalli, S., & Kaestner, K. H. (2009). The evolution of Fox genes and their role in development and disease. *Nature Reviews Genetics, 10*(4), 233.

Hauser, M. D., Yang, C., Berwick, R. C., Tattersall, I., Ryan, M. J., Watumull, J., … Lewontin, R. C. (2014). The mystery of language evolution. *Frontiers in Psychology, 5*, 401.

Hoff, E. (2013). Language development. *Cengage Learning*.

Holland, S. K., Vannest, J., Mecoli, M., Jacola, L. M., Tillema, J. M., Karunanayaka, P. R., … Byars, A. W. (2007). Functional MRI of language lateralization during development in children. *International Journal of Audiology, 46*(9), 533–551.

Jarrold, C., Baddeley, A. D., Hewes, A. K., & Phillips, C. (2001). A longitudinal assessment of diverging verbal and non-verbal abilities in the Williams syndrome phenotype. *Cortex, 37*(3), 423–431.

Järvinen, A., Korenberg, J. R., & Bellugi, U. (2013). The social phenotype of Williams syndrome. *Current Opinion in Neurobiology, 23*(3), 414–422.

Johns Hopkins University. (2018a) # 190685 Down syndrome. In Johns Hopkins University. OMIM: online mendelian inheritance in man 1966–2019. Baltimore: Johns Hopkins University. Retrieved from: https://www.omim.org/entry/190685.

Johns Hopkins University. (2018b). # 194050 Williams syndrome. In Johns Hopkins University. OMIM: online mendelian inheritance in man 1966–2019. Baltimore: Johns Hopkins University. Retrieved from: https://www.omim.org/entry/194050.

Johns Hopkins University. (2018c). # 300624 Fragile X syndrome. In Johns Hopkins University. OMIM: online mendelian inheritance in man 1966–2019. Baltimore: Johns Hopkins University. Retrieved from: https://www.omim.org/entry/300624.

Johns Hopkins University. (2018d). # 606711 Specific language impairment 1. In Johns Hopkins University. OMIM: online mendelian inheritance in man 1966–2019. Baltimore: Johns Hopkins University. Retrieved from: https://www.omim.org/entry/606711.

Johns Hopkins University. (2018e). # 606712 Specific language impairment 2. In Johns Hopkins University. OMIM: online mendelian inheritance in man 1966–2019. Baltimore: Johns Hopkins University. Retrieved from: https://www.omim.org/entry/606712.

Johns Hopkins University. (2018f). # 607134 Specific language impairment 3. In Johns Hopkins University. OMIM: online mendelian inheritance in man 1966–2019. Baltimore: Johns Hopkins University. Retrieved from: https://www.omim.org/entry/607134.

Johns Hopkins University. (2018g). # 612514 Specific language impairment 4. In Johns Hopkins University.

OMIM: online mendelian inheritance in man 1966–2019. Baltimore: Johns Hopkins University. Retrieved from: https://www.omim.org/entry/612514.

Johns Hopkins University. (2018h). # 615432 Specific language impairment 5. In Johns Hopkins University. OMIM: online mendelian inheritance in man 1966–2019. Baltimore: Johns Hopkins University. Retrieved from: https://www.omim.org/entry/615432.

Johns Hopkins University. (2018i). # 605317 Forkhead Fox P2. In Johns Hopkins University. OMIM: online mendelian inheritance in man 1966–2019. Baltimore: Johns Hopkins University. Retrieved from: https://www.omim.org/entry/605317.

Johns Hopkins University. (2018j). # 602081 Speech-Language disorder 1. In Johns Hopkins University. OMIM: online mendelian inheritance in man 1966–2019. Baltimore: Johns Hopkins University. Retrieved from: https://www.omim.org/entry/602081.

Johns Hopkins University. (2018k). # 608445 Speech-Sound disorder. In Johns Hopkins University. OMIM: online mendelian inheritance in man 1966–2019. Baltimore: Johns Hopkins University. Retrieved from: https://www.omim.org/entry/608445.

Johns Hopkins University. (2018l). # 184450 Stuttering familial persistent 1. In Johns Hopkins University. OMIM: online mendelian inheritance in man 1966–2019. Baltimore: Johns Hopkins University. Retrieved from: https://www.omim.org/entry/184450.

Johns Hopkins University. (2018m). # 609261 Stuttering familial persistent 2. In Johns Hopkins University. OMIM: online mendelian inheritance in man 1966–2019. Baltimore: Johns Hopkins University. Retrieved from: https://www.omim.org/entry/609261.

Johns Hopkins University. (2018n). # 614655 Stuttering familial persistent 3. In Johns Hopkins University. OMIM: online mendelian inheritance in man 1966–2019. Baltimore: Johns Hopkins University. Retrieved from: https://www.omim.org/entry/614655.

Johns Hopkins University. (2018o). # 614668 Stuttering familial persistent 4. In Johns Hopkins University. OMIM: online mendelian inheritance in man 1966–2019. Baltimore: Johns Hopkins University. Retrieved from: https://www.omim.org/entry/614668.

Järvinen-Pasley, A., Bellugi, U., Reilly, J., Debra, L., Galaburda, A., Reiss, A. L., & Korenberg, J. R. (2008). Defining the social phenotype in Williams syndrome: a model for linking gene, the brain, and behavior. *Development and Psychopathology, 20*(1), 1–35.

Karmiloff-Smith, A. (1992). *Beyond modularity: a developmental approach to cognitive science*. Cambridge: MIT Press.

Karmiloff-Smith, A. (1997). Crucial differences between developmental cognitive neuroscience and adult neuropsychology. *Developmental Neuropsychology, 13*(4), 513–524.

Karmiloff-Smith, A. (2018). Development itself is the key to understanding developmental disorders. In *Thinking developmentally from constructivism to neuroconstructivism* (pp. 97–117). Abingdon: Routledge.

Karmiloff-Smith, A., Brown, J. H., Grice, S., & Paterson, S. (2003). Dethroning the myth: Cognitive dissociations and innate modularity in Williams syndrome. *Developmental Neuropsychology, 23*(1-2), 227–242.

Kazemi, N., Estiar, M. A., Fazilaty, H., & Sakhinia, E. (2018). Variants in GNPTAB, GNPTG and NAGPA genes are associated with stutterers. *Gene, 647*, 93–100.

Klein-Tasman, B. P., van der Fluit, F., & Mervis, C. B. (2018). Autism Spectrum Symptomatology in Children with Williams Syndrome Who Have Phrase Speech or Fluent Language. *Journal of Autism and Developmental Disorders*, 1–14.

Knowland, V. C., & Thomas, M. S. (2011). Developmental trajectories in genetic disorders. In *International review of research in developmental disabilities* (Vol. 40, pp. 43–73). Cambridge, MA: Academic Press.

Krishnan, S., Bergström, L., Alcock, K. J., Dick, F., & Karmiloff-Smith, A. (2015). Williams syndrome: A surprising deficit in oromotor praxis in a population with proficient language production. *Neuropsychologia, 67*, 82–90.

Kumin, L. (2006). Speech intelligibility and childhood verbal apraxia in children with Down syndrome. *Down Syndrome Research and Practice, 10*(1), 10–22.

Lai, C. S., Fisher, S. E., Hurst, J. A., Vargha-Khadem, F., & Monaco, A. P. (2001). A forkhead-domain gene is mutated in a severe speech and language disorder. *Nature, 413*(6855), 519.

Lai, C. S., Gerrelli, D., Monaco, A. P., Fisher, S. E., & Copp, A. J. (2003). FOXP2 expression during brain development coincides with adult sites of pathology in a severe speech and language disorder. *Brain, 126*(11), 2455–2462.

Laing, E., Butterworth, G., Ansari, D., Gsödl, M., Longhi, E., Panagiotaki, G., … Karmiloff-Smith, A. (2002). Atypical development of language and social communication in toddlers with Williams syndrome. *Developmental Science, 5*(2), 233–246.

Langen, M., Durston, S., Kas, M. J., van Engeland, H., & Staal, W. G. (2011). The neurobiology of repetitive behavior:… and men. *Neuroscience & Biobehavioral Reviews, 35*(3), 356–365.

Laws, G., & Bishop, D. V. (2004). Pragmatic language impairment and social deficits in Williams syndrome: A comparison with Down's syndrome and specific language impairment. *International Journal of Language & Communication Disorders, 39*(1), 45–64.

Leonard, L. B. (2014). *Children with specific language impairment*. Cambridge, MA: MIT Press.

Lewis, B. A., Freebairn, L. A., Hansen, A. J., Iyengar, S. K., & Taylor, H. G. (2004). School-age follow-up of children with childhood apraxia of speech. *Language, Speech, and Hearing Services in Schools, 35*(2), 122–140.

Lorang, E., Sterling, A., & Schroeder, B. (2018). Maternal responsiveness to gestures in children with Down syndrome. *American Journal of Speech-Language Pathology, 27*(3), 1018–1029.

Losh, M., Bellugi, U., & Reilly, J. (2000). Narrative as a social engagement tool: The excessive use of evaluation in narratives from children with Williams syndrome. *Narrative Inquiry, 10*(2), 265–290.

Luyster, R. J., Seery, A., Talbott, M. R., & Tager-Flusberg, H. (2011). Identifying Early-Risk Markers and Developmental Trajectories for Language Impairment in Neurodevelopmental Disorders. *Developmental Disabilities Research Reviews, 17*(2), 151–159.

Martens, M. A., Wilson, S. J., & Reutens, D. C. (2008). Research review: Williams syndrome: a critical review of the cognitive, behavioral, and neuroanatomical phenotype. *Journal of Child Psychology and Psychiatry, 49*(6), 576–608.

Martin, G. E., Barstein, J., Hornickel, J., Matherly, S., Durante, G., & Losh, M. (2017). Signaling of noncomprehension in communication breakdowns in fragile X syndrome, Down syndrome, and autism spectrum disorder. *Journal of Communication Disorders, 65*, 22–34.

McDuffie, A., Thurman, A. J., Hagerman, R. J., & Abbeduto, L. (2015). Symptoms of autism in males with fragile X syndrome: A comparison to nonsyndromic ASD using current ADI-R scores. *Journal of Autism and Developmental Disorders, 45*(7), 1925–1937.

McKusick, V. A. (2007). Mendelian Inheritance in Man and its online version, OMIM. *The American Journal of Human Genetics, 80*(4), 588–604.

Mervis, C. B., & Becerra, A. M. (2007). Language and communicative development in Williams syndrome. *Mental Retardation and Developmental Disabilities Research Reviews, 13*(1), 3–15.

Mervis, C. B., & Robinson, B. F. (2005). Designing measures for profiling and genotype/phenotype studies of individuals with genetic syndromes or developmental language disorders. *Applied Psycholinguistics, 26*(1), 41–64.

Mervis, C. B., Robinson, B. F., Bertrand, J., Morris, C. A., Klein-Tasman, B. P., & Armstrong, S. C. (2000). The Williams syndrome cognitive profile. *Brain and Cognition, 44*(3), 604–628.

Miller, B. L., & Cummings, J. L. (Eds.). (2017). *The human frontal lobes: Functions and disorders*. London: Guilford Publications.

Newbury, D. F., Bishop, D. V., & Monaco, A. P. (2005). Genetic influences on language impairment and phonological short-term memory. *Trends in Cognitive Sciences, 9*(11), 528–534.

Newbury, D. F., Bonora, E., Lamb, J. A., Fisher, S. E., Lai, C. S., Baird, G., … Bolton, P. F. (2002). FOXP2 is not a major susceptibility gene for autism or specific language impairment. *The American Journal of Human Genetics, 70*(5), 1318–1327.

Nogueira, P. R., Oliveira, C. M. C. D., Giacheti, C. M., & Moretti-Ferreira, D. (2015). Familial persistent developmental stuttering: disfluencies and prevalence. *Revista CEFAC, 17*(5), 1441–1448.

Oliveira, B. V. D., Domingues, C. E. F., Juste, F. S., Andrade, C. R. F. D., & Moretti-Ferreira, D. (2012). Gagueira desenvolvimental persistente familial: perspectivas genéticas. *Revista da Sociedade Brasileira de Fonoaudiologia, 17*(4), 489–494.

Online Mendelian Inheritance in Man, OMIM®. (2018). McKusick-Nathans Institute of Genetic Medicine, Johns Hopkins University, Baltimore, MD, (July, 19, 2018). Retrieved from https://omim.org/

Petrin, A. L., Giacheti, C. M., Maximino, L. P., Abramides, D. V., Zanchetta, S., Rossi, N. F., … Murray, J. C. (2010). Identification of a microdeletion at the 7q33-q35 disrupting the CNTNAP2 gene in a Brazilian stuttering case. *American Journal of Medical Genetics Part A, 152*(12), 3164–3172.

Philofsky, A., Fidler, D. J., & Hepburn, S. (2007). Pragmatic language profiles of school-age children with autism spectrum disorders and Williams syndrome. *American Journal of Speech-Language Pathology, 16*(4), 368–380.

Poeppel, D. (2014). The neuroanatomic and neurophysiological infrastructure for speech and language. *Current opinion in Neurobiology, 28*, 142–149.

Potter, H. (2016). Beyond trisomy 21: phenotypic variability in people with Down syndrome explained by further chromosome mis-segregation and mosaic aneuploidy. *Journal of Down Syndrome & Chromosome Abnormalities, 2*(1), 109.

Rague, L., Caravella, K., Tonnsen, B., Klusek, J., & Roberts, J. (2018). Early gesture use in fragile X syndrome. *Journal of Intellectual Disability Research, 62*(7), 625–636.

Reilly, J., Losh, M., Bellugi, U., & Wulfeck, B. (2004). "Frog, where are you?" Narratives in children with specific language impairment, early focal brain injury, and Williams syndrome. *Brain and Language, 88*(2), 229–247.

Reilly, S., Bishop, D. V., & Tomblin, B. (2014). Terminological debate over language impairment in children: Forward movement and sticking points. *International Journal of Language & Communication Disorders, 49*(4), 452–462.

Riby, D. M., & Hancock, P. J. (2008). Viewing it differently: Social scene perception in Williams syndrome and autism. *Neuropsychologia, 46*(11), 2855–2860.

Rice, D., & Barone, S., Jr. (2000). Critical periods of vulnerability for the developing nervous system: evidence from humans and animal models. *Environmental Health Perspectives, 108*(Suppl 3), 511.

Rice, M. L., Warren, S. F., & Betz, S. K. (2005). Language symptoms of developmental language disorders: An overview of autism, Down syndrome, fragile X, specific language impairment, and Williams syndrome. *Applied Psycholinguistics, 26*(1), 7–27.

Roby-Brami, A., Hermsdörfer, J., Roy, A. C., & Jacobs, S. (2012). A neuropsychological perspective on the link between language and praxis in modern humans. *Philosophical Transactions of the Royal Society B: Biological Sciences, 367*(1585), 144–160.

Rodenas-Cuadrado, P., Ho, J., & Vernes, S. C. (2014). Shining a light on CNTNAP2: Complex functions to complex disorders. *European Journal of Human Genetics, 22*(2), 171.

Rodenhiser, D., & Mann, M. (2006). Epigenetics and human disease: Translating basic biology into clinical applications. *Canadian Medical Association Journal, 174*(3), 341–348.

Ross, L. A., Del Bene, V. A., Molholm, S., Woo, Y. J., Andrade, G. N., Abrahams, B. S., & Foxe, J. J. (2017). Common variation in the autism risk gene CNTNAP2, brain structural connectivity and multisensory speech integration. *Brain and Language, 174*, 50–60.

Rossi, N. F. (2016) Uso das técnicas de ressonância magnética funcional e eletroencefalografia nos estudos sobre o desenvolvimento da linguagem. *Avaliação da fala e da linguagem*, 109.

Rossi, N. F., Garayzábal-Heinze, E., Sampaio, A., Gonçalves, Ó. F., & Giacheti, C. M. (2015). Narrativa oral na síndrome de Williams e no desenvolvimento típico: estudo transcultural Brasil, Portugal e Espanha. Anais XXIII Congresso Brasileiro de Fonoaudiologia e IX Internacional de Fonoaudiologia, 14 a 16 de Outubro de 2015.

Rossi, N. F., & Giacheti, C. M. (2017). Association between speech–language, general cognitive functioning and behaviour problems in individuals with Williams syndrome. *Journal of Intellectual Disability Research, 61*(7), 707–718.

Rossi, N. F., Moretti-Ferreira, D., & Giacheti, C. M. (2007). Perfil comunicativo de indivíduos com a síndrome de Williams-Beuren. *Revista da Sociedade Brasileira de Fonoaudiologia*, 1–9.

Rossi, N. F., Sampaio, A., Gonçalves, Ó. F., & Giacheti, C. M. (2011). Analysis of speech fluency in Williams syndrome. *Research in Developmental Disabilities, 32*(6), 2957–2962.

Royston, R., Oliver, C., Moss, J., Adams, D., Berg, K., Burbidge, C., … Waite, J. (2018). Brief report: Repetitive behaviour profiles in williams syndrome: Cross syndrome comparisons with Prader–Willi and Down syndromes. *Journal of Autism and Developmental Disorders, 48*(1), 326–331.

Rupela, V., Velleman, S. L., & Andrianopoulos, M. V. (2016). Motor speech skills in children with Down syndrome: A descriptive study. *International Journal of Speech-Language Pathology, 18*(5), 483–492.

Shriberg, L. D., Strand, E. A., & Mabie, H. L. (2016). Prevalence estimates for three types of motor speech disorders in complex neurodevelopmental disorders (CND). In *Eighteenth Biennial Conference on Motor Speech: Motor Speech Disorders & Speech Motor Control, Newport Beach, CA*.

Skeide, M. A., & Friederici, A. D. (2016). The ontogeny of the cortical language network. *Nature Reviews Neuroscience, 17*(5), 323.

Smith, A., & Weber, C. (2016). Childhood Stuttering–Where are we and Where are we going? *Seminars in Speech and Language, 37*(4), 291.

Spinelli, M., Rocha, A. C. D. O., Giacheti, C. M., & Richieri-Costa, A. (1995). Word-finding difficulties, verbal paraphasias, and verbal dyspraxia in ten individuals with fragile x syndrome. *American Journal of Medical Genetics, 60*(1), 39–43.

Strømme, P., Bjømstad, P. G., & Ramstad, K. (2002). Prevalence estimation of Williams syndrome. *Journal of Child Neurology, 17*(4), 269–271.

Tomasello, M. (2010). *Origins of human communication*. Cambridge, MA: MIT Press.

Udwin, O., & Yule, W. (1990). Expressive language of children with Williams syndrome. *American Journal of Medical Genetics, 37*(S6), 108–114.

Verendeev, A., & Sherwood, C. C. (2017). Human brain evolution. *Current Opinion in Behavioral Sciences, 16*, 41–45.

Vicari, S., Bellucci, S., & Carlesimo, G. A. (2006). Evidence from two genetic syndromes for the independence of spatial and visual working memory. *Developmental Medicine and Child Neurology, 48*(2), 126–131.

Volterra, V., Caselli, M. C., Capirci, O., Tonucci, F., & Vicari, S. (2003). Early linguistic abilities of Italian children with Williams syndrome. *Developmental Neuropsychology, 23*(1-2), 33–58.

Whitehouse, C. M., & Lewis, M. H. (2015). Repetitive behavior in neurodevelopmental disorders: Clinical and translational findings. *The Behavior Analyst, 38*(2), 163–178.

Yang, Y., Jia, F., Fox, P. T., Siok, W. T., & Tan, L. H. (2019). Abnormal neural response to phonological working memory demands in persistent developmental stuttering. *Human Brain Mapping, 40*(1), 214–225.

Yeger, H. (2015). The evolution of human communication. *Journal of cell communication and signaling, 9*(3), 289–290.

Yeh, Z. T., Tsai, M. C., Tsai, M. D., Lo, C. Y., & Wang, K. C. (2017). The relationship between theory of mind and the executive functions: Evidence from patients with frontal lobe damage. *Applied Neuropsychology: Adult, 24*(4), 342–349.

Zhang, Y. E., Landback, P., Vibranovski, M. D., & Long, M. (2011). Accelerated recruitment of new brain development genes into the human genome. *PLoS Biology, 9*(10), e1001179.

Zingerevich, C., Greiss-Hess, L., Lemons-Chitwood, K., Harris, S. W., Hessl, D., Cook, K., & Hagerman, R. J. (2009). Motor abilities of children diagnosed with fragile X syndrome with and without autism. *Journal of Intellectual Disability Research, 53*(1), 11–18.

19

Social and Psychological Stressors

Elizabeth Halstead, Justin Stanley,
and Joanna Greer

Children with IDD are at greater risk for psychopathology than their typically developing peers (de Ruiter, Dekker, Verhulst, & Koot, 2007). Developmental factors and their interaction with an individual's social environment offer a framework for intervention (Kiddle & Dagnan, 2011). In this chapter, stressors are discussed as risk factors for development and exacerbation of emotional and behavioral problems in individuals with IDD. The literature has shown the negative impact of environmental stress factors on individuals with IDD. However, the broader literature, not specific to individuals with IDD, shows although children experience adversity and stress, their mental health and well-being can still be positive (e.g., Alvord & Grados, 2005).

Why some individuals respond better than others to stress has been debated, but one theory is that this variability can be conceptualized as resilience. Resilience is generally seen as a positive construct that can be built upon and improved and encompasses many personal attributes and environmental factors. Rutter has defined resilience as "an interactive concept that is with the combination of serious risk experiences and a relatively positive psychological outcome despite those experiences" (Rutter, 2006, p. 2). Garmezy, Masten, and Tellegen (1984) presented a three-model approach to stress, compensatory, challenge, and protective factor models. Protective factors are defined as having a positive influence on behavior and moderate the relationship between risk and problem behaviors (Jessor, Van Den Bos, Vanderryn, Costa, & Turbin, 1995). Garmezy (1985) organized the protective factors into personality factors, family cohesion, and external support systems.

Resilience is being explored in individuals with IDD and their families, and attributes and skills related to resilience have begun to be identified. A study by Miller (2002) used open-ended questions to determine resilience in students with IDD. Resilience was described as a person experiencing achievement despite others having a low expectation of them. The students identified several attributes which they believed made them resilient, such as identifiable success experiences (having a job), areas of strength (e.g., being good at art/sport), having self-determination, having special friendships and encouraging teachers, and acknowledging the learning disability.

E. Halstead (✉)
Department of Psychology and Human Development,
Institute of Education, University Central London,
London, UK
e-mail: l.halstead@ucl.ac.uk

J. Stanley
Department of Psychology, Rutgers University,
New Jersey, USA

J. Greer
Department of Psychology, Northumbria University,
Newcastle upon Tyne, UK

J. L. Matson (ed.), *Handbook of Intellectual Disabilities*, Autism and Child Psychopathology
Series, https://doi.org/10.1007/978-3-030-20843-1_19

Similarly, Raskind, Goldberg, Higgins, and Herman (1999) found six attributes in individuals with IDD who had been successful: self-awareness, proactivity, perseverance, emotional stability, appropriate goal setting, and the presence and use of effective support systems. Conversely, a lack of self-awareness, reactivity, lack of perseverance, emotional instability, lack of appropriate goal setting and self-directedness, and lack of presence or use of effective support systems led to more negative outcomes. Gilmore, Campbell, Shochet, and Roberts (2013) compared resilience in children with IDD to their typically developing peers (TD), and found children with IDD reported lower levels of tolerance, fewer future goals and higher levels of emotional sensitivity compared to TD peers, though other factors like optimism and self-esteem were reported at a similar level to TD peers.

In addition to building resilience, reviewing potential environmental risk factors and how they could be modified may lead to better outcomes in individuals with IDD (Fletcher, Loschen, Stavrakaki, & First, 2007; Hawkins, Catalano, & Miller, 1992). Dykens (2000) emphasized the need for researchers to go beyond measurement and prevalence studies of psychopathology in children with IDD, and identify the reasons why these children are prone to psychopathology to begin with. More recent research has turned to intervention on this basis of identified risk factors (Fisher, Moskowitz, & Hodapp, 2013; Jacola, Hickey, Howe, Esbensen, & Shear, 2014). Therefore, each section of this chapter considers not only how to reduce risk factors that impact on mental health and well-being, but also how to increase protective factors.

Rutter (1987) proposed resistance to stress is relative; a risk factor in one situation can be a protective factor in another (e.g., social support). Of particular importance is the family environment, reflecting elements such as parental stress, bidirectional relationships between emotional expressions of distress and child behavior problems, and the role of interventions targeting protective factors such as family cohesion and social support. Implications for intervention will be discussed.

Stress and Coping

Perceptions of stress predict current and future symptoms of psychopathology among adults with IDD (Hartley & MacLean Jr, 2005; Hastings, Hatton, Taylor, & Maddison, 2004). Lazarus (1991) describes “stress as harms, threats, and challenges,” the reaction to which depends on “what a person has at stake with an encounter with the environment or in life in general” (Lazarus, 1991, p.1 35) Individuals feel more stress when they perceive a threat to self-esteem or are not confident in their coping abilities (Roe & Gray, 1991).

The literature suggests perceptions of stress may be higher in individuals with IDD. Bramston, Fogarty, and Cummins (1999) found when administering the Lifestress Inventory to individuals with IDD that they reported stressors less than their nondisabled peers. It was found that the IDD group tended to assign higher impact values to the stressors they did report experiencing. In addition, Hatton and Emerson found that individuals with IDD experience stress at a similar, if not greater level to the general population (Hatton & Emerson, 2004). Perceptions of stress may vary based on subtypes of IDD; adults with ASD experience higher levels of perceived stress than their peers with other forms of IDD (Gillott & Standen, 2007).

Coping is related to depression in adults with mild IDD (Hartley & MacLean Jr, 2005). Lunsky (2003) found depressed females with mild IDD used fewer coping strategies than their nondepressed peers. Hartley and MacLean Jr (2005) found that symptoms of depression were related to infrequent use of active coping and later found that a group of IDD participants with depression reported a higher frequency of stressful social interactions than the nondepressed group. Given our knowledge of high prevalence rates of depression in the IDD population, as discussed in Chap. 48, coping mechanisms are likely to be impaired in a considerable proportion of this population. Coping mechanisms are discussed further in Chap. 38.

However, in the context of protective factors buffering risk, coping could be a protective factor in the IDD population as it is a skill that can be developed and positive coping mechanisms have been demonstrated to protect against negative outcomes in the general literature and with parents of children with IDD (Hastings & Brown, 2002; Heiman, 2002; Rutter, 1987; Werner, 2000). Rutter suggested the concept of resilience "focuses attention on coping mechanisms, mental sets, and the operation of personal agency. In other words, it requires a move from a focus on external risks to a focus on how these external risks are dealt with by the individual" (Rutter, 2006, p. 8). Few studies have focused on coping strategies used by individuals with IDD; however, Hartley and McLean Jr (2009) found that use of active coping vs. avoidant coping accounted for a significant portion of the variance in psychological distress experienced by individuals with IDD in stressful social situations. Janssen, Schuengel, and Stolk (2002) found in addition to developing low self-esteem and a general feeling of incompetence, individuals with IDD are at risk of developing learned helplessness, a process they compare to development of learned helplessness in people affected by the demoralizing conditions of chronic poverty (Olson & Schober, 1993). These factors could lead to ineffective coping mechanisms.

A final consideration is that stress measured subjectively can be difficult to assess. Subjective stress is often measured by self-report questionnaires, and as much as reliability can be a matter of concern for the general population (Turner & Wheaton, 1995), this concern may be even greater among the IDD population (Heal & Sigelman, 1995). Bramston, Bostock, and Tehan (1993) acknowledged many stressors individuals with mild ID experience may not be covered in scales like the Lifestress Inventory (Fogarty, Bramston, & Cummins, 1997). However, the Lifestress Inventory has been shown to be internally reliable, and the informant version a suitable parallel instrument (Lunsky & Bramston, 2006). Typically, past researchers have used a combination of qualitative and quantitative methodologies to measure stress, more recently, the Bangor Life Events Schedule for Intellectual Disabilities (BLESID-SR: Hulbert-Williams, Hastings, Crowe, & Pemberton, 2011), has been designed as a measure of significant life events occurring in the life of someone with IDD. In the BLESID-SR an interviewer uses a semi-structured interview to guide the conversation and reflect in the scoring what the individual has said about their life events. The BLESID-SR is available in both a self-report and proxy version. Research exploring caregiver reports of stress and self-reports of stress has suggested that it is important to assess stress from two perspectives when possible (Cummins, 2005).

Life Events

The Impact of Life Events

The connection between life events and mental health problems has been found in the general population (Tennant, 2002). Similar patterns are found in adults with IDD, as life events experienced by this population are associated with emotional disorders, depression, and behavior problems (Esbensen & Benson, 2006; Hastings et al., 2004). Cooper, Smiley, Morrison, Williamson, and Allan (2007) assessed mental health in 1023 adults with intellectual disabilities and found as a significant factor the number of life events experienced in the prior 12-month period. Hulbert-Williams and Hastings (2008) conducted a narrative review of research on exposure to adverse life events as a risk factor for psychological problems and found association of life events with clinically significant depression. Research links experience of life events to behavior difficulties in adults with IDD (Esbensen & Benson, 2006; Hulbert-Williams et al., 2011; Owen et al., 2004). Stress is often associated with negative outcomes in individuals with IDD, including symptoms of mental illness in adults with mild IDD and depression in adults with moderate IDD (Hastings et al., 2004; Lunsky, 2003).

Unfortunately, a gap in the literature concerns the relationship between pre-existing psychological problems in adults with IDD and the susceptibility of this population to trauma and other psychological problems (Hulbert-Williams & Hastings, 2008). Research is needed to investigate further temporal associations between psychological problems and life events (Hulbert-Williams & Hastings, 2008; Kraemer et al., 1997). One study provided tentative evidence that life events may be a risk factor for psychological problems in adults with IDD using a longitudinal approach (Esbensen & Benson, 2006). Additional research is needed into the mechanisms by which experience of adverse events becomes a risk factor for psychological problems, and whether this is mediated by protective factors such as social support.

Major Life Events

The link between major life events (e.g., loss of a spouse, loss of a job, major illness or injury, moving) and the experience of stress and related physical and mental conditions is established in the general population (Harkness & Monroe, 2016). Research has suggested individuals with IDD are more susceptible to psychological distress, and the development of comorbid psychiatric conditions or behavioral problems, after experiencing major life events (Magiati, Tay, & Howlin, 2014). The pattern of increased susceptibility of individuals with IDD to life events begins in childhood. Children with IDD experience a wider range of life events and are reported to experience such events more frequently than children without IDD (Hatton & Emerson, 2004). The discrepancy in risk exposure to adverse life events between individuals with IDD and TD peers continues into adulthood, and may be explained by aspects such as increased dependency on paid and family caregivers, increased negative social interactions, and a relatively restricted social circle compared to nondisabled peers in the same age range (Bramston et al., 1999; Fogarty et al., 1997; Levitas & Gilson, 2001). The greater dependency of adults with IDD on family and paid caregivers is a source of additional risk for major life events, as the loss of a friend or family member may also equate to the loss of a home or a carer (Hollins & Esterhuyzen, 1997; Levitas & Gilson, 2001). Gilrane-McGarry and Taggart (2007) suggest lack of support during bereavement is a major factor in individuals with IDD susceptibility to a complicated grief process, where the failure of grief to resolve over a long time period results in the development of psychopathology including chronic depression.

In retrospective studies, significant major life events and their relationship to mental health crises in those with IDD has been well documented (Ghaziuddin, 1988; Hastings et al., 2004; Hatton & Emerson, 2004). Although the literature shows associations between life event frequency and mental health in individuals with IDD, there is still variability in the psychopathology of individuals with IDD when experiencing life events. One explanation for this may be variation among subgroups. Maag and Reid (2006) found moderators such as gender, age, and ethnicity seem to cause significant variability in depression as an outcome. Taylor, Hatton, Dixon, and Douglas (2004) found significant differences in the rates for particular disorders across gender, age, and residence type. Studies suggest the prevalence of mental disorders may peak at the moderate level of IDD, at least for depression, anxiety, and psychosis (Holden & Gitlesen, 2009; Myrbakk & von Tetzchner, 2008). Psychopathology models in IDD should consider the level of IDD and demographics in the variability of results. There is a need for more differentiation in research regarding gender and severity of disability (Wong, 2003).

Cumulative Life Events

The extent to which a person with IDD is exposed to negative life events is an important factor in development of related mental health problems. Cumulative exposure to life events on the mental health of people with IDD has been studied, with multiple exposure found to correlate with development of affective disorders such as depression (Tsakanikos et al., 2006). Accumulated exposure to life events, and risk factors that are related to cumulative exposure such as poverty and greater presence of physical disability, is linked to poor physical and mental health in this population (Brooks, 2006; Masten, 2001).

What makes a person vulnerable to poor mental health outcomes after a life event will differ based on persons and circumstances (Spini, Hanappi, Bernardi, Oris, & Bickel, 2013). Abravanel and Sinha (2015) conducted a population-based study of 745 adults exposed to cumulative life events, distinguishing between those "at risk" based on pre-morbid mood disorder, and those "never depressed." They found that emotion regulation, assessed using the Difficulties with Emotional Regulation Scale (Gratz & Roemer, 2004), accounted for 50% of the variance in depressive symptomology independent of risk status. Emotion regulation is a complex process drawing on use of cognitive, experiential, and emotion-based strategies to affect the way adverse life events are interpreted across the life course (Garnefski, Kraaij, & Spinhoven, 2001). Relative impoverishment of effective emotion regulation strategies in individuals with IDD compared to their TD peers may begin in childhood and continue throughout the life course (McClure, Hapern, Wolper, & Donahue, 2009) indicating this is a prominent risk factor and suitable for targeting through early intervention. Janssen et al. (2002) argue ineffective attachment models developed by children with IDD are a major risk factor for experience of chronic stress, depression, and problem behaviors during social interactions, persistent through adulthood. This provides one example of how a specific failure in emotion regulation strategies can serve as a risk factor for developing psychopathology during the experience of life adverse events.

Trauma

The development of post-traumatic stress disorder from exposure to life events has been studied in individuals with IDD (Murphy, O'Callaghan, & Clare, 2007) though research is constrained by a lack of ways to measure trauma in this cohort (Wigham, Taylor, & Hatton, 2014). Other than a handful of studies that are largely record reviews concerning sexual and physical assault (see Bowman, Scotti, & Morris, 2010), a dearth of research exists on the types of trauma occurring in the lives of individuals with IDD. Martorell and Tsakanikos (2008) emphasize separating between "non-traumatic" life events and "traumatic" life events may be difficult for this population, as difficulty understanding a stressful situation can lead the person with IDD to experience greater levels of distress than nondisabled peers.

Research suggests a disproportionate number of children with IDD are victims of abuse, with more severely disabled children at greatest risk (Fisher, Hodapp, & Dykens, 2008). There is a tendency for individuals with IDD who have been abused to receive psychiatric diagnosis, such as schizophrenia and depression (e.g., Ryan, 1994). Therefore, it is unclear whether the apparently strong association between trauma and psychiatric disabilities is due to increased risk of abuse in individuals with psychiatric disorders, thus the possibility of a shared etiology between the two.

As in the general population, one of the most prominent consequences of violent victimization in persons with IDD is PTSD. The limited data suggests that, as expected, individuals with disabilities are at increased risk for developing PTSD. For example, Ryan (1994) performed evaluations on 310 consecutive admissions to a team consultation service for persons with IDD and found that 16.5% met criteria for current PTSD. This rate is approximately double the rate of lifetime PTSD in the general population (e.g., Kessler, Sonnega, Bromet, Hughes, & Nelson, 1995).

For example, post-traumatic stress reactions, up to and including the onset of post-traumatic stress disorder, are highly prevalent in adults with IDD when exposed to environmental stressors as well as to abuse, neglect, and violence (Wigham & Emerson, 2015). Adults with IDD experience more traumatic life events compared to the TD population, and have fewer psychological and social resources for dealing with these stressors (Magiati et al., 2014). Adults with IDD frequently "suffer in silence" the higher rate of traumatic life events that they are prone to experience, and have less control over their environment so as to allow them to avoid (whether through internal or external means) reminders of past trauma (Wigham & Emerson, 2015). Staff training on the affective experience of adults with IDD, promoting the idea that they are aware of, and suffer from, life stressors, are needed to address this problem. However, a finding posited by Wigham et al. (2014) is the lack of moderation between social support and significant behavioral disturbances after traumatic life events, which may contribute to the onset of PTSD after experience of trauma. The difficulty people with IDD have in keeping social support may make a difference (Gilrane-McGarry & Taggart, 2007).

Protective factors that mitigate exposure to traumatic life events and adverse psychological outcomes include intelligence, perceived self-efficacy, and optimism, as studied in both disabled and nondisabled children exposed to trauma (Masten, Narayan, Silverman, & Osofsky, 2015) and intellectually disabled children in this situation (Finzi-Dottan & Karu, 2006). Social support is shown in many studies to be protective against adverse effects of trauma (Ozbay et al., 2007) though a distinction has been made between types of support which are more protective, with higher-quality relationships (i.e., allowing for more emotional expression and shared problem-solving) having greater protective qualities (Platt, Keyes, & Koenen, 2014).

Transition

Resilience has been posed as being not a fixed attribute, but as changing with circumstances (Rutter, 1987; Werner, 1989). There are certain points in an individual's life when they may be more vulnerable to stress and this may contribute to changeability in coping and perceptions of stress in individuals with IDD. The transition to adulthood, during which young adults with IDD leave behind the relative structure of the school environment to pursue uncertain employment and residential outcomes, is a period in which many individuals with IDD report being vulnerable to feelings of stress and anxiety (Williams & Heslop, 2005). This is in keeping with Raskind et al. (1999) who theorized the change over time from childhood to adolescence may be due to participants having more power in decision-making concerning their own education at the time.

The transition period is often perceived as a stressful time for young adults with IDD and their families. Many individuals with IDD face poorer outcomes than their TD peers across measures of transition success, such as social relationships, supported and competitive employment, and the ability to live independently (Foley, Dyke, Girdler, Bourke, & Leonard, 2012). Increased rates of psychological distress experienced during transition increases the risk for problem behaviors (Williams & Heslop, 2006). A qualitative interview study of young people with IDD found many young adults with IDD reported subjective feelings of being pressured by parents and teachers to make particular decisions related to their schooling and post-secondary community participation choices, as opposed to feeling free to make their own decisions (Williams & Heslop, 2005). The feeling that one has lost control over decision-making is exacerbated by loss of social networks that occurs when young adults with IDD transition from school environments (Raghavan & Pawson, 2008).

A second stressor faced by young adults with IDD and their families during transition is the unclear pathway of post-secondary supports. Transition-related changes in social roles and lack of supports for families can negatively impact family well-being (Neece, Kraemer, & Blacher, 2009). This combined with loss of social networks can increase problem behaviors during transition (Williams & Heslop, 2006). Williams and Heslop (2006) recommend approaching problem behaviors during transition from the stance of strengthening the individual's social networks. Forte, Jahoda and Dagnan (2011) used a mixed methodology approach to explore resilience in young people with IDD to examine the worries experienced by young people aged 17–20 with IDD during transition into adulthood. Their worries differed significantly in type as well as intensity. Young people with IDD were most worried about making friends, being bullied, and losing people they were dependent upon like parents, vs. TD peers who were most worried about getting a job, not having enough money, and having to make decisions about future choices. The IDD group also reported significantly greater levels of rumination over these worries. Forte, Jahoda, and Dagnan (2011) suggest obtaining insight into these patterns can strength a resilience-based intervention. For example, psychological interventions that target rumination could mitigate development of depression during this period.

Social Environment

Social Competence

The ability to navigate social interactions and benefit from positive aspects of social communication varies in the IDD population. Social competence has been defined as an individual's ability to successfully adapt to his/her social environment (Vaughn & Haager, 1994). Schafer and Semrud-Clikeman (2008) review difficulties that children with IDD experience with social competence.

Individuals with certain IDD diagnoses, such as Down syndrome (DS) and Williams syndrome (WS), are often characterized by having proficient social skills, including empathy and social communication, and in turn enables positive responses to others in distress (Di Nuovo & Buono, 2011). Conversely, individuals with IDD with an autism spectrum disorder (ASD) diagnosis demonstrate more social difficulties than individuals without comorbid ASD. Kau et al. (2004) found greater impact of social communication difficulties on problem behavior, in the case of individuals with Fragile X syndrome (FXS) with comorbid ASD as opposed to individuals with FXS without comorbid ASD. Individuals with Down syndrome (DS) who meet the screening criteria for comorbid ASD report milder social difficulties than children with ASD without any diagnosis of DS (Howlin, Wing & Gould, 1995). Diagnosis type and associated differences in social competence may create differential outcomes when attempting to integrate children with IDD into mainstream classroom settings. For example, integrated with TD peers in mainstream classrooms, children with ASD may be at high risk for social isolation (Locke, Ishijima, Kasari, & London, 2010).

Social difficulties encountered by this population extend beyond difficulties from the increased behavior disorders; also implicated are deficiencies in peer-related social competence. Guralnick (2006) estimates 60–65% of children with IDD fail to use appropriate social strategies during social tasks. Social isolation may be found in children besides the 25% of those with IDD who have behavior problems.

Larkin, Jahoda, MacMahon, and Pert (2012) found correlations between hostile attributional style, and increased experience of social conflict, in young adults with IDD. Increased likeliness of perceiving threat in ambiguous interactions, and a lack of coping strategies for use when faced with social threats, may be responsible for the higher rates of aggressive and affective disorders often observed in adults with IDD (Emerson & Jahoda, 2013).

As social interactions have long been identified as a potential stress factor for individuals with IDD, there is evidence of several interventions aimed at improving social skills for children and adults with IDD as is discussed further in Chap. 38. Szumski, Smogorzewska, Grygiel, and Orlando (2017) identified a significant improvement in the social skills of pre-school children with ASD using a "Play Time / Social Time" intervention designed to develop social skills compared to a problem-solving intervention. A meta-analysis of 55 social intervention skills studies with individuals with ASD (Bellini, Peters, Benner, & Hopf, 2007) found only minimal effect of these interventions; however, they did highlight that the setting of the interventions, such as resource rooms, were frequently inappropriate. In contrast, greater efficacy was observed when interventions took place in classroom / playtime settings. A meta-analysis of 19 studies (Gates, Kang, & Lerner, 2017) found that group social skills interventions led to moderate overall improvements in social competence. The authors also identified a large self-report effect, which they attributed to increased social knowledge, but not perceived changes in the individuals' own social behavior. These effects were found in studies including data from parents, children, observers, and tasks. The same trend was not observed in studies based on teachers' observations. Gates et al. (2017) emphasized that differences in efficacy rates are likely to reflect the variations by reporting sources.

Peer Interactions

Adults with mild IDD report more frequent and severe stress from negative social interactions than from other categories of stress events, and have more difficulty adaptively coping with everyday stressful social situations than other stressful events (e.g., Bramston et al., 1999; Hartley & MacLean Jr, 2005).

Children with IDD experience greater social isolation than TD peers and find it harder to develop positive peer relationships (Guralnick, 2006). Behavior problems are a prominent reason why children with IDD face social isolation (Crnic, Hoffman, Gaze, & Edelbrock, 2004; Guralnick, 2006). Children with IDD whose disabilities heighten the risk of aggression (such as autism spectrum disorder) are more likely to experience social isolation than children with IDD with lower levels of aggression, like Williams syndrome (Odom et al., 2006). Children with IDD with behavior problems and poor social skills are at greater risk of being bullied (Reiter & Lapidot-Lefler, 2007) and engage in bullying themselves (Christensen, Fraynt, Neece, & Baker, 2012).

The social isolation experienced by individuals with IDD often persists into adulthood (Lippold & Burns, 2009). Scott and Havercamp (2014) found a significant number of adults with IDD reported receiving less emotional support from others than nondisabled peers. Adults with IDD are more likely to receive social support from paid caregivers (Amado, 1993). These relationships are not reciprocal and are less likely to buffer against loneliness than other sources of support.

Social stressors such as negative social interactions are linked to physical and mental health problems in adults with IDD, including depression. Nezu, Nezu, Rothenberg, DelliCarpini, and Groag (1995) found associations between symptoms of depression and reported negative social interactions in a study of 107 adults with mild IDD. Hartley and MacLean Jr (2005) found that stress from negative social interactions predicted symptoms of depression in adults with IDD. Lunsky and colleagues also found a correlation between stress from negative social interactions and depressive symptoms; furthermore, continuing stress predicted the persistence of depressive symptoms 6 months later (Lunsky & Benson, 2001; Lunsky & Havercamp, 1999). Bender, Rosenkrans, and Crane (1999) found nonverbal individuals with IDD face a greater risk of suicide and posited that one reason for this may be social isolation.

There is a high prevalence of loneliness reported by adults with IDD. Stancliffe, Wilson, Bigby, Balandin, and Craig (2014) estimate up to 50% of adults with IDD are chronically lonely. Research by Cacioppo and colleagues (Cacioppo & Cacioppo, 2014) suggests loneliness contributes to poorer health through myriad pathways. Other risks for loneliness throughout the lifespan are family ecological variables like childhood attachment styles (Janssen et al., 2002). Adults with IDD, who lack coping resources or social supports to promote resilience during traumatic life events, are at greater risk for suicide (Gilmore & Cuskelly, 2014).

Social Support

It has been established that adults with mild to moderate ID are more vulnerable to stressful social interactions than nondisabled peers, and they report social stressors to have greater negative impact than other life events (Bramston et al., 1999; Fogarty et al., 1997; Hartley & MacLean Jr, 2005). Social support and positive peer interactions can be a protective factor for individuals with IDD as they promote resilience in the face of adverse life events (Bonanno, 2004) and moderate against development of PTSD (Brewin, Andrews, & Valentine, 2000).

A model linking social support to stress-buffering is provided by Cohen and Wills (1985). The buffering effects of social support persist across the lifespan. Rose and Espelage (2012) identify the ways that supportive peer relationships protect children with IDD from experiencing victimization during childhood. However, social support may not always moderate the relationship of trauma and behavioral changes (Wigham et al., 2014). It is likely that the stress-buffering effects of social support is affected by social-cognitive aspects such as whether one perceives such support as positive or negative (Lunsky, 2008).

Interventions to improve social interactions within persons with intellectual disabilities may be most effective in childhood, when social competencies are still developing (Guralnick, 2006). Carter and Hughes (2005) reviewed 26 interventions aimed at improving social interactions between students with IDD and nondisabled peers, and concluded additional research is needed to probe how interventions may have an impact on some but not all aspects of social interaction, such as improving social competence, reducing aggression, or facilitating environmental supports. In a similar light, Amado (1993) considers how to create a conceptual framework for studying interventions for adults with IDD, including bolstering social competencies.

Social Support and the Family

One stress-buffering resource is the availability and/or quality of social support in parents of children with IDD (Ekas, Lickenbrock, & Whitman, 2010; White & Hastings, 2004). Targeting social support can improve aspects of parental well-being including reduced mental health problems (Lovell, Moss, & Wetherell, 2012), reduced spousal problems (White & Hastings, 2004) and greater self-efficacy and coping (Kersh, Hedvat, Hauser-Cram, & Warfield, 2006).

The role of informal support in the reduction of stress in parents of individuals with IDD is widely documented (Robinson, Weiss, Lunsky, & Ouellette-Kuntz, 2016; White & Hastings, 2004). Mothers' ratings of grandparent support in the care of their child are negatively correlated with stress (Hastings, Allen, McDermott, & Still, 2002). Parents' perceptions of the helpfulness of informal sources of support, rather than the number of supports, has the most consistent associations with parental well-being (Zaidman-Zait et al., 2017). There are also changes in the type of support most beneficial with age. Practical support is more associated than emotional support with greater parental well-being in parents of adolescents with ASD (White & Hastings, 2004). By the time a child reaches adolescence, parents might have come to terms with being a parent of a child with an IDD, and need less emotional support.

The role of formal support sources in parental well-being seems to be less clear. Support services for families of children with disabilities more commonly focus on the child's needs (White & Hastings, 2004); therefore parental support needs are often unmet (Freedman & Boyer, 2000). Furthermore, several studies revealed unhelpful relationships and disagreements with professional services, for example, an inability to provide an individualized service adding to stress (Sloper & Turner, 1992).

Family Environment

Parenting Stress

High parenting stress is an important environmental risk variable. It is associated with many undesirable outcomes, including parent depression (Hastings, Daley, Burns, & Beck, 2006), marital conflict (Kersh et al., 2006), poor physical health (Eisenhower, Baker, & Blacher, 2009), less effective parenting (Crnic, Gaze, & Hoffman, 2005), and increased child behavior problems (Baker et al., 2003). Parents of children with IDD typically report more stress than parents of TD children (Baker et al., 2003; Emerson, 2003).

Parenting a child with an IDD is frequently a lifelong responsibility. Robinson et al. (2016) found nearly 50% of family members of individuals with IDD reported more caregiving responsibilities than they could handle, notably if there are more children living at home. Increased burden on parents of individuals with IDD are linked to the severity of symptoms and level of disability (Irazábal et al., 2012). High levels of behavioral support needs are linked to increased parental burden (Al-Krenawi, Graham, & Al Gharaibeh, 2011). This is also evident when the individual with IDD has health complications (Valicenti-McDermott et al., 2015) or a mental health problem (Irazábal et al., 2012). These profiles are reported consistently cross-culturally irrespective of majority / minority ethnicity or the country's level of income (Blacher & McIntyre, 2006; but also see Valicenti-McDermott et al., 2015).

There is a wealth of literature reporting greater deficits in psychosocial functioning and a greater prevalence of psychological distress in parents of children with IDD (Blacher & Hatton, 2007; Hastings, 2016; Singer, 2006). These include psychological distress, anxiety, depression, demoralization, and loss of freedom due to caregiving duties (Robinson et al., 2016). However, a population-based sample found 40% of mothers of children with IDD reported clinically significant emotional problems, whereas 60% did not (Totsika, Hastings, Emerson, Lancaster, & Berridge, 2011). Though the prevalence of parental depression is greater than the general population, evidence suggests most parents of children with IDD do not suffer from clinically significant depression.

Although there is evidence that stress experienced by parents of children with IDD can be chronic, there is marked individual variation in its trajectory over the life course (Glidden & Schoolcraft, 2003). Parents' psychological health and children's developmental health is especially vulnerable during the early years of a child's schooling (Fontil & Petrakos, 2015). Parental concerns include acceptance of their child by new teachers and peers and whether schools can meet the child's learning needs (Hirst, Jervis, Visagie, Sojo, Cavanagh, 2011). There is also evidence of declines in parental psychological well-being with increasing age of the child (Orsmond, Seltzer, Greenberg, & Krauss, 2006), though some research reports decreases in parental stress in adolescence (Woodman, 2014). Young adulthood may be especially challenging due to impending transition from school to work, and cause concerns about the child's future (Lueckling & Fabian, 1997; but see Goff et al., 2016). Social support may reduce during this time (Shattuck, Wagner, Narendorf, Sterzing, & Hensley, 2011).

Another concern is the continued co-residence of the child in the family home, often well past the age when most TD children become independent. Seltzer, Floyd, Song, Greenberg, and Hong (2011) reported a higher rate of co-residence of adults with IDD when their parents were in midlife and in early old age than in a comparison group. An adult child living at home is generally associated with poorer well-being for parents in midlife and early old age, more so in mothers (Pudrovska, 2009). A longitudinal study of 117 families before and after residential relocation (Seltzer, Krauss, Hong, & Orsmond, 2001) found levels of direct caregiving decreased and parents had less frequent contact with residential staff during the first 3 years after the adult child moved from home, resulting positively on parental well-being.

Sociodemographic Variables

Higher education level (Al-Krenawi et al., 2011), higher income (Santiago, Wadsworth, & Stump, 2011), and more effective coping style (Mak & Ho, 2007; Stuart & McGrew, 2009) are factors associated with less parental burden. Families supporting a child with IDD are more likely to be exposed to adverse socioeconomic circumstances (Emerson, 2007; Emerson & Hatton, 2009). They have lower rates of employment, return to employment later after childbirth, and are less likely to work full-time (Parish, Seltzer, Greenberg, & Floyd, 2004), and thus have less financial security later in life.

The relationship of family circumstances to psychopathology in children with IDD appears to be bidirectional. It has been found children with IDD often reside in single-parent, lower-socioeconomic families (Emerson, 2007). Emerson and Hatton (2007) found significant associations between gender, age and eight social variables and the risk of having behavioral disorders as well as hyperkinesis. Emerson and Hatton (2009) found that socioeconomic position moderated associations between child behavior problems and maternal anxiety, depression and self-assessed health.

However, some research suggests that raising a child with IDD contributes to parental stress and depression regardless of the buffering effects of socioeconomic status. Hoare, Harris, Jackson, and Kerley (1998) did not find parental depression to be related to either marital or socioeconomic status (also see Olsson & Hwang, 2001). In contrast, low levels of socioeconomic support along with parents' negative perceptions of their ability to influence their children's behavior are associated with increased stress and depression (Falk, Norris, & Quinn, 2014). Zaidman-Zait, Most, Tarrasch, and Haddad (2018) found during transition, more disadvantaged parents experienced more stress and depression, and their children had lower adaptive behavior and more behavior problems than comparison groups. However, when controlling for between-group differences in family socioeconomic circumstances, poorer well-being among parents of children with disabilities may be attenuated or even eliminated (Emerson, Baines, Allerton, & Welch, 2010; Olsson & Hwang, 2008). As such, socioeconomic status may not serve as a buffer against depression in parents of children with IDD (but also see Stoneman, 2007).

Minority status (Luthar, 1991; Masten, 2011) and racial discrimination (Brooks, 2006) also predict poor outcomes for children and youth. Children of African and Hispanic descent are often disadvantaged due to living in severely distressed neighborhoods with reduced accessibility to social supports, community services, employment opportunities, and high-quality schools. Age of the individual with IDD is important when considering life course vulnerabilities. Raskind et al. (1999) found a reduction in the perceived intensity of stressors from childhood to adolescence, particularly perception of having an IDD. Older women with IDD remain the least studied members of the disability population, and yet they often live well into late adulthood. LeRoy, Walsh, Kulik, and Rooney (2004) found older women with IDD to have limited resources, and social networks; yet they reported that their health was good, though it often limited their activities. Despite their societal limitations, these women reported this is the happiest period of their lives.

Residential placement can have a significant impact on stress and health in adults with IDD. Taggart (2014) found adults with IDD who live in supported residential placements, especially in institution-like settings, are at greater risk of psychopathology than adults who live with family members. Hastings et al. (2004) found people with IDD living in staffed accommodation have significantly more life events. Owen et al. (2004) found participants were exposed to at least three adverse life events the past year, and life event exposure was linked with increased aggressive behavior and greater incidence of emotional disorders.

Adults with IDD often become emotionally as well as practically dependent on the paid support staff assisting them with community access and daily living activities (Lunsky, 2008). When support staff leave due to job changes, or when adults with IDD leave behind support persons with whom they had positive experiences, feelings of bereavement can result (Hartley & MacLean Jr, 2005). Accumulation of multiple stressors combined with an impersonal staff attitude has been found to perpetuate challenging behaviors in people with IDD (Griffith, Hutchinson, & Hastings, 2013).

Family Environment

Along with the established relationship between socioeconomic position within a family, and the health of the child with IDD, there are a number of factors associated with nonoptimal child development (Guralnick, 2005). It is well established that parents of children with IDD show increased stress compared to parents of children without IDD (Emerson, 2003; Hastings & Beck, 2004). Experiencing child behavioral problems with a child with IDD has been shown to be a high risk for poorer health and well-being in cross-sectional research (Baker et al. 2002; Totsika et al., 2013).

However, there has been variability shown in parental well-being and other outcomes as seen similarly in the psychopathology of individuals with IDD. Totsika et al. (2011) found 60% of mothers of children with IDD did not report concerning clinical levels of their emotional problems. Positive outcomes are also established in the literature (e.g., Hastings & Taunt, 2002; Scorgie & Sobsey, 2000). Researchers have explored moderating or mediating factors in the relationship between child behavioral problems and maternal outcomes such as Plant and Sanders (2007) who explored behavioral problems and parental stress, with social support as a moderator. They found social support had a positive effect on this relationship. In addition, Halstead, Ekas, Hastings, and Griffith (2018) found perceived social support functioned as a protective factor, when perceived social support was higher, social support moderated the relationship between child behavioral and emotional problems and maternal outcomes, such as depression and life satisfaction. Social support has been found to be a protective factor between life events and parenting stress and outcomes such as depression, and social isolation (Dunn, Burbine, Bowers, & Tantledd-Dunn, 2001).

Family climate has an impact on child behavioral and emotional problems, including cohesiveness, expressiveness and conflict (Greenberg et al., 2012). Adverse characteristics associated with poorer child mental health include low parental intellectual level (Feldman, 1997), mental illness, particularly depression (Seifer & Dickstein, 2000), and parental neglect or abuse (Barnett, 1997). Guralnick (1998, 2005) suggests family climate risk factors are mediated by nonoptimal patterns of family interaction, such as high levels of expressed emotion and family conflict. Many family interaction patterns are affected by family risk factors, and contribute to poorer outcomes for the child with IDD (e.g., Phelps, Belsky, & Crnic, 1998). Individual risk factors might not exert an adverse impact on child outcomes in the absence of nonoptimal patterns of family interaction, suggesting optimal patterns can protect against the effect of other risk factors (Werner, 1995).

Family Cohesion

Family cohesion relates to positive outcomes for parents and children. Maintaining cohesion may be challenging for families of individuals with IDD (Mitchell, Szczerepa, & Hauser-Cram, 2016), and parents may be at greater risk of relationship breakdown. Research has found somewhat higher divorce rates in parents of individuals with IDD compared with parents of TD children (Hartley et al., 2010). Certain groups may be more susceptible to marital breakdown, such as parents of individuals with ASD (Hartley et al., 2010). Greater depressive symptoms have been found in single mothers of children with IDD (Olsson & Hwang, 2001).

Preventing domestic discord may promote parental mental health in families with children with IDD, as a positive marital relationship can predict lower parenting stress (Benson & Kersh, 2011). The well-being of one parent affects the stress experienced by the other parent (Hastings et al., 2005). This is important for parents of individuals with IDD as they may differ in their perceptions of family life. Studies have found mothers to be more stressed than fathers in having a child with an IDD (Dabrowska & Pisula, 2010; Oelofsen & Richardson, 2006), though this is not a consistent finding (Hastings, 2003; McCarthy, Cuskelly, van Kraayenoord, & Cohen, 2006). Alternatively, parents may differ in their predictors of stress (Hastings, 2003). While parents of children with ASD reported no differences in overall stress, children's problem behaviors and their partner's mental health were predictive of mothers' levels of stress

(Hastings, 2003). However, marital satisfaction positively predicted cohesion for both parents (Mitchell et al., 2016).

Expressed Emotion

Recently, researchers have focused on expressed emotion as an explanatory variable among families of children with disabilities. Expressed emotion is defined by high levels of criticism and/or emotional over-involvement from one family member directed toward another family member with a disability or illness. Expressed emotion is associated with relapse among adults with schizophrenia (Butzlaff & Hooley, 1998) and has been recently examined in families of children with intellectual disabilities (Hastings et al., 2006) as well as autism spectrum disorder (Greenberg, Seltzer, Hong, & Orsmond, 2006).

Research from Hastings et al. (2006) and Greenberg et al. (Greenberg et al., 2006; Greenberg et al., 2012) suggests a bidirectional relationship between child behavior problems and expressed emotion. Hastings et al. (2006) studied mothers of children with IDD at two time points, assessing maternal distress, mental health, expressed emotion, and child behavior problems. Criticism in particular was cross-sectionally, but not longitudinally, related to child externalizing behavior and maternal distress. Greenberg found links between high expressed emotion and maladaptive behavior in autism (Greenberg et al., 2006) and Fragile X syndrome (Greenberg et al., 2012). The latter study also found significant correlations between lack of criticism and fewer behavior problems. Smith et al. (2012) studied behavior problems and disability-related symptoms in adolescents and adults with FXS over a 3-year period, and found an association between maternal warmth, lower levels of symptoms, and higher levels of adaptive behavior. Maternal depression and criticism were associated with more psychological symptoms in individuals with FXS.

However, only examining the negative aspects of the family affective climate may be limiting for understanding the full range of family processes that influence development. Negative and positive well-being, for instance, are not necessarily on the same continuum of mental functioning. There remains a need to examine the positive aspects of emotional expression and other family processes among families with children with autism spectrum disorder.

Family Resilience

Family resilience can be conceptualized as a “renewable power” (Sobotková, 2004), a dynamic property of families’ strengths which allow families to adapt despite stressors. Sobotková (2004) identifies multiple factors that support family resilience: family functioning, stress appraisals, problem-solving skills, coping strategies, and sources of familial durability. Sources of familial durability can be external, such as socioeconomic status or social support networks, or internal, such as mutual value orientation and shared belief systems (Sobotková, 2004).

Several potential resilience factors have been explored, including hope (Lloyd & Hastings, 2009) and self-efficacy (Hastings & Brown, 2002). More recently, a systematic review by Peer and Hillman (2014) suggested that coping style, optimism, and social support may all be factors that influence resilience in parents of children with IDD. Ruiz-Robledillo, De Andrés-García, Pérez-Blasco, González-Bono, and Moya-Albiol (2014) found resilience showed associations with overall general physical and psychological health of caregivers of children with ASD. Research suggests that resilience is associated with mental health, such as anxiety, insomnia, and depression in caregivers (e.g., Tang et al., 2013). The Resilience Scale for Adults (RSA: Friborg, Hjemdal, Rosenvinge, & Martinussen, 2003) has been used in a small population of parents of children with IDD, as part of a comparison between parents of children with IDDs and parents of children with Sanfilippo syndrome (Grant et al., 2013). Parents rated social resources as their highest protective factor, and planned future as their lowest protective factor, both of which are subscales of the RSA.

Resilience research in the IDD field typically uses cross-sectional research. However, Bayat (2007) suggested resilience is a process that can only be shown over time, thus the need for longitudinal studies. One longitudinal study with parents of children with IDD (Gerstein, Crnic, Blacher, & Baker, 2009) studied trajectories of daily parenting stress in parents of young children with IDD, and found factors such as psychological well-being, marital adjustment, and positive relationships affected parenting stress differently in mothers and fathers, and concluded that parents affect each other's resilience. In contrast, Halstead, Griffith, and Hastings (2018) found no longitudinal association between resilience and maternal well-being outcomes, and little evidence of the role of resilience as a protective factor between child behavior problems and maternal well-being. However, they did observe maternal resilience functioned as a compensatory factor, having a significant independent main effect relationship with well-being outcomes in mothers of children with IDD and ASD.

Family Interventions

The Double ABCX model (McCubbin & Patterson, 1983) supports analysis of factors (i.e., protective resources, or risk factors including family discord) related to family adaptation (Saloviita, Itälinna, & Leinonen, 2003). The Double ABCX Model examines stressors that "pile up" post-crisis along with the original stressor (aA), post-crisis resources and those first used to meet the crisis (bB), post-crisis perceptions and those first applied to the crisis (cC) and the overall likelihood of adaptation to future crises on the basis of these developments (Xx).

Multiple components of family intervention programs can contribute to external support of family adaptation. Guralnick (1997, 1998) categorizes these intervention components as resource supports, social supports, and information and services. These components can be adapted to specific family needs to create a successful intervention capable of improving family patterns of interaction. Guralnick (1997, 1998) emphasizes that such interventions are primarily successful when they are delivered consistently with a family's goals, values, priorities and routines.

Mindfulness is typically described as being in the present moment, and responding without judgment to the unfolding of physical, mental, and emotional experiences (Kabat-Zinn, 2003). MBIs may help parents develop non-judgmental ways to focus on their children's behaviors, improving acceptance (de Bruin, Blom, Smit, van Steensel, & Bögels, 2015). Recent research demonstrates the efficacy of MBIs to improve well-being in caregivers of children with IDD (Neece, 2014). For parents of children with IDD, mindfulness is associated with reduced stress and anxiety, and increased positive parent-child interactions (Beer, Ward, & Moar, 2013). Chapters 9 and 42 explore further interventions for parents of children with IDD.

Conclusion

This chapter has reviewed risk and protective factors in the IDD field to date. Our approach considers multiple dimensions over the life course, with particular attention paid to social support, family environment, and individual differences. Moreover, our resilience-based approach has led us to consider how specific factors, such as the family environment, can present as both risk factors and protective factors based on differences in persons and circumstances. We believe that development of problem behaviors and poor mental health outcomes is best understood as a transactional process between the individual and complicated social systems including the family environment, school, and post-secondary outcomes such as residential placement. Exploring the interplay of risk and protective factors during these transactional processes is how best to identify targets for intervention. Continuing research is required to find new ways to promote resilience for people with IDD, and this research should focus on precise identification of risk and protective factors, conceived dynamically as changing over time, and changing in its significance for the individual with IDD based on persons and circumstances.

References

Abravanel, B. T., & Sinha, R. (2015). Emotion dysregulation mediates the relationship between lifetime cumulative adversity and depressive symptomatology. *Journal of Psychiatric Research, 61*, 89–96.

Al-Krenawi, A., Graham, J. R., & Al Gharaibeh, F. (2011). The impact of intellectual disability, caregiver burden, family functioning, marital quality, and sense of coherence. *Disability & Society, 26*(2), 139–150.

Alvord, M. K., & Grados, J. J. (2005). Enhancing resilience in children: A proactive approach. *Professional Psychology: Research and Practice, 36*(3), 238.

Amado, A. N. E. (1993). *Friendships and community connections between people with and without developmental disabilities*. Baltimore, MD: Paul H Brookes Publishing.

Baker, B. L., Blacher, J., Crnic, K. A., & Edelbrock, C. (2002). Behavior problems and parenting stress in families of three-year-old children with and without developmental delays. *American Journal on Mental Retardation*, 107(6), 433–444. November 2002.

Baker, B. L., McIntyre, L. L., Blacher, J., Crnic, K., Edelbrock, C., & Low, C. (2003). Pre-school children with and without developmental delay: Behaviour problems and parenting stress over time. *Journal of Intellectual Disability Research, 47*(4–5), 217–230.

Barnett, D. (1997). The effects of early intervention on maltreating parents and their children. In M. J. Guralnick (Ed.), *The effectiveness of early intervention* (pp. 147–170). Baltimore, MD: Brookes.

Bayat, M. (2007). Evidence of resilience in families of children with autism. *Journal of Intellectual Disability Research, 51*(9), 702–714.

Beer, M., Ward, L., & Moar, K. (2013). The relationship between mindful parenting and distress in parents of children with an autism spectrum disorder. *Mindfulness, 4*(2), 102–112.

Bellini, S., Peters, J. K., Benner, L., & Hopf, A. (2007). A meta-analysis of school-based social skills interventions for children with autism spectrum disorders. *Remedial and Special Education, 28*(3), 153–162.

Bender, W. N., Rosenkrans, C. B., & Crane, M. K. (1999). Stress, depression, and suicide among students with learning disabilities: Assessing the risk. *Learning Disability Quarterly, 22*(2), 143–156.

Benson, P. R., & Kersh, J. (2011). Marital quality and psychological adjustment among mothers of children with ASD: Cross-sectional and longitudinal relationships. *Journal of Autism and Developmental Disorders, 41*(12), 1675–1685.

Blacher, J., & McIntyre, L. L. (2006). Syndrome specificity and behavioural disorders in young adults with intellectual disability: Cultural differences in family impact. *Journal of Intellectual Disability Research, 50*(3), 184–198.

Blacher, J., & Hatton, C. (2007). Families in context: Influences on coping. In S. L. Odom, R. H. Horner, M. E. Snell, & J. Blacher (Eds.), *Handbook of developmental disabilities* (pp. 531–551). New York: The Guilford Press.

Bowman, R. A., Scotti, J. R., & Morris, T. L. (2010). Sexual abuse prevention: A training program for developmental disabilities service providers. *Journal of Child Sexual Abuse, 19*(2), 119–127.

Bramston, P., Fogarty, G., & Cummins, R. A. (1999). The nature of stressors reported by people with an intellectual disability. *Journal of Applied Research in Intellectual Disabilities, 12*(1), 1–10.

Bramston, P., Bostock, J., & Tehan, G. (1993). The measurement of stress in people with an intellectual disability: A pilot study. *International Journal of Disability, Development and Education, 40*(2), 95–104.

Brewin, C. R., Andrews, B., & Valentine, J. D. (2000). Meta-analysis of risk factors for posttraumatic stress disorder in trauma-exposed adults. *Journal of Consulting and Clinical Psychology, 68*(5), 748.

Bonanno, G. A. (2004). Loss, trauma, and human resilience: Have we underestimated the human capacity to thrive after extremely aversive events? *American Psychologist, 59*(1), 20.

Brooks, J. E. (2006). Strengthening resilience in children and youths: Maximizing opportunities in the schools. *Children and Schools, 28*(2), 69–76.

Butzlaff, R. L., & Hooley, J. M. (1998). Expressed emotion and psychiatric relapse: A meta-analysis. *Archives of General Psychiatry, 55*(6), 547–552.

Cacioppo, J. T., & Cacioppo, S. (2014). Social relationships and health: The toxic effects of perceived social isolation. *Social and Personality Psychology Compass, 8*(2), 58–72.

Carter, E. W., & Hughes, C. (2005). Increasing social interaction among adolescents with intellectual disabilities and their general education peers: Effective interventions. *Research and Practice for Persons with Severe Disabilities, 30*(4), 179–193.

Christensen, L. L., Fraynt, R. J., Neece, C. L., & Baker, B. L. (2012). Bullying adolescents with intellectual disability. *Journal of Mental Health Research in Intellectual Disabilities, 5*(1), 49–65.

Cohen, S., & Wills, T. A. (1985). Stress, social support, and the buffering hypothesis. *Psychological Bulletin, 98*(2), 310.

Cooper, S. A., Smiley, E., Morrison, J., Williamson, A., & Allan, L. (2007). Mental ill-health in adults with intellectual disabilities: Prevalence and associated factors. *The British Journal of Psychiatry, 190*(1), 27–35.

Crnic, K., Hoffman, C., Gaze, C., & Edelbrock, C. (2004). Understanding the emergence of behavior problems in young children with developmental delays. *Infants & Young Children, 17*(3), 223–235.

Crnic, K. A., Gaze, C., & Hoffman, C. (2005). Cumulative parenting stress across the preschool period: Relations to maternal parenting and child behaviour at age 5. *Infant and Child Development: An International Journal of Research and Practice, 14*(2), 117–132.

Cummins, R. A. (2005). Caregivers as managers of subjective wellbeing: A homeostatic perspective. *Journal of Applied Research in Intellectual Disabilities, 18*(4), 335–344.

Dabrowska, A., & Pisula, E. (2010). Parenting stress and coping styles in mothers and fathers of preschool children with autism and Down syndrome. *Journal of Intellectual Disability Research, 54*(3), 266–280.

de Bruin, E. I., Blom, R., Smit, F. M., van Steensel, F. J., & Bögels, S. M. (2015). MYmind: Mindfulness training for youngsters with autism spectrum disorders and their parents. *Autism, 19*(8), 906–914.

De Ruiter, K. P., Dekker, M. C., Verhulst, F. C., & Koot, H. M. (2007). Developmental course of psychopathology in youths with and without intellectual disabilities. *Journal of Child Psychology and Psychiatry, 48*(5), 498–507.

Di Nuovo, S., & Buono, S. (2011). Behavioral phenotypes of genetic syndromes with intellectual disability: Comparison of adaptive profiles. *Psychiatry Research, 189*(3), 440–445.

Dunn, M. E., Burbine, T., Bowers, C. A., & Tantledd-Dunn, S. (2001). Moderators of stress in parents of children with autism. *Community Mental Health Journal, 37*(1), 39–51.

Dykens, E. M. (2000). Annotation: Psychopathology in children with intellectual disability. *The Journal of Child Psychology and Psychiatry and Allied Disciplines, 41*(4), 407–417.

Einfeld, S. L., Ellis, L. A., & Emerson, E. (2011). Comorbidity of intellectual disability and mental disorder in children and adolescents: A systematic review. *Journal of Intellectual and Developmental Disability, 36*(2), 137–143.

Eisenhower, A. S., Baker, B. L., & Blacher, J. (2009). Children's delayed development and behavior problems: Impact on mothers' perceived physical health across early childhood. *Social Science & Medicine, 68*(1), 89–99.

Ekas, N. V., Lickenbrock, D. M., & Whitman, T. L. (2010). Optimism, social support, and well-being in mothers of children with autism spectrum disorder. *Journal of Autism and Developmental Disorders, 40*(10), 1274–1284.

Emerson, E. (2003). Mothers of children and adolescents with intellectual disability: Social and economic situation, mental health status, and the self-assessed social and psychological impact of the child's difficulties. *Journal of Intellectual Disability Research, 47*(4–5), 385–399.

Emerson, E. (2007). Poverty and people with intellectual disabilities. *Mental Retardation and Developmental Disabilities Research Reviews, 13*(2), 107–113.

Emerson, E., Baines, S., Allerton, L., & Welch, V. (2010). *Health inequalities and people with learning disabilities in the UK: 2010*. Durham: *Improving Health & Lives: Learning Disabilities Observatory*.

Emerson, E., & Hatton, C. (2007). Mental health of children and adolescents with intellectual disabilities in Britain. *The British Journal of Psychiatry, 191*(6), 493–499.

Emerson, E., & Hatton, C. (2009). Socioeconomic position, poverty, and family research. *International Review of Research in Mental Retardation, 37*, 97–129.

Emerson, E., & Jahoda, A. J. (2013). Social and psychological factors as determinants of emotional and behavioral difficulties. *Psychological Therapies for Adults with Intellectual Disabilities*, 15–30.

Esbensen, A. J., & Benson, B. A. (2006). A prospective analysis of life events, problem behaviours and depression in adults with intellectual disability. *Journal of Intellectual Disability Research, 50*(4), 248–258.

Falk, N. H., Norris, K., & Quinn, M. G. (2014). The factors predicting stress, anxiety and depression in the parents of children with autism. *Journal of Autism and Developmental Disorders, 44*(12), 3185–3203.

Feldman, M. A. (1997). The effectiveness of early intervention for children of parents with mental retardation. In M. J. Guralnick (Ed.), *The effectiveness of early intervention* (pp. 171–191). Baltimore, MD: Brookes.

Finzi-Dottan, R., & Karu, T. (2006). From emotional abuse in childhood to psychopathology in adulthood: A path mediated by immature defense mechanisms and self-esteem. *The Journal of Nervous and Mental Disease, 194*(8), 616–621.

Fisher, M. H., Moskowitz, A. L., & Hodapp, R. M. (2013). Differences in social vulnerability among individuals with autism spectrum disorder, Williams syndrome, and Down syndrome. *Research in Autism Spectrum Disorders, 7*(8), 931–937.

Fisher, M. H., Hodapp, R. M., & Dykens, E. M. (2008). Child abuse among children with disabilities: What we know and what we need to know. *International Review of Research in Mental Retardation, 35*, 251–289.

Fletcher, R., Loschen, E., Stavrakaki, C., & First, M. (2007). DM-ID: Diagnostic manual—intellectual disability: A textbook of diagnosis of mental disorders in persons with intellectual disability. *Journal of Mental Health Research in Intellectual Disabilities*, 29.

Fogarty, G. J., Bramston, P., & Cummins, R. A. (1997). Validation of the Lifestress Inventory for people with a mild intellectual disability. *Research in Developmental Disabilities, 18*(6), 435–456.

Foley, K. R., Dyke, P., Girdler, S., Bourke, J., & Leonard, H. (2012). Young adults with intellectual disability transitioning from school to post-school: A literature review framed within the ICF. *Disability and Rehabilitation, 34*(20), 1747–1764.

Fontil, L., & Petrakos, H. H. (2015). Transition to school: The experiences of Canadian and immigrant families of children with autism spectrum disorders. *Psychology in the Schools, 52*(8), 773–788.

Forte, M., Jahoda, A., & Dagnan, D. (2011). An anxious time? Exploring the nature of worries experienced by young people with a mild to moderate intellectual disability as they make the transition to adulthood. *British Journal of Clinical Psychology, 50*(4), 398–411.

Freedman, R. I., & Boyer, N. C. (2000). The power to choose: Supports for families caring for individuals with developmental disabilities. *Health & Social Work, 25*(1), 59–68.

Friborg, O., Hjemdal, O., Rosenvinge, J. H., & Martinussen, M. (2003). A new rating scale for adult resilience: What are the central protective resources behind healthy adjustment? *International Journal of Methods in Psychiatric Research, 12*(2), 65–76.

Garmezy, N., Masten, A. S., & Tellegen, A. (1984). The study of stress and competence in children: A building block for developmental psychopathology. *Child Development*, 97–111.

Garmezy, N. (1985). Stress-resistant children: The search for protective factors. *Recent Research in Developmental Psychopathology, 4*, 213–233.

Garnefski, N., Kraaij, V., & Spinhoven, P. (2001). Negative life events, cognitive emotion regulation and emotional problems. *Personality and Individual Differences, 30*(8), 1311–1327.

Gates, J. A., Kang, E., & Lerner, M. D. (2017). Efficacy of group social skills interventions for youth with autism spectrum disorder: A systematic review and meta-analysis. *Clinical Psychology Review, 52*, 164–181.

Gerstein, E. D., Crnic, K., Blacher, J., & Baker, B. L. (2009). Resilience and the course of daily parenting stress in families of young children with intellectual disabilities. *Journal of Intellectual Disability Research, 53*(12), 981–997.

Ghaziuddin, M. (1988). Behavioural disorder in the mentally handicapped: The role of life events. *The British Journal of Psychiatry, 152*(5), 683–686.

Gillott, A., & Standen, P. J. (2007). Levels of anxiety and sources of stress in adults with autism. *Journal of Intellectual Disabilities, 11*(4), 359–370.

Gilmore, L., & Cuskelly, M. (2014). Vulnerability to loneliness in people with intellectual disability: An explanatory model. *Journal of Policy and Practice in Intellectual Disabilities, 11*(3), 192–199.

Gilmore, L., Campbell, M., Shochet, I., & Roberts, C. (2013). Resiliency profiles of children with intellectual disability and their typically developing peers. *Psychology in the Schools, 50*(10), 1032–1043.

Gilrane-McGarry, U., & Taggart, L. (2007). An exploration of the support received by people with intellectual disabilities who have been bereaved. *Journal of Research in Nursing, 12*(2), 129–144.

Glidden, L. M., & Schoolcraft, S. A. (2003). Depression: Its trajectory and correlates in mothers rearing children with intellectual disability. *Journal of Intellectual Disability Research, 47*(4–5), 250–263.

Goff, B. S., Monk, J. K., Malone, J., Staats, N., Tanner, A., & Springer, N. P. (2016). Comparing parents of children with Down syndrome at different life span stages. *Journal of Marriage and Family, 78*(4), 1131–1148.

Grant, S., Cross, E., Wraith, J. E., Jones, S., Mahon, L., Lomax, M., … Hare, D. (2013). Parental social support, coping strategies, resilience factors, stress, anxiety and depression levels in parents of children with MPS III (Sanfilippo syndrome) or children with intellectual disabilities (ID). *Journal of Inherited Metabolic Disease, 36*(2), 281–291.

Gratz, K. L., & Roemer, L. (2004). Multidimensional assessment of emotion regulation and dysregulation: Development, factor structure, and initial validation of the difficulties in emotion regulation scale. *Journal of Psychopathology and Behavioral Assessment, 26*(1), 41–54.

Greenberg, J. S., Seltzer, M. M., Hong, J., & Orsmond, G. I. (2006). Bidirectional effects of expressed emotion and behavior problems and symptoms in adolescents and adults with autism. *American Journal on Mental Retardation, 111*(4), 229–249.

Greenberg, J. S., Seltzer, M. M., Baker, J. K., Smith, L. E., Warren, S. F., Brady, N., & Hong, J. (2012). Family environment and behavior problems in children, adolescents, and adults with fragile X syndrome. *American Journal on Intellectual and Developmental Disabilities, 117*(4), 331–346.

Griffith, G. M., Hutchinson, L., & Hastings, R. P. (2013). "I'm not a patient, I'm a person": The experiences of individuals with intellectual disabilities and challenging behavior—A thematic synthesis of qualitative studies. *Clinical Psychology: Science and Practice, 20*(4), 469–488.

Guralnick, M. J. (1997). Effectiveness of early intervention for vulnerable children: A developmental perspective. *American Journal on Mental Retardation, 102*(4), 319–345.

Guralnick, M. J. (1998). The effectiveness of early intervention for vulnerable children: A developmental perspective. *American Journal on Mental Retardation, 102*, 319–345.

Guralnick, M. J. (2005). Early intervention for children with intellectual disabilities: Current knowledge and future prospects. *Journal of Applied Research in Intellectual Disabilities, 18*(4), 313–324.

Guralnick, M. J. (2006). Peer relationships and the mental health of young children with intellectual delays. *Journal of Policy and Practice in Intellectual Disabilities, 3*(1), 49–56.

Halstead, E., Ekas, N., Hastings, R. P., & Griffith, G. M. (2018). Associations between resilience and the well-being of mothers of children with autism spectrum disorder and other developmental disabilities. *Journal of Autism and Developmental Disorders, 48*(4), 1108–1121.

Halstead, E. J., Griffith, G. M., & Hastings, R. P. (2018). Social support, coping, and positive perceptions as potential protective factors for the well-being of mothers of children with intellectual and developmental disabilities. *International Journal of Developmental Disabilities, 64*(4-5), 288–289.

Harkness, K. L., & Monroe, S. M. (2016). The assessment and measurement of adult life stress: Basic premises, operational principles, and design requirements. *Journal of Abnormal Psychology, 125*(5), 727.

Hartley, S. L., & MacLean, W. E., Jr. (2005). Perceptions of stress and coping strategies among adults with mild mental retardation: Insight into psychological distress. *American Journal on Mental Retardation, 110*(4), 285–297.

Hartley, S. L., & MacLean, W. E., Jr. (2009). Depression in adults with mild intellectual disability: Role of stress, attributions, and coping. *American Journal on Intellectual and Developmental Disabilities, 114*(3), 147–160.

Hartley, S. L., Barker, E. T., Seltzer, M. M., Floyd, F., Greenberg, J., Orsmond, G., & Bolt, D. (2010). The relative risk and timing of divorce in families of children with an autism spectrum disorder. *Journal of Family Psychology, 24*(4), 449.

Hastings, R. P. (2003). Child behaviour problems and partner mental health as correlates of stress in mothers and fathers of children with autism. *Journal of Intellectual Disability Research, 47*(4–5), 231–237.

Hastings, R. P. (2016). Do children with intellectual and developmental disabilities have a negative impact on other family members? The case for rejecting a negative narrative. In *International review of research in developmental disabilities* (Vol. 50, pp. 165–194). New York, NY: Academic Press.

Hastings, R. P., Allen, R., McDermott, K., & Still, D. (2002). Factors related to positive perceptions in mothers of children with intellectual disabilities. *Journal of Applied Research in Intellectual Disabilities, 15*(3), 269–275.

Hastings, R. P., & Brown, T. (2002). Behavior problems of children with autism, parental self-efficacy, and mental health. *American Journal on Mental Retardation, 107*(3), 222–232.

Hastings, R. P., & Taunt, H. M. (2002). Positive perceptions in families of children with developmental disabilities. *American Journal on Mental Retardation, 107*(2), 116–127.

Hastings, R. P., & Beck, A. (2004). Practitioner review: Stress intervention for parents of children with intellectual disabilities. *Journal of Child Psychology and Psychiatry, 45*(8), 1338–1349.

Hastings, R. P., Hatton, C., Taylor, J. L., & Maddison, C. (2004). Life events and psychiatric symptoms in adults with intellectual disabilities. *Journal of Intellectual Disability Research, 48*(1), 42–46.

Hastings, R. P., Kovshoff, H., Ward, N. J., Degli Espinosa, F., Brown, T., & Remington, B. (2005). Systems analysis of stress and positive perceptions in mothers and fathers of pre-school children with autism. *Journal of Autism and Developmental Disorders, 35*(5), 635.

Hastings, R. P., Daley, D., Burns, C., & Beck, A. (2006). Maternal distress and expressed emotion: Cross-sectional and longitudinal relationships with behavior problems of children with intellectual disabilities. *American Journal on Mental Retardation, 111*(1), 48–61.

Hastings, R. P., & Lloyd, T. (2007). Expressed emotion in families of children and adults with intellectual disabilities. *Mental Retardation and Developmental Disabilities Research Reviews, 13*(4), 339–345.

Hatton, C., & Emerson, E. (2004). The relationship between life events and psychopathology amongst children with intellectual disabilities. *Journal of Applied Research in Intellectual Disabilities, 17*(2), 109–117.

Hawkins, J. D., Catalano, R. F., & Miller, J. Y. (1992). Risk and protective factors for alcohol and other drug problems in adolescence and early adulthood: Implications for substance abuse prevention. *Psychological Bulletin, 112*(1), 64.

Heal, L. W., & Sigelman, C. K. (1995). Response biases in interviews of individuals with limited mental ability. *Journal of Intellectual Disability Research, 39*(4), 331–340.

Heiman, T. (2002). Parents of children with disabilities: Resilience, coping, and future expectations. *Journal of Developmental and Physical Disabilities, 14*(2), 159–171.

Hirst, M., Jervis, N., Visagie, K., Sojo, V., & Cavanagh, S. (2011). *Transition to primary school: A review of the literature*. Canberra: Department of Health and Ageing.

Hoare, P., Harris, M., Jackson, P., & Kerley, S. (1998). A community survey of children with severe intellectual disability and their families: Psychological adjustment, carer distress and the effect of respite care. *Journal of Intellectual Disability Research, 42*(3), 218–227.

Hodapp, R. M., & Dykens, E. M. (2009). Intellectual disabilities and child psychiatry: Looking to the future. *Journal of Child Psychology and Psychiatry, 50*(1–2), 99–107.

Holden, B., & Gitlesen, J. P. (2009). The overlap between psychiatric symptoms and challenging behaviour: A preliminary study. *Research in Developmental Disabilities, 30*(2), 210–218.

Hollins, S., & Esterhuyzen, A. (1997). Bereavement and grief in adults with learning disabilities. *The British Journal of Psychiatry, 170*(6), 497–501.

Howlin, P., Wing, L., & Gould, J. (1995). The recognition of autism in children with Down syndrome-implications for intervention and some speculations about pathology. *Developmental Medicine & Child Neurology, 37*(5), 406–414.

Hulbert-Williams, L., Hastings, R. P., Crowe, R., & Pemberton, J. (2011). Self-reported life events, social support and psychological problems in adults with intellectual disabilities. *Journal of Applied Research in Intellectual Disabilities, 24*(5), 427–436.

Hulbert-Williams, L., & Hastings, R. P. (2008). Life events as a risk factor for psychological problems in individuals with intellectual disabilities: A critical review. *Journal of Intellectual Disability Research, 52*(11), 883–895.

Irazábal, M., Marsà, F., García, M., Gutiérrez-Recacha, P., Martorell, A., Salvador-Carulla, L., & Ochoa, S. (2012). Family burden related to clinical and functional variables of people with intellectual disability with and without a mental disorder. *Research in Developmental Disabilities, 33*(3), 796–803.

Jacola, L. M., Hickey, F., Howe, S. R., Esbensen, A., & Shear, P. K. (2014). Behavior and adaptive functioning in adolescents with Down syndrome: Specifying targets for intervention. *Journal of Mental Health Research in Intellectual Disabilities, 7*(4), 287–305.

Janssen, C. G. C., Schuengel, C., & Stolk, J. (2002). Understanding challenging behaviour in people with severe and profound intellectual disability: A stress-attachment model. *Journal of Intellectual Disability Research, 46*(6), 445–453.

Jessor, R., Van Den Bos, J., Vanderryn, J., Costa, F. M., & Turbin, M. S. (1995). Protective factors in adolescent problem behavior: Moderator effects and developmental change. *Developmental Psychology, 31*(6), 923.

Kabat-Zinn, J. (2003). Mindfulness-based interventions in context: Past, present, and future. *Clinical Psychology: Science and Practice, 10*(2), 144–156.

Kau, A. S., Tierney, E., Bukelis, I., Stump, M. H., Kates, W. R., Trescher, W. H., & Kaufmann, W. E. (2004). Social behavior profile in young males with fragile X syndrome: Characteristics and specificity. *American Journal of Medical Genetics Part A, 126*(1), 9–17.

Kersh, J., Hedvat, T. T., Hauser-Cram, P., & Warfield, M. E. (2006). The contribution of marital quality to the well-being of parents of children with developmental disabilities. *Journal of Intellectual Disability Research, 50*(12), 883–893.

Kessler, R. C., Sonnega, A., Bromet, E., Hughes, M., & Nelson, C. B. (1995). Posttraumatic stress disorder in the National Comorbidity Survey. *Archives of General Psychiatry, 52*(12), 1048–1060.

Kiddle, H., & Dagnan, D. (2011). Vulnerability to depression in adolescents with intellectual disabilities. *Advances in Mental Health and Intellectual Disabilities, 5*(1), 3–8.

Kraemer, H. C., Kazdin, A. E., Offord, D. R., Kessler, R. C., Jensen, P. S., & Kupfer, D. J. (1997). Coming to terms with the terms of risk. *Archives of General Psychiatry, 54*(4), 337–343.

Larkin, P., Jahoda, A., MacMahon, K., & Pert, C. (2012). Interpersonal sources of conflict in young people with and without mild to moderate intellectual disabilities at transition from adolescence to adulthood. *Journal of Applied Research in Intellectual Disabilities, 25*(1), 29–38.

Lazarus, R. S. (1991). *Emotion and adaptation*. New York: Oxford University Press on Demand.

Levitas, A. S., & Gilson, S. F. (2001). Predictable crises in the lives of people with mental retardation. *Mental Health Aspects of Developmental Disabilities, 4*(3), 89–100.

LeRoy, B. W., Walsh, P. N., Kulik, N., & Rooney, M. (2004). Retreat and resilience: Life experiences of older women with intellectual disabilities. *American Journal on Mental Retardation, 109*(5), 429–441.

Lippold, T., & Burns, J. (2009). Social support and intellectual disabilities: A comparison between social networks of adults with intellectual disability and those with physical disability. *Journal of Intellectual Disability Research, 53*(5), 463–473.

Lloyd, T. J., & Hastings, R. (2009). Hope as a psychological resilience factor in mothers and fathers of children with intellectual disabilities. *Journal of Intellectual Disability Research, 53*(12), 957–968.

Locke, J., Ishijima, E. H., Kasari, C., & London, N. (2010). Loneliness, friendship quality and the social networks of adolescents with high-functioning autism in an inclusive school setting. *Journal of Research in Special Educational Needs, 10*(2), 74–81.

Lovell, B., Moss, M., & Wetherell, M. A. (2012). With a little help from my friends: Psychological, endocrine and health corollaries of social support in parental caregivers of children with autism or ADHD. *Research in Developmental Disabilities, 33*(2), 682–687.

Lueckling, R. G., & Fabian, E. S. (1997). Transition from school to work. In S. M. Pueschel & M. Sustrova (Eds.), *Adolescents with down syndrome: Toward a more fulfilling life* (pp. 235–243). Baltimore, MD: Brookes.

Lunsky, Y. (2003). Depressive symptoms in intellectual disability: Does gender play a role? *Journal of Intellectual Disability Research, 47*(6), 417–427.

Lunsky, Y. (2008). The impact of stress and social support on the mental health of individuals with intellectual disabilities. *Salud Pública de México, 50*, s151–s153.

Lunsky, Y., & Bramston, P. (2006). A preliminary study of perceived stress in adults with intellectual disabilities according to self-report and informant ratings. *Journal of Intellectual and Developmental Disability, 31*(1), 20–27.

Lunsky, Y., & Benson, B. A. (2001). Association between perceived social support and strain, and positive and negative outcome for adults with mild intellectual disability. *Journal of Intellectual Disability Research, 45*(2), 106–114.

Lunsky, Y., & Havercamp, S. M. (1999). Distinguishing Low Levels of Social Implications for Dual Diagnosis. *American Journal on Mental Retardation, 104*(2), 200–204.

Luthar, S. S. (1991). Vulnerability and resilience: A study of high-risk adolescents. *Child Development, 62*(3), 600–616.

Maag, J. W., & Reid, R. (2006). Depression among students with learning disabilities: Assessing the risk. *Journal of Learning Disabilities, 39*(1), 3–10.

Magiati, I., Tay, X. W., & Howlin, P. (2014). Cognitive, language, social and behavioural outcomes in adults with autism spectrum disorders: A systematic review of longitudinal follow-up studies in adulthood. *Clinical Psychology Review, 34*(1), 73–86.

Mak, W. W., & Ho, G. S. (2007). Caregiving perceptions of Chinese mothers of children with intellectual disability in Hong Kong. *Journal of Applied Research in Intellectual Disabilities, 20*(2), 145–156.

Martorell, A., & Tsakanikos, E. (2008). Traumatic experiences and life events in people with intellectual disability. *Current Opinion in Psychiatry, 21*(5), 445–448.

Masten, A. S. (2001). Ordinary magic: Resilience processes in development. *American Psychologist, 56*(3), 227–238.

Masten, A. S. (2011). Resilience in children threatened by extreme adversity: Frameworks for research, practice, and translational synergy. *Development and Psychopathology, 23*(2), 493–506.

Masten, A. S., Narayan, A. J., Silverman, W. K., & Osofsky, J. D. (2015). Children in war and disaster. *Handbook of Child Psychology and Developmental Science, 4*, 704–745.

McCarthy, A., Cuskelly, M., van Kraayenoord, C. E., & Cohen, J. (2006). Predictors of stress in mothers and fathers of children with fragile X syndrome. *Research in Developmental Disabilities, 27*(6), 688–704.

McClure, K. S., Hapern, J., Wolper, P., & Donahue, J. J. (2009). *Emotion regulation and intellectual disability*. Philadelphia, PA: Journal on Developmental Disabilities.

McCubbin, H. I., & Patterson, J. M. (1983). The family stress process: The double ABCX model of adjustment and adaptation. *Marriage & Family Review, 6*(1–2), 7–37.

Miller, M. (2002). Resilience elements in students with learning disabilities. *Journal of Clinical Psychology, 58*(3), 291–298.

Mitchell, D. B., Szczerepa, A., & Hauser-Cram, P. (2016). Spilling over: Partner parenting stress as a predictor of family cohesion in parents of adolescents with developmental disabilities. *Research in Developmental Disabilities, 49*, 258–267.

Myrbakk, E., & von Tetzchner, S. (2008). Psychiatric disorders and behavior problems in people with intellectual disability. *Research in Developmental Disabilities, 29*(4), 316–332.

Murphy, G. H., O'Callaghan, A. C., & Clare, I. C. (2007). The impact of alleged abuse on behaviour in adults with severe intellectual disabilities. *Journal of Intellectual Disability Research, 51*(10), 741–749.

Neece, C. L. (2014). Mindfulness-based stress reduction for parents of young children with developmental delays: Implications for parental mental health and child behavior problems. *Journal of Applied Research in Intellectual Disabilities, 27*(2), 174–186.

Neece, C. L., Kraemer, B. R., & Blacher, J. (2009). Transition satisfaction and family well being among parents of young adults with severe intellectual disability. *Intellectual and Developmental Disabilities, 47*(1), 31–43.

Nezu, C. M., Nezu, A. M., Rothenberg, J. L., DelliCarpini, L., & Groag, I. (1995). Depression in adults with mild mental retardation: Are cognitive variables involved? *Cognitive Therapy and Research, 19*(2), 227–239.

Oelofsen, N., & Richardson, P. (2006). Sense of coherence and parenting stress in mothers and fathers of preschool children with developmental disability. *Journal of Intellectual and Developmental Disability, 31*(1), 1–12.

Olson, G. I., & Schober, B. I. (1993). The satisfied poor. *Social Indicators Research, 28*(2), 173–193.

Olsson, M. B., & Hwang, C. P. (2001). Depression in mothers and fathers of children with intellectual disability. *Journal of Intellectual Disability Research, 45*(6), 535–543.

Olsson, M. B., & Hwang, C. P. (2008). Socioeconomic and psychological variables as risk and protective factors for parental well-being in families of children with intellectual disabilities. *Journal of Intellectual Disability Research, 52*(12), 1102–1113.

Odom, S. L., Zercher, C., Li, S., Marquart, J. M., Sandall, S., & Brown, W. H. (2006). Social acceptance and rejection of preschool children with disabilities: A mixed-method analysis. *Journal of Educational Psychology, 98*(4), 807.

Orsmond, G. I., Seltzer, M. M., Greenberg, J. S., & Krauss, M. W. (2006). Mother–child relationship quality among adolescents and adults with autism. *American Journal on Mental Retardation, 111*(2), 121–137.

Owen, D. M., Hastings, R. P., Noone, S. J., Chinn, J., Harman, K., Roberts, J., & Taylor, K. (2004). Life events as correlates of problem behavior and mental health in a residential population of adults with developmental disabilities. *Research in Developmental Disabilities, 25*(4), 309–320.

Ozbay, F., Johnson, D. C., Dimoulas, E., Morgan, C. A., III, Charney, D., & Southwick, S. (2007). Social support and resilience to stress: From neurobiology to clinical practice. *Psychiatry (Edgmont), 4*(5), 35.

Parish, S. L., Seltzer, M. M., Greenberg, J. S., & Floyd, F. (2004). Economic implications of caregiving at midlife: Comparing parents with and without children who have developmental disabilities. *Mental Retardation, 42*(6), 413–426.

Peer, J. W., & Hillman, S. B. (2014). Stress and resilience for parents of children with intellectual and developmental disabilities: A review of key factors and recommendations for practitioners. *Journal of Policy and Practice in Intellectual Disabilities, 11*(2), 92–98.

Phelps, J. L., Belsky, J., & Crnic, K. (1998). Earned security, daily stress, and parenting: A comparison of five alternative models. *Development and Psychopathology, 10*, 21–38.

Plant, K. M., & Sanders, M. R. (2007). Predictors of caregiver stress in families of preschool-aged children with developmental disabilities. *Journal of Intellectual Disability Research, 51*(2), 109–124.

Platt, J., Keyes, K. M., & Koenen, K. C. (2014). Size of the social network versus quality of social support: Which is more protective against PTSD? *Social Psychiatry and Psychiatric Epidemiology, 49*(8), 1279–1286.

Pudrovska, T. (2009). Parenthood, stress, and mental health in late midlife and early old age. *The International Journal of Aging and Human Development, 68*(2), 127–147.

Raghavan, R., & Pawson, N. (2008). Transition and social networks of young people with learning disabilities. *Advances in Mental Health and Learning Disabilities, 2*(3), 25–28.

Raskind, M. H., Goldberg, R. J., Higgins, E. L., & Herman, K. L. (1999). Patterns of change and predictors of success in individuals with learning disabilities: Results from a twenty-year longitudinal study. *Learning Disabilities Research & Practice, 14*(1), 35–49.

Reiter, S., & Lapidot-Lefler, N. (2007). Bullying among special education students with intellectual disabilities: Differences in social adjustment and social skills. *Intellectual and Developmental Disabilities, 45*(3), 174–181.

Robinson, S., Weiss, J. A., Lunsky, Y., & Ouellette-Kuntz, H. (2016). Informal support and burden among parents of adults with intellectual and/or developmental disabilities. *Journal of Applied Research in Intellectual Disabilities, 29*(4), 356–365.

Roe, J., & Gray, A. (1991, November). Teachers' professional self-esteem in the light of occupational stress factors. In *Australian Association for Research in Education Annual Conference, Surfers Paradise, Gold Coast, Australia (26–30 November).*

Rose, C. A., & Espelage, D. L. (2012). Risk and protective factors associated with the bullying involvement of students with emotional and behavioral disorders. *Behavioral Disorders, 37*(3), 133–148.

Ruiz-Robledillo, N., De Andrés-García, S., Pérez-Blasco, J., González-Bono, E., & Moya-Albiol, L. (2014). Highly resilient coping entails better perceived health, high social support and low morning cortisol levels in parents of children with autism spectrum disorder. *Research in Developmental Disabilities, 35*(3), 686–695.

Rutter, M. (1987). Psychosocial resilience and protective mechanisms. *American Journal of Orthopsychiatry, 57*(3), 316–331.

Rutter, M. (2006). Implications of resilience concepts for scientific understanding. *Annals of the New York Academy of Sciences, 1094*(1), 1–12.

Ryan, R. (1994). Post-traumatic stress disorder in persons with developmental disabilities. *Community Mental Health Journal*, (1), 45–54.

Saloviita, T., Itälinna, M., & Leinonen, E. (2003). Explaining the parental stress of fathers and mothers caring for a child with intellectual disability: A double ABCX model. *Journal of Intellectual Disability Research, 47*(4–5), 300–312.

Santiago, C. D., Wadsworth, M. E., & Stump, J. (2011). Socioeconomic status, neighborhood disadvantage, and poverty-related stress: Prospective effects on psychological syndromes among diverse low-income families. *Journal of Economic Psychology, 32*(2), 218–230.

Schafer, V., & Semrud-Clikeman, M. (2008). Neuropsychological functioning in subgroups of children with and without social perception deficits and/or hyperactivity—Impulsivity. *Journal of Attention Disorders, 12*(2), 177–190.

Scorgie, K., & Sobsey, D. (2000). Transformational outcomes associated with parenting children who have disabilities. *Mental Retardation, 38*(3), 195–206.

Scott, H. M., & Havercamp, S. M. (2014). Mental health for people with intellectual disability: The impact of stress and social support. *American Journal on Intellectual and Developmental Disabilities, 119*(6), 552–564.

Seifer, R. & Dickstein, S. (2000) Parental mental illness and infant development. In Handbook of Infant Mental Health 2nd Edition, C. H. Zeanah Jr), pp. 145–160. The Guilford Press, New York.

Seltzer, M., Krauss, M. W., Hong, J., & Orsmond, G. I. (2001). Continuity or discontinuity of family involvement following residential transitions of adults who have mental retardation. *Mental Retardation, 39*(3), 181–194.

Seltzer, M. M., Floyd, F., Song, J., Greenberg, J., & Hong, J. (2011). Midlife and aging parents of adults with intellectual and developmental disabilities: Impacts of lifelong parenting. *American Journal on Intellectual and Developmental Disabilities, 116*(6), 479–499.

Shattuck, P. T., Wagner, M., Narendorf, S., Sterzing, P., & Hensley, M. (2011). Post–high school service use among young adults with an autism spectrum disorder. *Archives of Pediatrics & Adolescent Medicine, 165*(2), 141–146.

Singer, G. H. (2006). Meta-analysis of comparative studies of depression in mothers of children with and without developmental disabilities. *American Journal on Mental Retardation, 111*(3), 155–169.

Sloper, P., & Turner, S. (1992). Service needs of families of children with severe physical disability. *Child: Care, Health and Development, 18*(5), 259–282.

Smith, L. E., Barker, E. T., Seltzer, M. M., Abbeduto, L., & Greenberg, J. S. (2012). Behavioral phenotype of fragile X syndrome in adolescence and adulthood. *American Journal on Intellectual and Developmental Disabilities, 117*(1), 1–17.

Sobotková, I. (2004). Rodinná resilience. *Československá psychologie*, (3).

Spini, D., Hanappi, D., Bernardi, L., Oris, M., & Bickel, J. F. (2013). Vulnerability across the life course: A theoretical framework and research directions. *LIVES Working Papers, 2013*(27), 1–35.

Stancliffe, R. J., Wilson, N. J., Bigby, C., Balandin, S., & Craig, D. (2014). Responsiveness to self-report questions about loneliness: A comparison of mainstream and intellectual disability-specific instruments. *Journal of Intellectual Disability Research, 58*(5), 399–405.

Stoneman, Z. (2007). Examining the Down syndrome advantage: Mothers and fathers of young children with disabilities. *Journal of Intellectual Disability Research, 51*(12), 1006–1017.

Stuart, M., & McGrew, J. H. (2009). Caregiver burden after receiving a diagnosis of an autism spectrum disorder. *Research in Autism Spectrum Disorders, 3*(1), 86–97.

Szumski, G., Smogorzewska, J., Grygiel, P., & Orlando, A. M. (2017). Examining the effectiveness of naturalistic social skills training in developing social skills and theory of mind in preschoolers with ASD. *Journal of Autism and Developmental Disorders*, 1–16.

Taggart, L. (2014). Health promotion for people with intellectual disabilities. *Journal of Applied Research in Intellectual Disabilities, 27*(4), 290–290.

Tang, S. T., Huang, G. H., Wei, Y. C., Chang, W. C., Chen, J. S., & Chou, W. C. (2013). Trajectories of caregiver depressive symptoms while providing end-of-life care. *Psycho-Oncology, 22*(12), 2702–2710.

Taylor, J. L., Hatton, C., Dixon, L., & Douglas, C. (2004). Screening for psychiatric symptoms: PAS-ADD Checklist norms for adults with intellectual disabilities. *Journal of Intellectual Disability Research, 48*(1), 37–41.

Tennant, C. (2002). Life events, stress and depression: A review of recent findings. *Australian & New Zealand Journal of Psychiatry, 36*(2), 173–182.

Totsika, V., Hastings, R. P., Emerson, E., Lancaster, G. A., & Berridge, D. M. (2011). A population-based investigation of behavioural and emotional problems and maternal mental health: Associations with autism spectrum disorder and intellectual disability. *Journal of Child Psychology and Psychiatry, 52*(1), 91–99.

Totsika, V., Hastings, R. P., Emerson, E., Lancaster, G. A., Berridge, D. M., & Vagenas, D. (2013). Is there a bidirectional relationship between maternal well-being and child behavior problems in autism spectrum disorders? Longitudinal analysis of a population-defined sample of young children. *Autism Research, 6*(3), 201–211.

Tsakanikos, E., Costello, H., Holt, G., Bouras, N., Sturmey, P., & Newton, T. (2006). Psychopathology in adults with autism and intellectual disability. *Journal of Autism and Developmental Disorders, 36*(8), 1123–1129.

Turner, R. J., & Wheaton, B. (1995). Checklist measurement of stressful life events. *Measuring Stress: A Guide for Health and Social Scientists*, 29–58.

Valicenti-McDermott, M., Lawson, K., Hottinger, K., Seijo, R., Schechtman, M., Shulman, L., & Shinnar, S. (2015). Parental stress in families of children with autism and other developmental disabilities. *Journal of Child Neurology, 30*(13), 1728–1735.

Vaughn, S., & Haager, D. (1994). Social competence as a multifaceted construct: How do students with learning disabilities fare? *Learning Disability Quarterly, 17*(4), 253–266.

Werner, E. E. (1989). High-risk children in young adulthood: A longitudinal study from birth to 32 years. *American Journal of Orthopsychiatry, 59*(1), 72–81.

Werner, E. E. (1995). Resilience in development. *Current Directions in Psychological Science, 4*, 81–85.

Werner, E. E. (2000). Protective factors and individual resilience. In S. J. Meisels & J. P. Shonkoff (Eds.), *Handbook of early childhood intervention* (pp. 97–116). New York, NY: Cambridge University Press.

Wigham, S., Taylor, J. L., & Hatton, C. (2014). A prospective study of the relationship between adverse life events and trauma in adults with mild to moderate intellectual disabilities. *Journal of Intellectual Disability Research, 58*(12), 1131–1140.

Wigham, S., & Emerson, E. (2015). Trauma and life events in adults with intellectual disability. *Current Developmental Disorders Reports, 2*(2), 93–99.

Williams, V., & Heslop, P. (2005). Mental health support needs of people with a learning difficulty: A medical or a social model? *Disability & Society, 20*(3), 231–245.

Williams, V., & Heslop, P. (2006). Filling the emotional gap at transition: Young people with learning difficulties and friendship. *Tizard Learning Disability Review, 11*(4), 28–37.

White, N., & Hastings, R. P. (2004). Social and professional support for parents of adolescents with severe intellectual disabilities. *Journal of Applied Research in Intellectual Disabilities, 17*(3), 181–190.

Woodman, A. C. (2014). Trajectories of stress among parents of children with disabilities: A dyadic analysis. *Family Relations, 63*(1), 39–54.

Wong, B. Y. (2003). General and specific issues for researchers' consideration in applying the risk and resilience framework to the social domain of learning disabilities. *Learning Disabilities: Research & Practice, 18*(2), 68–76.

Zaidman-Zait, A., Most, T., Tarrasch, R., & Haddad, E. (2018). Mothers' and fathers' involvement in intervention programs for deaf and hard of hearing children. *Disability and Rehabilitation, 40*(11), 1301–1309.

Zaidman-Zait, A., Mirenda, P., Duku, E., Vaillancourt, T., Smith, I. M., Szatmari, P., … Zwaigenbaum, L. (2017). Impact of personal and social resources on parenting stress in mothers of children with autism spectrum disorder. *Autism, 21*(2), 155–166.

20

Environmental Chemical Exposures and Intellectual Disability in Children

David C. Bellinger

Although environmental chemical exposures impair the function of many organ systems, the central nervous system (CNS) is considered to be especially vulnerable, in large part because of the potential life-long consequences of perturbations in its development. Six of the 10 chemicals identified by the WHO as being of greatest public health concern globally are known to adversely affect brain-based functions and their development (air pollution, arsenic, dioxin- and dioxin-like compounds, lead, mercury, pesticides) (http://www.who.int/ipcs/assessment/public_health/chemicals_phc/en/). Emerging data suggests that at least two others (cadmium, fluoride) might do so as well. The goal of this chapter is to describe, in brief, how exposure to an environmental chemical during a critical stage of brain development process might increase a child's risk of an intellectual disability or other developmental disorders. Many of the examples provided involve lead because the body of data available for this chemical is much more extensive than that for any other. Principles derived from the study of lead will generally apply to other chemicals, however.

One reason for the enhanced vulnerability of the developing CNS to exogenous insults is the sheer complexity of the processes it involves, which occur over a protracted period beginning shortly after conception and lasting for at least two decades. Brain development is characterized by an exquisite temporal and spatial choreography of processes that must unfold properly in order to produce an organ consisting of billions of precisely located, highly interconnected, specialized cells. Of course, many factors other than environmental chemicals can interfere with these developmental processes during the prenatal and postnatal years. Examples of such pathogens include viruses (e.g., zika, cytomegalovirus, rubella), bacteria (e.g., syphilis, Neisseria meningitidis), and protozoa (e.g., Toxoplasma gondii, schistosoma).

In the sixteenth century, Paracelsus published what remains the most fundamental dictum of toxicology: "Solely the dose determines that a thing is not a poison." In other words, any chemical can become toxic to biological systems. Because timing is critically important for proper brain development, implying the existence of "critical windows of vulnerability," it is evident that this dictum must be amended to indicate that in addition to dose, the stage of brain development at the time of exposure is an important consideration in determining whether exposure to a chemical is neurotoxic. Nevertheless, Paracelsus' contribution provided the foundation for key toxicological concepts, including as the dose-response relationship and threshold dose. Thus,

D. C. Bellinger (✉)
Boston Children's Hospital, Harvard Medical School,
Harvard T.H. Chan School of Public Health,
Boston, MA, USA
e-mail: david.bellinger@childrens.harvard.edu

J. L. Matson (ed.), *Handbook of Intellectual Disabilities*, Autism and Child Psychopathology
Series, https://doi.org/10.1007/978-3-030-20843-1_20

in evaluating the extent to which exposure to an environmental chemical poses a public health problem, it is critical to determine whether the doses incurred by members of the general population at critical times in brain development exceed the doses known to cause various forms of harm. The following section briefly describes brain development and identifies chemicals which can have deleterious impact on different aspects of the processes involved.

During prenatal life, the key stages of brain development are primary neurulation (weeks 3–4), development of the forebrain (prosencephalon) (months 2–3), neuronal proliferation (months 3–4), neuronal migration (months 3–5), and neuronal organization (later gestation and continuing postnatally). Myelination begins in mid-pregnancy and continues into young adulthood. The coordination of these complex processes is regulated by myriad signaling pathways that must work properly if an individual is to end up with a species-typical brain. Although minor, inconsequential variations occur among individuals in the fidelity of these processes, some variations can result in abnormalities that have substantial impact on an individual's abilities to carry out brain-based functions. Which aspects of CNS development a chemical perturbs and thus, when exposure is likely to be most deleterious, depends in part on its mechanism of action. Many chemicals affect multiple aspects of brain development. Alcohol, methylmercury, and chlorpyrifos (a pesticide) disturb neural cell proliferation, while methylmercury, alcohol, and X-irradiation affect neural cell migration. Differentiation of neuroblasts is affected by alcohol, nicotine, methylmercury, and lead. The creation of glial cells and subsequent myelination of neurons is affected by endocrine disrupting chemicals, alcohol, and lead. Synaptogenesis is affected by alcohol, polychlorinated biphenyls, triethyltin, and the pesticides parathion and permethrin, while apoptosis (the orderly process of programmed cell death) is affected by lead, alcohol, and methylmercury. Many chemicals affect neurotransmitter systems, including organophosphate pesticides, alcohol, lead, methylmercury, aluminum, and pharmaceutical agents such as some anti-depressants (e.g., selective serotonin reuptake inhibitors).

One reason for the enhanced vulnerability of developing CNS is that the placenta and blood-brain barrier do not fully protect the fetus from exposure to all chemicals. Some, such as lead, passively diffuse across the placenta so that the concentration in fetal blood is approximately the same as in maternal blood (Aylward et al., 2014). In the case of methylmercury, the concentration in fetal blood is approximately 70% greater than in maternal blood (Stern & Smith, 2003), suggesting the possibility that it is actively transported across the placenta. In a nationally representative sample of pregnant U.S. women some chemicals, including polychlorinated biphenyls, organochlorine pesticides, perfluorinated chemicals, phenols (e.g., bisphenol A), polybrominated diphenyl ethers (PBDEs, which are flame retardants), phthalates (plasticizers), polycyclic aromatic hydrocarbons (products of the combustion of organic materials), and perchlorate, were detected in 99–100% of pregnant women (Woodruff, Zota, & Schwartz, 2011).

Moreover, the toxicokinetics of chemicals in the maternal-fetal unit can be complex. For example, in addition to being exposed to whatever lead a woman is exposed to during her pregnancy, the fetus is also exposed to lead to which the woman was exposed in the past. Approximately 90% of the lead in an adult's body is stored in bones. To support the development of a fetus' skeleton, large quantities of calcium are mobilized from maternal bone during pregnancy, especially the third trimester. Lead and calcium share certain key chemical properties, viz., both are divalent cations (valence of +2). As a result, lead is mobilized from maternal bone by the same processes that mobilize calcium. In fact, lead that reflects maternal exposure that pre-dates her pregnancy accounts for a substantial fraction of the lead in a fetus's cord blood (Gulson, Mizon, Korsch, Palmer, & Donnelly, 2003). Cadmium does not cross the placenta, but it can accumulate in it and impair its critical support functions.

In adults, the blood-brain barrier, which consists of tight junctions between the endothelial cells lining cerebral microvessels, prevents larger water-soluble chemicals from entering the brain (Zheng, Aschner, & Ghersi-Egea, 2003). This barrier is not fully developed at birth, however, and studies in nonhuman primates using radioactive tracers demonstrate that many chemicals pass more readily from the circulating blood into the brain in immature animals than in adult animals.

Certain behavioral and physiologic factors also place a developing child at greater risk than an adult to the deleterious effects of chemical exposures (Selevan, Kimmel, & Mendola, 2000). First, certain pathways of exposure are unique to children, including placental transfer and breastfeeding. It is primarily fat-soluble chemicals (e.g., dioxins, PCBs, perfluorinated compounds) that are of concern with regard to passage into breast milk. Certain behaviors that are more common in children than adults bring them into more intimate contact than adults with some chemicals. These include hand-to-mouth activity, oral exploration of objects, and non-nutritive ingestion (i.e., pica). Such behaviors can result in greater exposure to chemicals, such as lead and PBDEs, that might be present in household dust and soil. On a body weight basis, children consume more food and breathe a greater volume of air than do adults, so they tend to experience greater exposures than adults to foodborne and airborne hazards. Children and adults also experience different breathing zones, with children spend more time near the floor, where chemical concentrations in the air might be greater (e.g., after residential pesticide application). Differences between the diets of children and adults can also be important. Because children's relative consumption of fruit juices is typically greater than that of adults, the presence of pesticide residues on these products can pose a greater risk to them than to adults. Certain micronutrient deficiencies, such as iron and calcium, are more common in children and can result in greater fractional absorption of chemicals with which these essential metals share binding sites in the gut. For example, children absorb up to 50% of ingested lead, whereas adults absorb approximately 10%. Finally, some liver detoxification pathways are not fully developed in the early postnatal years. For example, the enzymes that convert lipid-soluble compounds into water-soluble metabolites that can be excreted in the urine are less effective in children, with the result that parent compounds that can damage cellular processes remain present for a longer period following exposure.

Although acute poisonings as a result of a single, high-dose exposure to a chemical can be serious and even fatal, such events are, fortunately, fairly rare. A broader concern is the potential neurological impact of children's chronic, low-dose exposure to chemicals that, in some cases, are nearly ubiquitous in the environment, affecting large numbers of children. The specific concern is that although such exposures might not produce clinical signs of intoxication, they nevertheless adversely impact brain function, increasing a child's risk of reduced intellectual capacities. The following sections discuss in more detail what is known about the effects of particular chemicals or classes of chemicals on children's intellectual development.

Metals

Mercury. Mercury exists in three forms: elemental, inorganic, and organic. The latter forms are considered to be the most important from the standpoint of public health. Ethylmercury was long used as a preservative (as a component of thimerosal) in multi-use vials of vaccines, but its use has largely been discontinued in pediatric formulations because concerns were raised around 2000 about the potential toxicity of the cumulative doses young children received. The following discussion focuses largely on methylmercury.

In the general population, consumption of seafood is the major pathway of exposure to methylmercury. Inorganic mercury that is dispersed into the environment by both natural (e.g., volcanoes, forest fires) and industrial processes (e.g., combustion of fossil fuels) settles into waterbodies where it is bio-transformed (specifically methylated to form methylmercury) by microbes in the sediments. It enters the aquatic food chain and bio-concentrates in tissues as it ascends trophic levels. The concentrations of methylmercury are therefore greatest in long-lived predatory fish (e.g., whales, shark, swordfish, albacore tuna).

Methylmercury provides a striking example of age-dependence of vulnerability to neurotoxicity. Industrial discharge of mercury salts into Minamata Bay in Japan resulted in heavy contamination of the seafood in the region. Children born to women who, during pregnancy, consumed large amounts of this seafood suffered a distinctive constellation of neurologic signs that came to be called, "Congenital Minamata Disease" (CMD) (Harada, 1995). They included growth disturbances, retention of primitive reflexes, sensory impairments, intellectual disability (tenfold increase in risk), cerebral palsy (50-fold increase in risk), and movement and coordination disorders (e.g., cerebellar ataxia, chorea, athetosis, dysarthria). A special hospital was built in Minamata City solely to provide lifelong care for these children, who were unable to function independently. Many of the mothers of children with CMD manifested no symptoms of mercury intoxication or only mild sensory symptoms such as transient parathesias. Neuropathological examination of individuals who were of different ages when their mercury exposure began revealed strikingly different patterns of brain abnormalities (Choi, 1989). In individuals who had already reached adulthood at the onset of exposure, the lesions were highly localized, clustering in the pre- and post-central gyri, the calcarine fissure of the occipital cortex, and the cerebellum. This is consistent with the clinical signs of mercury intoxication in adults, which include movement disorders, tremors, sensory disturbances, and constriction of the visual fields. In individuals exposed throughout gestation, however, lesions were found throughout the brain, with no apparent localization. This is consistent with the global developmental impairments characteristic of patients with CMD. One reason the impacts are diffuse rather than focal in fetuses is that exposure to methylmercury arrests mitotic cells in metaphase, impairing the cytoskeletal proteins (microtubule assemblies) that form the mitotic spindle. As a result, cell proliferation and migration are perturbed, producing widespread abnormalities including heterotopias (islands of cells in the wrong location), reduced cell densities, anomalous cytoarchitecture, disturbance in laminar pattern of cerebral cortex, incomplete myelination, glial proliferation, and limited gyral differentiation.

The devastating neurological effects observed in poisoning episodes such as occurred in Minamata and elsewhere stimulated concern that low-level chronic prenatal exposure to methylmercury has less serious but still deleterious effects on children's brain development. Therefore, numerous studies have been conducted in cohorts from regions in which seafood is an important component of the diet. A large prospective study of children from the Faroe Islands (North Atlantic Ocean) showed that the performance of children on tests of language, attention, and memory, was inversely related to their mothers' mercury exposure during pregnancy (Grandjean et al., 1997). In this cohort, each increase of 1 μg/g (part per million) in the maternal hair-mercury was associated with, on average, a loss of about one-half of an IQ point (Bellanger et al., 2013). Neuropsychological deficits were found at follow-up evaluations of the children at 14 (Debes, Budtz-Jørgensen, Weihe, White, & Grandjean, 2006) and 22 years of age (Debes, Weihe, & Grandjean, 2016), although they were diminished in magnitude. Functional MRI studies of a subset of the cohort showed dose-related alterations in patterns of activation (White et al., 2011). Studies conducted in areas in which local seafood is heavily contaminated by the use of mercury as an amalgamator in artisanal gold mining have found substantial intellectual deficits in the children with the greatest prenatal exposure to methylmercury (Gibb & O'Leary, 2014).

Some large studies of fish-consuming populations have not found significant associations between children's prenatal methylmercury exposure and their later intellectual development (e.g., Davidson et al., 1998). One potential explanation has to do with the pathway or vehicle of exposure. Seafood is a source not only of methylmercury but also a variety of important macronutrients (e.g., protein) and micronutrients (e.g., long-chain polyunsaturated fatty acids such as omega-3, selenium, iron, choline) that promote brain development. Failure to structure statistical analyses in such a way that adjustments are made for the fact that increased intake of these nutrients often accompanies increased intake of methylmercury can obscure the detection of both the toxicity of methylmercury and the benefits of the nutrients, i.e., produce negative confounding (Choi, Cordier, Weihe, & Grandjean, 2008). Analyses that address this issue have shown that by careful selection of the particular species of fish that are consumed, it is possible both to achieve the benefits of these nutrients and minimize exposure to methylmercury, improving the cognitive outcomes of children (e.g., Oken et al., 2005).

Bellinger, O'Leary, Rainis, and Gibb (2016) estimated that the global incidence of methylmercury-associated intellectual disability is approximately 225,000 cases per year. Based on country-specific data on exposure, the incidence rate varied more than 20-fold across different regions of the world. The highest rate of mercury-associated ID was in the low-income region of the Americas (7 cases per 10,000 population) (subregion D in the WHO classification, which includes Bolivia, Ecuador, Guatemala, Haiti, Nicaragua, and Peru). The largest number of cases, however, was estimated to be in countries in the highly populated Western Pacific B region (87,445 cases). This includes all countries in the region except Australia, Brunei Darussalam, Japan, New Zealand, and Singapore. As would be expected, island nations and those in which artisanal gold mining is common tended to have the highest incidence rates, most likely due to the prominence of seafood in the diet. Nearly all of the ID cases associated with methylmercury would be expected to be mild in severity.

Mercury in its elemental form has long been used in dentistry, as dental amalgam, which is used to restore caries, is 50% elemental mercury by weight. These amalgams can release mercury, and individuals with a larger number of such fillings have been shown to have somewhat higher urinary mercury concentrations. The question of whether the concentrations are sufficient to cause neurotoxicity has been controversial. Although there have been numerous case reports and observational studies suggesting that it is, two large trials comparing the neuropsychological and renal outcomes of children with substantial unmet dental needs who were randomized to receive either dental amalgam or composite resin restorations of caries, did not find evidence that children in the composite resin group performed better than the children in the amalgam group in the 5–7 year follow-up period (Bellinger et al., 2006; Lauterbach et al., 2008).

Lead. Lead can produce devastating effects on the developing brain by interfering with myriad processes. Depending on the dose, it increases apoptosis (programmed cell death), excitotoxicity (neuronal damage caused by over-activation of receptors), reduces cellular energy metabolism by impairing mitochondrial function, reduces heme synthesis and the oxygen-carrying capacity of red blood cells, increases oxidative stress and lipid peroxidation thus damaging cell membrane lipids, alters the activity of first and second messenger systems in neurons, receptor densities, and dendritic branching patterns, impairs the development and function of oligodendroglia resulting in abnormal myelin formation, disturbs neurotrophic processes including thyroid transport into brain, and alters the regulation of gene transcription. Severe lead poisoning can cause brain hemorrhage, edema, seizures, and coma. Even children whose lead poisoning is not so severe as to cause such an encephalopathy are left with a variety of residual difficulties. In an early case series, Byers and Lord (1943) demonstrated the error of the view, widespread at the time, that children fully recover from sub-encephalopathic lead poisoning. They observed that, "after recovery from their lead poisoning, these…children made an extremely poor record in competition with their fellows," insofar as, "with one definite and a second possible exception, none of the 20 children succeeded in school" (p.479). They also noted that the children exhibited severe behavioral pathologies, including hyperactivity, reduced impulse control, and aggression (e.g., fire-setting, biting others, stealing supplies, repeatedly dancing on desks, attacking classmates). Other problem behaviors noted included cruelty to animals and lack of response to punishment.

It is now recognized that because lead is so ubiquitous in the environment, intellectual impairment due to excessive exposure is a frequent rather than a rare occurrence. Indeed, some lead can be detected in the blood of virtually everyone. Based on 2015 data, the Institute for Health Metrics and Evaluation estimated that lead exposure accounts for 12.4% of the cases of idiopathic intellectual disability in the world, concentrating in low- and middle-income countries.

Based on the substantial amount of evidence that accrued on lead neurotoxicity over the past few decades, the blood lead concentration considered to be the "upper limit of normal" by bodies such as the World Health Organization and the U.S. Centers for Disease Control and Prevention (CDC) steadily dropped. In the 1960s a value of 60 μg/dL was considered the limit but, at present, because of the apparent absence of a threshold for the appearance of adverse effects on children's cognition and behavior, no level is considered to be "safe." A set of analyses of IQ data collected in seven prospective studies (N = 1333) conducted in several countries provided much of the justification for the present consensus (Lanphear et al., 2005). The motivation for pooling the data from these studies was to increase the precision with which the shape of the dose-effect relationship could be ascertained for blood lead concentrations <10 μg/dL. At the time of these analyses, 10 μg/dL was the U.S. CDC "action" level, at which intervention activities were triggered to reduce a child's lead exposure. The relationships between IQ, measured at age 5–10 years, and various indices of lead exposure were evaluated. A variety of statistical models were compared in terms of goodness-of-fit to the data, adjusting for 10 covariates, such as maternal IQ and home quality. A log-linear model using concurrent blood lead concentration as the exposure biomarker provided the best fit and indicated that an increase in blood lead from 2.4 to 30 μg/dL (the fifth and 95th percentiles of the blood lead distribution in the pooled dataset) was associated with an IQ reduction of 6.9 points. One of the most important findings was that the association was nonlinear, insofar as much of the total reduction, 3.9 points, occurred over the range of 2.4 to 10 μg/dL. The increase from 10 to 20 μg/dL was associated with a further reduction of 1.9 points, and the increase from 20 to 30 μg/dL with an additional reduction of 1.1 points. Although the mechanism(s) by which the proportional loss in IQ are greater at lower than at higher blood lead concentrations remain unknown, similar supra-linear relationships between blood lead and other cognitive outcomes were subsequently reported (e.g., Kordas et al., 2006; Téllez-Rojo et al., 2006).

Epidemiological studies have generally included assessment of other aspects of children's neuropsychological functioning. IQ is an apical measure, integrating children's performance in diverse verbal and nonverbal domains. If the effects of a neurotoxicant are focal, limited to only certain domains, one would expect that tests that focus more narrowly on the vulnerable domains would be more strongly related to lead biomarkers than is IQ. It is somewhat surprising, therefore, that, IQ is more consistently associated with lead biomarkers than is performance on tests that focus on more narrowly defined domains (e.g., memory, language). While this might, in part, be attributable to psychometric differences in the tests used, it might also provide some insight into the mechanism of lead-associated neurotoxicity, suggesting diffuse rather than focal neuronal and/or white matter injury, thereby producing global impairments of higher cortical functioning. It is also possible that the domains most affected vary across studies because of the joint effect of cohort differences in the timing of lead exposure and differences across domains in the timing of greatest susceptibility. The domains vulnerable to lead exposure also depend, to some extent, on cohort characteristics or context specific aspects such as genetic susceptibility, social environment, mixed chemical exposures.

Many studies have reported significant inverse associations, in the general population of children, between lead exposure and success in school, expressed as lower scores on standardized tests, receipt of special education services, grade retention, and failure to complete qualifications (e.g., Amato et al., 2012; Delgado et al., 2017; Fergusson, Horwood, & Lynskey, 1997; Magzamen et al., 2013, 2015; Needleman, Schell, Bellinger, Leviton, & Allred, 1990). In ecologic (i.e., aggregate-level) analyses, Nevin (2009) reported an inverse relationship between preschool blood lead concentrations and SAT scores, lagged by 17 years. Evens et al. (2015) evaluated the relationship between blood lead concentration and performance on the Illinois Standard Achievement Test in a cohort of 47,168 Chicago schoolchildren. Adjusting for covariates, they found dose-dependent reductions, extending below 10 μg/dL, in both reading and math scores, with each 5 μg/dL increase in blood lead associated with an increase of 1.3 in the risk of reading and math scores defined as "failure." A nonlinear dose-response relationship was found for reading failure, with the slope steeper for blood lead concentrations less than 10 μg/dL compared to greater than 10.

In a similar study, Miranda, Kim, Reiter, Galeano, and Maxson (2009) combined a North Carolina blood lead surveillance database with a database containing scores on a reading test given to school children in the state at the end of fourth grade. In the sample of 57,678 children, blood lead concentration ranged from 1 to 16 μg/dL, with a 75th percentile of 6 μg/dL. A significant dose-response effect relationship was found between blood lead and test score, without apparent threshold. Quantile regression analyses indicated that the adverse impact was more pronounced at the lower end of the test score distribution than at the higher end, indicating that the impact of lead is disproportionally greater on children who are already at academic risk than on children who are at low academic risk. This might be attributable to contextual factors that influence a child's resilience or susceptibility to lead exposure, such as nutrition, social environment, and other chemical exposures.

Skerfving, Lofmark, Lundh, Mikoczy, and Stromberg (2015) evaluated the performance of 3176 Swedish children on an examination taken at the end of compulsory schooling (age 16 years) in relation to blood lead concentration measured when the children were in primary school (age 7–12 years). They found an inverse nonlinear association between blood lead concentration (range 0.6 to 16.2 μg/dL, 90th percentile 6 μg/dL) and school performance, with a steeper slope at concentrations below 5 μg/dL than above 5. They estimated that the adverse impact of an increase in blood lead concentration from 2.5 to 5 μg/dL was similar in magnitude to the impact of having a mother with a university versus a primary school education. Surkan et al. (2007) reported that children with a blood lead concentration of 5–10 μg/dL, compared to children with a concentration of 1–2 μg/dL, had significantly lower reading and mathematics scores, even after adjustment for Full-Scale IQ score. This suggests that the achievements of children with greater exposures were not at a level commensurate with their ability.

The effects of lead are persistent as children age, with greater childhood exposure associated with reduced success in life success. In the large Dunedin Multidisciplinary Health and Development Study, Reuben et al. (2017) found that blood lead concentration measured at 11 years of age was inversely related to IQ at age 38 years, even after adjustment for IQ at age 11 years. Furthermore, early lead exposure was inversely related to socioeconomic status at age 38, suggesting that children who were more highly exposed enjoyed lower upward social mobility and, in fact, failed to match the socioeconomic achievements of their parents.

One aspect of lead neurotoxicity that receives little attention is the possible role of early-life exposure to lead as, itself, an effect modifier that increases the adverse impacts of later events and exposures or even normal aging processes. Weiss, Clarkson, and Simon (2002) speculated that if early-life exposure to a neurotoxicant increases the annual rate of neuronal loss by less than 1%, clinical signs of neurodegeneration would appear several years earlier than they would in the absence of such an exposure. Animal studies suggest that lead exposure reduces the ability of rats to weather an unrelated neurological insult in adulthood. Rats exposed to lead early in development but not thereafter were less successful than control rats in recovering function (beam walking, limb placing) after a laser-induced stroke in the somatosensory cortex in adulthood (Schneider & DeKamp, 2007). Early exposure to lead might also influence function in adulthood by altering the speed of aging. Provocative studies in rodents and nonhuman primates suggest that exposure in infancy initiates epigenetic processes, perhaps involving altered patterns of DNA methylation, that result in adult-onset overexpression of proteins involved in neurodegenerative processes characteristic of Alzheimer's Disease (viz., increased deposition of β-amyloid, increased hyper-phosphorylation of tau protein) (Gąssowska et al., 2016).

In the 1970s, the observations of Byers and Lord (1943) regarding the behavioral pathologies were followed up, with case-control and chelation challenge studies suggesting that children diagnosed with hyperactivity had greater lead burdens (e.g., David, Clark, & Voeller, 1972). Subsequent studies indicated that even in children who were neither clinically lead poisoned nor diagnosed with hyperactivity, a greater lead burden was associated with increased distractibility, reduced ability to follow directions, disorganization, daydreaming, and lack of task persistence (e.g., Needleman et al., 1979). A meta-analysis of 33 studies conducted between 1972 and 2010, involving more than 10,000 children, found significant effect sizes linking greater exposure and dimensional measures of both inattentive and hyperactive/impulsive symptoms (Goodlad, Marcus, & Fulton, 2013).

It is now established that increased childhood lead exposure also increased the risk that a child meets diagnostic criteria for ADHD. Using data from NHANES 1999–2002, Braun, Kahn, Froehlich, Auinger, and Lanphear (2006) found that the adjusted odds ratio for parent-reported ADHD among 6–16-year old children with a blood lead concentration in the fifth quintile (>2 μg/dL) was 4.1, compared to children in the first quintile (<0.8 μg/dL). A dose-response relationship was observed, with adjusted odds ratios of 1.1, 2.1, and 2.7 for children in the second quintile, third, and fourth quintiles, respectively. Among the limitations of this study are the absence of a clinician-confirmed diagnosis of ADHD and the fact that data on important covariates, such as family history of ADHD, were not available. A study by Froehlich et al. (2009), using NHANES 2001–2004 data, addressed the first issue. The Diagnostic Interview Schedule for Children, a clinician-administered diagnostic interview based on DSM-IV, was used to confirm a diagnosis of ADHD. Children with a blood lead concentration in the upper tertile (>1.3 μg/dL) were 2.3 times more likely to meet diagnostic criteria than were children with a concentration in the lowest tertile.

This association has been replicated in several subsequent case-control studies (Choi, Kwon, Lim, Lim, & Ha, 2016; Park et al., 2016; Wang et al., 2008). Nigg et al. (2008) clarified possible behavioral mechanisms of the association between lead and ADHD symptoms. Children 8–17 years old who met rigorous criteria for the diagnosis of ADHD had a significantly higher blood lead concentration than controls, even though concentration for all participants ranged only from 0.4 to 3.5 μg/dL, mean of 1.03). A significant relationship was found between blood lead concentration and total ADHD symptoms. In this study, IQ was measured and a Stop task was administered, providing assessments of a child's ability to suppress a prepared response (stop signal reaction time) and variability of reaction time on the "go" trials (response variability, readiness, and control). Mediation analyses suggested that lead exposure might increase a child's risk of ADHD by impairing cognitive control abilities, and that the association between blood lead and IQ was mediated by the association between blood lead and hyperactivity-impulsivity, not vice versa. Nigg et al. (2008) argued that the plausibility of the link between lead and ADHD is supported by the evidence that lead disrupts midbrain dopamine circuitry (striatum and fronto-striatal networks), the same circuitry that is thought to underlie ADHD. Nigg, Nikolas, Knottnerus, Cavanagh, and Friderici (2010) replicated the associations between blood lead concentration and ADHD symptoms in a larger sample, including adjustment for additional covariates, and at even lower blood lead concentrations (range 0.3–2.2 μg/dL).

Several studies in the last decade have suggested that children with greater early-life lead exposure are at increased risk of social pathologies such as criminal activities (e.g., Boutwell et al., 2017; Wright et al., 2008). If this association is true, it seems most likely to reflect a developmental cascade, representing a possible end-state for individuals with reduced intelligence, reduced school success, behavioral impairments such as reduced impulse control and ADHD, executive function deficits such as inability to anticipate consequences or to implement long-term plans, and possible substance abuse (Bellinger, Matthews-Bellinger, & Kordas, 2016). Of course, not all children with excessive early-life lead exposure follow this developmental trajectory, as there are many points along the way at which timely developmental supports might reduce the likelihood that the cascade is fully expressed.

Neuroimaging studies have explored the associations between lead exposure history and brain structure in individuals from the general population. Most of the data are from the Cincinnati Prospective Lead Study in which participants were enrolled prenatally and followed into young adulthood (19–24 years of age). Volumetric imaging revealed significant inverse associations between annual mean blood lead concentration between 3 and 6 years of age and gray matter volume, particularly in the frontal regions of the brain, including the anterior cingulate cortex and the ventrolateral prefrontal cortex, areas usually considered to be related to executive functions, mood regulation, and decision-making (Cecil et al., 2008). On diffusion tensor imaging, reduced fractional anisotropy and axial diffusivity were also associated with greater childhood lead exposure (Brubaker et al., 2009). These findings suggest impaired myelination and reduced axonal integrity in regions that regulate executive functions.

Brain function in adulthood is also inversely related to childhood lead exposure. Proton magnetic resonance spectroscopy studies showed that greater childhood blood lead concentration was associated with reductions in several metabolites, including *N*-acetyl aspartate and creatine and phosphocreatine, in both gray and white matter (Cecil et al., 2011). Functional magnetic resonance (fMRI) imaging showed that during a verb generation task, individuals with greater blood lead concentrations in childhood showed dose-dependent changes in activation pattern in the left frontal cortex and the left middle temporal gyrus (Yuan et al., 2006). In another cohort, individuals with greater lead exposure showed fMRI showed reduced activation in the dorsolateral prefrontal cortex, ventrolateral prefrontal cortex, pre-supplementary motor areas, and inferior parietal cortex on the Wisconsin Card Sorting Test and the n-back task (particularly on trials that imposed the greatest memory load) (Seo et al., 2015). These findings suggest that exposure to lead impairs the fronto-parietal working memory network.

Arsenic

Although arsenic is well known for its acute toxicity, chronically elevated exposure has many effects that would be expected to affect brain function. These effects include increasing oxidative stress, free radicals, and neuronal apoptosis, impaired neurogenesis in the hippocampus and disruption of the expression of the NMDA receptor, disruption of the hypothalamo-pituitary-adrenal axis, reductions in neurotransmitter levels, interference with the expression of thyroid hormone receptor genes, and others (Bellinger, 2013).

The potential consequences of clinical arsenic poisoning became evident from an episode that occurred in Japan in 1955, when milk powder produced by a manufacturer was contaminated with arsenic. Infants who consume formula made from this powder could have had an arsenic intake of as much as 5 milligrams per day, and signs of poisoning appeared after a total intake of approximately 60 milligrams. One-quarter of the infants residing in the prefectures of western Japan were affected. As many as 130 children died, and their brains showed signs of edema, hemorrhage, and white matter degeneration. Those who survived were at an increased risk of epilepsy, severe intellectual disability, and hearing deficit (Dakeishi, Murata, & Grandjean, 2006).

Diverse neuropsychological deficits were still apparent 50 years later, even among those not previously recognized as having any disability (Yorifuji et al., 2016).

For a large number of people in many areas of the world, arsenic naturally occurs at relatively high concentrations in the water used for cooking and drinking. Studies have been conducted in several areas of South Asia (e.g., Bangladesh, West Bengal, Taiwan) to determine whether chronic exposure to elevated, but not acutely toxic, concentrations of arsenic are associated with reduced intellectual capacities of children.

Fluoride

Fluoride differs from most other environmental chemicals in that, in many areas, public health policies insure that most children are exposed to it for its assumed benefit in reducing dental caries. It is added to public water supplies and put in toothpastes and supplements. Because of its addition to water, fluoride also is present in various dietary elements, such as fruit juices, which are commonly consumed by children. A large number of basic neuroscience studies raise concern about the potential effects of excess exposure to developing animals. In some areas of world, however, the concentration of fluoride in groundwater is naturally high enough to cause observable harm (fluorosis). Depending on the dose, the signs range from small white opaque spots on the enamel of teeth to severe pitting and discoloration of teeth, as well as to skeletal fluorosis. According to the CDC, in 1999–2004, nearly 40% of U.S. children 12–15 years of age show signs of at least mild fluorosis, nearly a 100% increase over the rate observed in 1986–1987 (Beltran-Aguilar, Barker, & Dye, 2010). A review of nearly three dozen studies conducted in China, mostly ecologic in design and comparing children from a low-exposure village to a high-exposure village, concluded that they provide suggestive evidence of modest fluoride-associated reductions in children's IQ scores (Choi, Sun, Zhang, & Grandjean, 2012). For several reasons, however, such studies provide only weak evidence. The most important are the fact that only data on external exposure are available (i.e., water fluoride concentration) rather than data on internal exposures (i.e., blood concentrations of fluoride in individual participants or severity of dental fluorosis), and the likelihood that children resident in the villages being compared differ not only in the fluoride concentration in their water supplies, but in also in terms of other factors that might affect the distributions of their IQ scores (e.g., socioeconomic status, access to medical care, quality of schools, etc.). Recently, a few studies that address at least some of these limitations have been conducted. Using dental fluorosis as the metric of exposure in a relatively small pilot study in Sichuan, China, Choi et al. (2015) found negative associations between fluorosis severity, reflecting lifetime exposure, and children's scores on some neuropsychological tests, Khan et al. (2015) found inverse associations between fluorosis severity and IQ scores in Indian children, and Bashash et al. (2017) found inverse associations between children's prenatal fluoride exposure (concentration in maternal urine during pregnancy) and their IQ scores at ages 4 and 6-to-12 years.

Pesticides

All pesticides are neurotoxic by design insofar as they act by targeting the functioning of the insect nervous system. Because the central nervous systems of insects and mammals operate on many of the same principles, the toxicity of these chemicals tends not to be species-selective. There are many different classes of pesticides, differing in their modes of action and in their toxicities. Organochlorine pesticides (e.g., DDT) were developed in the first half of the twentieth century. They are fat soluble, accumulate in the food chain, and persist in the environment for long periods. They act by altering the electrophysiological properties of cell membranes (particularly axons), disturbing sodium and potassium ion exchange. Because of their toxicity and persistence in the environment, their use has largely been banned or restricted in recent decades although their use continues in certain regions of the world.

Pesticides that degrade more rapidly, and therefore have shorter residence times than organochlorines in the environment, were introduced in the mid-twentieth century (e.g., organophosphates, carbamates). The organophosphates were originally developed as nerve gas agents in Germany during WWII. They are generally considered to be less toxic than organochlorines and are widely used on food crops, in homes, parks, schools, and golf courses. Organophosphate pesticides inhibit the activity of acetylcholinesterase, an enzyme that catalyzes the breakdown of the neurotransmitter acetylcholine. However, it is now known that certain OPs have adverse impacts on children's neurodevelopment at doses that do not cause acetylcholinesterase inhibition. They are thought to work by a different mechanism, which might include induction of inflammation, interference with C-reactive protein receptor signaling, insulin resistance, or nuclear transcription factor functioning. Another major class of pesticides, the pyrethroids, were developed in the 1970s. The neonicotinoids were introduced in the 1980s, although controversy quickly arose because they were implicated in colony collapse disorder, involving a massive die-off of workers in a honey bee colony with serious implications for the pollination of food crops.

In the last decades, a substantial number of studies, conducted in both cohorts presumed to have greater pesticide exposures due to the location of their residence and in general population cohorts, have reported that pesticide body burden is inversely related to children's IQ scores. In a group of children living in the agricultural Salinas Valley of California, Bouchard et al. (2011) showed that offspring of mothers with a concentration of the dialkyl phosphate metabolites of OP greater than 50 nmol/L during pregnancy had lower IQ scores at age 7 years than the children of mothers with concentrations less than 50 nmol/L, which corresponds to the mean in U.S. women of reproductive age. In an urban New York City cohort, Rauh et al. (2011) found an inverse association between the concentration of the OP pesticide chlorpyrifos in umbilical cord blood plasma and child IQ and working memory at age 7 years. In this same cohort, morphometric MRI analysis showed dose-related perturbations in the volumes of many regions of the brain (Rauh et al., 2012). Recently, Eskenazi et al. (2018) reported inverse relationships in a rural, agricultural cohort between prenatal exposure to pyrethroid pesticides and children's social-emotional scores at age 1 year and language/expressive communication scores at age 2 years.

Air Pollution

A rapidly developing body of literature on air pollutants indicates that greater exposures are associated with subclinical impacts on children's cognition (Clifford, Lang, Chen, Anstey, & Seaton, 2016). Air pollution is a complex mixture of diverse chemicals, and the composition can vary by site. Components of the mixture include polycyclic aromatic hydrocarbons (which are produced by the incomplete combustion of organic matter), oxides of sulfur, nitrogen, and carbon, ozone, and metals. In the United States, the concentrations of particulate matter have steadily declined in recent years, although the concentration of ozone has increased (www.lung.org/our-initiatives/healthy-air/sota/key-findings). Small particles (smaller than 2.5 micrometers in diameter) are generally considered to be the most hazardous because they can be inhaled and deposited deep in the lung, reaching terminal bronchioles and alveoli.

Approximately 95% of the world's population live in areas where outdoor fine particulate matter (particles less than 2.5 microns in diameter) concentrations (dust or soot particles) exceed the World Health Organization's Air Quality Guideline of 10 μg/m^3, with most areas of Africa, the Middle East, and South Asia exceeding 35 g/m^3 (www.stateofglobalair.org/air). In addition, more than one-third of the world's population is exposed to potentially hazardous indoor air pollution as a result of the combustion of biomass fuels (e.g., wood, dung, peat, crop, wastes) or coal. Because of the closed spaces and the large amount of time spent indoors, especially during the colder months, the indoor concentrations of particulate matter can be as much as 20-fold greater than outdoor concentrations.

It is only in recent years that the impacts of air pollution on the brain have been investigated, and several potential mechanisms have been identified, including oxidative stress/inflammation (viz., elevation of cytokines and reactive oxygen species), altered levels of dopamine and/or glutamate, and changes in synaptic plasticity/structure (Allen et al., 2017). Studies of children and young adults growing up in Mexico City have reported the emergence of exposure-related signs of neurodegeneration, including early stages of the development of neurofibrillary tangles (hyperphosphorylated tau protein) and neuritic plaques (beta-amyloid deposits), with 1 in 4 individuals showing later stages (Braak stages III-V) neurofibrillary tangles by the fourth decade of life (Calderón-Garcidueñas et al., 2018). They also show other abnormalities, including prefrontal white matter hyperintensities, damage to epithelial and endothelial barriers, tight junction and neural autoantibodies (Calderón-Garcidueñas et al., 2016). Studies of a cohort in Spain showed that, even in the absence of morphological changes in brain, greater airborne exposure to elemental carbon and nitrogen dioxide was associated with lower functional connectivity in key brain networks (e.g., the default mode network) as well as altered activation pattern on a sensory task (Pujol et al., 2016). A prospective study conducted in New York City found that greater prenatal exposure to polycyclic aromatic hydrocarbon (PAH) air pollutants was associated with lower IQ at age 5 (Lovasi et al., 2014) and slower processing speed, attention-deficit/hyperactivity disorder symptoms, and externalizing problems at age 7–9 years (Peterson et al., 2015). Morphometric neuroimaging indicated that these effects were mediated by disruptions of white matter in the left hemisphere. Greater postnatal exposure to PAHs was associated with disruptions of white matter in the dorsal prefrontal regions. Several studies suggest an association between various indicators of air pollution and a child's risk of either a diagnosis of an autism spectrum disorder or elements of its phenotype (Weisskopf, Kioumourtzoglou, & Roberts, 2015).

Synthetic Organic Chemicals

The research literatures on the impacts of exposure to synthetic organic chemicals are not as well-developed as those on the pollutants discussed above. These chemicals include polyhalogenated compounds such as polychlorinated biphenyls (PCBs), and polybrominated diphenyl ethers (PBDEs). PCBs were banned in the US in the 1970s but because of their resistance to degradation, they persist in the environment. PBDEs are used as flame retardants in a wide variety of products. Sharing many of the properties of PCBs, they accumulate and persist for long periods in the environment and in human fat tissue. A systematic review and meta-analysis of studies on children's intelligence and prenatal exposure to PBDEs at levels typical of the general population found a consistent inverse relationship (Lam et al., 2017). A ten-fold increase in PBDE exposure was associated with a decrement of nearly 4 IQ points.

Perfluorinated compounds (PFCs) are commonly used in a variety of consumer products (e.g., non-stick cookware, stain resistant fabrics, fast food packaging). To date, the evidence pertaining to the neurodevelopmental risks associated with such exposures are mixed (Liew, Goudarzi, & Oulhote, 2018).

Endocrine Disruptors

Concerns have been raised about exposure during development to chemicals that alters the function(s) of the hormonal system, causing adverse effects in an organism or its progeny (Braun, 2017). Such chemicals are called "endocrine disrupting chemicals" (EDCs) and can mimic the effects of endogenous hormones, antagonize the effects of endogenous hormones, disrupt the synthesis and metabolism of endogenous hormones, disrupt the synthesis of hormone receptors, and alter target cell sensitivity. Hormone levels in early development are critical in organizing brain development, and perturbations can have long-lasting effects on hormonal programming. For example, adequate levels of thyroid hormone are critical for various processes of brain development, including cell migration, differentiation, and signaling. Given that intellectual disability is a result of congenital hypothyroidism, and even subclinical reductions in thyroid function during pregnancy are associated with IQ deficits in children (Levie et al., 2018), it is plausible to hypothesize that prenatal exposures to chemicals that affect thyroid hormone levels produce more modest impacts on children's intelligence. Increased concentrations of chemicals such as phthalates are inversely associated with total serum thyroid hormone levels in pregnant women and neonates and thyroid stimulating hormone in neonates (Romano et al., in press). However, evidence that the alterations that occur at levels of phthalate exposure typical in the general population affect intelligence is presently inconsistent (Factor-Litvak et al., 2014; Nakiwala et al., 2018). Several studies have reported that early exposure to EDCs such as phthalates do, however, influence sexually dimorphic behaviors, that is, those that tend to differ between the sexes. For example, prenatal phthalate exposure might reduce masculine play in boys (Swan et al., 2010).

Estimating the Population Impact of Environmental Chemicals and the Burden of Disease

An argument frequently advanced by those skeptical about the importance of environmental chemicals is that their impact on the neurodevelopment of an individual child is modest, failing to reach the level of clinical significance. This argument fails to consider the issue in the context of population health. Effect estimates from epidemiologic data are in essence population average effects and should be interpreted in the context of a population and not at the individual level. Some individuals will be resistant and some will be more sensitive. Moreover, the impact of a factor at the population level depends not only on the effect size but also on the distribution of the factor or, in the case of a dichotomous factor, its incidence or prevalence. In a set of comparative analyses of pediatric disease and events, such as brain tumors, congenital heart disease, traumatic brain injury, iron deficiency, and lead exposure, Bellinger (2012) estimated the total number of IQ points lost among U.S. children younger than 5 years of age associated with each disease or event. The estimate for the loss associated with lead exposure was nearly 23 million IQ points, exceeded only by preterm birth. Among the reasons for this is the absence of a threshold for its inverse relationship with IQ and the fact that virtually every child has a blood lead concentration above the detection limit. As a result, and in contrast to most other diseases and events, every child contributes to the total IQ loss in the population that is associated with lead exposure. In fact, the greatest contribution to the total loss care is contributed by the very large proportion of children with blood lead concentrations at the low end of the distribution (because that's where most children fall). A similar calculation of the total IQ losses among the cohort of young U.S. children from the late 1970s produced the figure of approximately 125 million points, suggesting that the public health interventions implemented to reduce population lead exposure has produced a benefit of about 100 million IQ points. Given that approximately 25 million children fall into this age range, the average IQ benefit has been about 4 points, close to the estimate of 4–5 points reached by Kaufman et al. (2014) for the gain in adult IQ.

As the research reviewed in this chapter indicates, environmental chemicals cause adversities that extend well beyond a reduction in intelligence, affecting an exposed individual's success in many aspects of future life. Although current efforts to estimate the burden of disease associated with environmental chemicals consider only IQ deficit as the sequelae (GBD 2016 Risk Factors Collaborators, 2017), a full accounting of the burden of disease imposed by environmental chemicals must include these downstream impacts that can seriously impair quality of life (Bellinger, 2018).

Conclusion

The current approach to regulating chemicals is to impose restrictions on their use only after it becomes apparent that people who are exposed to them are demonstrably harmed. In effect, this results in large-scale natural experiments on the population. The fact that children are the most vulnerable subgroup of the population makes this approach particularly unconscionable. Unfortunately, even rudimentary toxicological data are available for only a small fraction of the approximately 80,000 chemicals in use, and most of these data pertain to rather crude health endpoints such as death, cancer, and birth defects rather than brain development and function. A more protective, proactive alternative would be to require a manufacturer to provide evidence of a chemical's safety before permission is granted to introduce it into the marketplace. It is our responsibility to future generations to reduce or, when possible, to eliminate the threats that these chemicals pose to their future well-being.

References

Allen, J. L., Oberdorster, G., Morris-Schaffer, K., Wong, C., Klocke, C., Sobolewski, M., … Cory-Slechta, D. A. (2017). Developmental neurotoxicity of inhaled ambient ultrafine particle air pollution: Parallels with neuropathological and behavioral features of autism and other neurodevelopmental disorders. *Neurotoxicology, 59*, 140–154.

Amato, M. S., Moore, C. F., Magzamen, S., Imm, P., Havlena, J. A., Anderson, H. A., & Kanarek, M. S. (2012). Lead exposure and educational proficiency: Moderate lead exposure and educational proficiency on end-of-grade examinations. *Annals of Epidemiology, 22*, 738–743.

Aylward, L. L., Hays, S. M., Kirman, C. R., Marchitti, S. A., Kenneke, J. F., English, C., … Becker, R. A. (2014). Relationships of chemical concentrations in maternal and cord blood: A review of available data. *Journal of Toxicology and Environmental Health B Critical Reviews, 17*, 175–203.

Bashash, M., Thomas, D., Hu, H., Martinez-Mier, E. A., Sanchez, B. N., Basu, N., … Hernández-Avila, M. (2017). Prenatal fluoride exposure and cognitive outcomes in children at 4 and 6-12 years of age in Mexico. *Environmental Health Perspectives, 125*, 097017.

Bellanger, M., Pichery, C., Aerts, D., Berglund, M., Castano, A., Cejchanova, M., … Grandjean, P. (2013). Economic benefits of methylmercury control in Europe: Monetary value of neurotoxicity prevention. *Environmental Health, 12*, 3.

Bellinger, D. C. (2012). A strategy for comparing the contributions of environmental chemicals and other risk factors to neurodevelopment of children. *Environmental Health Perspectives, 120*, 501–507.

Bellinger, D. C. (2013). Inorganic arsenic exposure and children's neurodevelopment: A review of the evidence. *Toxics, 1*, 2–17.

Bellinger, D. C. (2018). Applying methods of the global burden of diseases, injuries, and risk. Factors Study to developmental neurotoxicants: A commentary. *Environmental Health, 17*, 53.

Bellinger, D. C., Matthews-Bellinger, J. A., & Kordas, K. (2016). A developmental perspective on early-life exposure to neurotoxicants. *Environment International, 94*, 103–112.

Bellinger, D. C., O'Leary, K., Rainis, H., & Gibb, H. J. (2016). Country-specific estimates of the incidence of intellectual disability associated with prenatal exposure to methylmercury. *Environmental Research, 147*, 159–163.

Bellinger, D. C., Trachtenberg, F., Barregard, L., Tavares, M., Cernichiari, E., Daniel, D., & McKinlay, S. (2006). Neuropsychological and renal effects of dental amalgam in children: A randomized clinical trial. *Journal of the American Medical Association, 295*, 1775–1783.

Beltran-Aguilar, E. D., Barker, L., & Dye, B. A. (2010). Prevalence and severity of dental fluorosis in the United States, 1999-2004. *National Center for Health Statistics Data Brief, 53*, 1–8. Retrieved from https://www.cdc.gov/nchs/products/databriefs/db53.htm

Bouchard, M. F., Chevrier, J., Harley, K. G., Kogut, K., Vedar, M., Calderon, N., … Eskenazi, B. (2011). Prenatal exposure to organophosphate pesticides and IQ in 7-year-old children. *Environmental Health Perspectives, 119*, 1189–1195.

Boutwell, B. B., Nelson, E. J., Qian, Z., Vaughn, M. G., Wright, J. P., Beaver, K. M., … Rosenfeld, R. (2017). Aggregate-level lead exposure, gun violence, homicide, and rape. *PLoS One, 12*, e0187953.

Braun, J. (2017). Early-life exposure to EDCs: Role in childhood obesity and neurodevelopment. *Nature Reviews Endocrinology, 13*, 161–173.

Braun, J. M., Kahn, R. S., Froehlich, T., Auinger, P., & Lanphear, B. P. (2006). Exposures to environmental toxicants and attention deficit hyperactivity disorder in U.S. Children. *Environmental Health Perspectives, 114*, 1904–1909.

Brubaker, C. J., Schmithorst, V. J., Haynes, E. N., Dietrich, K. N., Egelhoff, J. C., Lindquist, D. M., … Cecil, K. M. (2009). Altered myelination and axonal integrity in adults with childhood lead exposure: A diffusion tensor imaging study. *Neurotoxicology, 30*, 867–875.

Byers, R. K., & Lord, E. E. (1943). Late effects of lead poisoning on mental development. *American Journal of Diseases of Children, 66*, 471–494.

Calderón-Garcidueñas, L., Gónzalez-Maciel, A., Reynoso-Robles, R., Delgado-Chávez, R., Mukherjee, P. S., Kulesza, R. J., … Villarreal-Ríos, R. (2018). Hallmarks of Alzheimer disease are evolving relentlessly in Metropolitan Mexico City infants, children and young adults. APOE4 carriers have higher suicide risk and higher odds of reaching NFT stage V at ≤ 40 years of age. *Environmental Research, 164*, 475–487.

Calderón-Garcidueñas, L., Reynoso-Robles, R., Vargas-Martínez, J., Gómez-Maqueo-Chew, A., Pérez-Guillé, B., Mukherjee, P. S., … Gónzalez-Maciel, A. (2016). Prefrontal white matter pathology in air pollution exposed Mexico City young urbanites and their potential impact on neurovascular unit dysfunction and the development of Alzheimer's disease. *Environmental Research, 146*, 404–417.

Cecil, K. M., Brubaker, C. J., Adler, C. M., Dietrich, K. N., Altaye, M., Egelhoff, J. C., … Lanphear, B. P. (2008). Decreased brain volume in adults with childhood lead exposure. *PLoS Medicine, 5*, e112.

Cecil, K. M., Dietrich, K. N., Altaye, M., Egelhoff, J. C., Lindquist, D. M., Brubaker, C. J., & Lanphear, B. P. (2011). Proton magnetic resonance spectroscopy in adults with childhood lead exposure. *Environmental Health Perspectives, 119*, 403–408.

Choi, A. L., Cordier, S., Weihe, P., & Grandjean, P. (2008). Negative confounding in the evaluation of toxicity: The case of methylmercury in fish and seafood. *Critical Reviews in Toxicology, 38*, 877–893.

Choi, A. L., Sun, G., Zhang, Y., & Grandjean, P. (2012). Developmental fluoride neurotoxicity: A systematic review and meta-analysis. *Environmental Health Perspectives, 120*, 1362–1368.

Choi, A. L., Zhang, Y., Sun, G., Bellinger, D. C., Wang, K., Yang, X. J., … Grandjean, P. (2015). Association of lifetime exposure to fluoride and cognitive functions in Chinese children: A pilot study. *Neurotoxicology and Teratology, 47*, 96–101.

Choi, B. H. (1989). The effects of methylmercury on the developing brain. *Progress in Neurobiology, 32*, 447–470.

Choi, W. J., Kwon, H. J., Lim, M. H., Lim, J. A., & Ha, M. (2016). Blood lead, parental marital status and the risk of attention-deficit/hyperactivity disorder in elementary school children: A longitudinal study. *Psychiatry Research, 236*, 42–46.

Clifford, A., Lang, L., Chen, R., Anstey, K. J., & Seaton, A. (2016). Exposure to air pollution and cognitive functioning across the life course—A systematic literature review. *Environmental Research, 147*, 383–398.

Dakeishi, M., Murata, K., & Grandjean, P. (2006). Long-term consequences of arsenic poisoning during infancy due to contaminated milk powder. *Environmental Health, 5*, 31.

David, O., Clark, J., & Voeller, K. (1972). Lead and hyperactivity. *Lancet, 2*(7783), 900–903.

Davidson, P. W., Myers, G. J., Cox, C., Axtell, C., Shamlaye, C., Sloane-Reeves, J., … Clarkson, T. W. (1998). Effects of prenatal and postnatal methylmercury exposure from fish consumption on neurodevelopment: Outcomes at 66 months of age in the Seychelles Child Development Study. *Journal of the American Medical Association, 280*, 701–707.

Debes, F., Budtz-Jørgensen, E., Weihe, P., White, R. F., & Grandjean, P. (2006). Impact of prenatal methylmercury exposure on neurobehavioral function at age 14 years. *Neurotoxicology and Teratology, 28*, 536–547.

Debes, F., Weihe, P., & Grandjean, P. (2016). Cognitive deficits at age 22 years associated with prenatal exposure to methylmercury. *Cortex, 74*, 358–369.

Delgado, C. F., Ullery, M. A., Jordan, M., Duclos, C., Rajagopalan, S., & Scott, K. (2017). Lead Exposure and developmental disabilities in preschool-aged children. *Journal of Public Health Management Practice, 24*, e10–e17.

Eskenazi, B., An, S., Rauch, S. A., Coker, E. S., Maphula, A., Obida, M., … Chevrier, J. (2018). Prenatal exposure to DDT and pyrethroids for malaria control and child neurodevelopment: The VHEMBE cohort, South Africa. *Environmental Health Perspectives, 126*, 047004.

Evens, A., Hryhorczuk, D., Lanphear, B. P., Rankin, K. M., Lewis, D. A., Forst, L., & Rosenberg, D. (2015). The impact of low-level lead toxicity on school performance among children in the Chicago public schools: A population-based retrospective cohort study. *Environmental Health, 14*, 21.

Factor-Litvak, P., Insel, B., Calafat, A. M., Liu, X., Perera, F., Rauh, V. A., & Whyatt, R. M. (2014). Persistent associations between maternal prenatal exposure to phthalates on child IQ at age 7 years. *PLoS One, 9*, e114003.

Fergusson, D. M., Horwood, L. J., & Lynskey, M. T. (1997). Early dentine lead levels and educational outcomes at 18 years. *Journal of Child Psychology and Psychiatry, 38*, 471–478.

Froehlich, T. E., Lanphear, B. P., Auinger, P., Hornung, R., Epstein, J. N., Braun, J., & Kahn, R. S. (2009). Association of tobacco and lead exposures with attention-deficit/hyperactivity disorder. *Pediatrics, 124*, e1054–e1063.

Gąssowska, M., Baranowska-Bosiacka, I., Moczydłowska, J., Tarnowski, M., Pilutin, A., Gutowska, I., … Adamczyk, A. (2016). Perinatal exposure to lead (Pb) promotes Tau phosphorylation in the rat brain in a GSK-3β and CDK5 dependent manner: Relevance to neurological disorders. *Toxicology, 347-349*, 17–28.

GBD 2016 Risk Factor Collaborators. (2017). Global, regional, and national comparative risk assessment of 84 behavioural, environmental and occupational, and metabolic risks or clusters of risks, 1990-2016: A systematic analysis for the global burden of disease study 2016. *Lancet, 390*, 1345–1442.

Gibb, H., & O'Leary, K. G. (2014). Mercury exposure and health impacts among individuals in the artisanal and small-scale goldmining community: A comprehensive review. *Environmental Health Perspectives, 122*, 667–672.

Goodlad, J. K., Marcus, D. K., & Fulton, J. J. (2013). Lead and attention-deficit/hyperactivity disorder (ADHD) symptoms: A meta-analysis. *Clinical Psychology Review, 33*, 417–425.

Grandjean, P., Weihe, P., White, R. F., Debes, F., Araki, S., Yokoyama, K., … Jørgensen, P. J. (1997). Cognitive deficit in 7-year-old children with prenatal exposure to methylmercury. *Neurotoxicology and Teratology, 19*, 417–428.

Gulson, B. L., Mizon, K. J., Korsch, M. J., Palmer, J. M., & Donnelly, J. B. (2003). Mobilization of lead from human bone tissue during pregnancy and lactation—A summary of long-term research. *Science of the Total Environment, 303*, 79–104.

Harada, M. (1995). Minamata disease: Methylmercury poisoning in Japan caused by environmental pollution. *Critical Reviews in Toxicology, 25*, 1–24.

Kaufman, A. S., Zhou, X., Reynolds, M. R., Kaufman, N. L., Green, G. P., & Weiss, L. G. (2014). The possible societal impact of the decrease in U.S. blood lead levels on adult IQ. *Environmental Research, 132*, 413–420.

Khan, S. A., Singh, R. K., Navit, S., Chadha, D., Johri, N., Navit, P., … Bahuguna, R. (2015). Relationship between dental fluorosis and intelligence quotient of school going children in and around Lucknow district: A cross-sectional study. *Journal of Clinical Diagnosis and Research, 9*, ZC10–ZC15.

Kordas, K., Canfield, R. L., López, P., Rosado, J. L., Vargas, G. G., Cebrián, M. E., … Stoltzfus, R. J. (2006). Deficits in cognitive function and achievement in Mexican first-graders with low blood lead concentrations. *Environmental Research, 100*, 371–386.

Lam, J., Lanphear, B. P., Bellinger, D., Axelrad, D. A., McPartland, J., Sutton, P., … Woodruff, T. J. (2017). Developmental PBDE exposure and IQ/ADHD in childhood: A systematic review and meta-analysis. *Environmental Health Perspectives, 125*, 086001.

Lanphear, B. P., Hornung, R., Khoury, J., Yolton, K., Baghurst, P., Bellinger, D. C., … Roberts, R. (2005). Low-level environmental lead exposure and children's intellectual function: An international pooled analysis. *Environmental Health Perspectives, 113*, 894–899.

Lauterbach, M., Martins, I. P., Castro-Caldas, A., Bernardo, M., Luis, H., Amaral, H., … Derouen, T. (2008). Neurological outcomes in children with and without amalgam-related mercury exposure: Seven years of longitudinal observations in a randomized trial. *Journal of the American Dental Association, 139*, 138–145.

Levie, D., Korevaar, T. I. M., Bath, S. C., Dalmau-Bueno, A., Murcia, M., Espada, M., … Guxens, M. (2018). Thyroid function in early pregnancy, child IQ, and autistic traits: A meta-analysis of individual-participant data. *Journal of Clinical Endocrinology and Metabolism, 103*(8), 2967–2979.

Liew, Z., Goudarzi, H., & Oulhote, Y. (2018). Developmental exposures to perfluoroalkyl substances (PFASs): An update of associated health outcomes. *Current Environmental Health Reports, 5*, 1–19.

Lovasi, G. S., Eldred-Skemp, N., Quinn, J. W., Chang, H. W., Rauh, V. A., Rundle, A., … Perera, F. P. (2014). Neighborhood social context and individual polycyclic aromatic hydrocarbon exposures associated with child cognitive test scores. *Journal of Child and Family Studies, 23*, 785–799.

Magzamen, S., Amato, M. S., Imm, P., Havlena, J. A., Coons, M. J., Anderson, H. A., … Moore, C. F. (2015). Quantile regression in environmental health: Early life lead exposure and end-of-grade exams. *Environmental Research, 137*, 108–119.

Magzamen, S., Imm, P., Amato, M. S., Havlena, J. A., Anderson, H. A., Moore, C. F., & Kanarek, M. S. (2013). Moderate lead exposure and elementary school end-of-grade examination performance. *Annals of Epidemiology, 23*, 700–707.

Miranda, M. L., Kim, D., Reiter, J., Galeano, M. A. O., & Maxson, P. (2009). Environmental contributors to the achievement gap. *Neurotoxicology, 30*, 1019–1024.

Nakiwala, D., Peyre, H., Heude, B., Bernard, J. Y., Béranger, R., Slama, R., & Philippat, C. (2018). EDEN mother-child study group. In-utero exposure to phenols and phthalates and the intelligence quotient of boys at 5 years. *Environmental Health, 17*(1), 17.

Needleman, H. L., Gunnoe, C., Leviton, A., Reed, R., Peresie, H., Maher, C., & Barrett, P. (1979). Deficits in psychologic and classroom performance of children with elevated dentine lead levels. *New England Journal of Medicine, 300*(13), 689–695.

Needleman, H. L., Schell, A., Bellinger, D., Leviton, A., & Allred, E. N. (1990). The long-term effects of exposure to low doses of lead in childhood. An 11-year follow-up report. *New England Journal of Medicine, 322*(2), 83–88.

Nevin, R. (2009). Trends in preschool lead exposure, mental retardation, and scholastic achievement: Association or causation? *Environmental Research, 109*(3), 301–310.

Nigg, J. T., Knottnerus, G. M., Martel, M. M., Nikolas, M., Cavanagh, K., Karmaus, W., & Rappley, M. D. (2008). Low blood lead levels associated with clinically diagnosed attention-deficit/hyperactivity disorder and mediated by weak cognitive control. *Biological Psychiatry, 63*, 325–331.

Nigg, J. T., Nikolas, M., Knottnerus, G. M., Cavanagh, K., & Friderici, K. (2010). Confirmation and extension of association of blood lead with attention-deficit/hyperactivity disorder (ADHD) and ADHD symptom domains at population-typical exposure levels. *Journal of Child Psychology and Psychiatry, 51*, 58–65.

Oken, E., Wright, R. O., Kleinman, K. P., Bellinger, D., Amarasiriwardena, C. J., Hu, H., … Gillman, M. W. (2005). Maternal fish consumption, hair mercury, and infant cognition in a U.S. Cohort. *Environmental Health Perspectives, 113*(10), 1376–1380.

Park, J. H., Seo, J. H., Hong, Y. S., Kim, Y. M., Kang, J. W., Yoo, J. H., … Choe, B. M. (2016). Blood lead concentrations and attention deficit hyperactivity disorder in Korean children: A hospital-based case control study. *BMC Pediatrics, 16*(1), 156.

Peterson, B. S., Rauh, V. A., Bansal, R., Hao, X., Toth, Z., Nati, G., … Perera, F. (2015). Effects of prenatal exposure to air pollutants (polycyclic aromatic hydrocarbons) on the development of brain white matter, cognition, and behavior in later childhood. *JAMA Psychiatry, 72*(6), 531–540.

Pujol, J., Martínez-Vilavella, G., Macià, D., Fenoll, R., Alvarez-Pedrerol, M., Rivas, I., … Sunyer, J. (2016). Traffic pollution exposure is associated with altered brain connectivity in school children. *NeuroImage, 129*, 175.

Rauh, V., Arunajadai, S., Horton, M., Perera, F., Hoepner, L., Barr, D. B., & Whyatt, R. (2011). Seven-year neurodevelopmental scores and prenatal exposure to chlorpyrifos, a common agricultural pesticide. *Environmental Health Perspectives, 119*, 1196–1201.

Rauh, V. A., Perera, F. P., Horton, M. K., Whyatt, R. M., Bansal, R., Hao, X., … Peterson, B. S. (2012). Brain anomalies in children exposed prenatally to a common organophosphate pesticide. *Proceedings of the National Academy of Sciences USA, 109*(20), 7871–7876.

Reuben, A., Caspi, A., Belsky, D. W., Broadbent, J., Harrington, H., Sugden, K., … Moffitt, T. E. (2017). Association of childhood blood lead levels with cognitive function and socioeconomic status at age 38 years and with IQ change and socioeconomic mobility between childhood and adulthood. *Journal of the American Medical Association, 317*(12), 1244–1251.

Schneider, J. S., & DeKamp, E. (2007). Postnatal lead poisoning impairs behavioral recovery following brain damage. *Neurotoxicology, 28*(6), 1153–1157.

Selevan, S. G., Kimmel, C. A., & Mendola, P. (2000). Identifying critical windows of exposure for children's health. *Environmental Health Perspectives, 108*(Suppl 3), 451–455.

Seo, J., Lee, B. K., Jin, S. U., Jang, K. E., Park, J. W., Kim, Y. T., … Chang, Y. (2015). Altered executive function in the lead-exposed brain: A functional magnetic resonance imaging study. *Neurotoxicology, 50*, 1–9.

Skerfving, S., Lofmark, L., Lundh, T., Mikoczy, Z., & Stromberg, U. (2015). Late effects of low blood lead concentrations in children on school performance and cognitive functions. *Neurotoxicology, 49*, 114–120.

Stern, A. H., & Smith, A. E. (2003). An assessment of the cord blood: Maternal blood methylmercury ratio: Implications for risk assessment. *Environmental Health Perspectives, 111*, 1465–1470.

Surkan, P. J., Zhang, A., Trachtenberg, F., Daniel, D. B., McKinlay, S., & Bellinger, D. C. (2007). Neuropsychological function in children with blood lead levels <10 microg/dL. *Neurotoxicology, 28*(6), 1170–1177.

Swan, S. H., Liu, F., Hines, M., Kruse, R. L., Wang, C., Redmon, J. B., … Weiss, B. (2010). Prenatal phthalate exposure and reduced masculine play in boys. *International Journal of Andrology, 33*(2), 259–269.

Téllez-Rojo, M. M., Bellinger, D. C., Arroyo-Quiroz, C., Lamadrid-Figueroa, H., Mercado-García, A., Schnaas-Arrieta, L., … Hu, H. (2006). Longitudinal associations between blood lead concentrations lower than 10 microg/dL and neurobehavioral development in environmentally exposed children in Mexico City. *Pediatrics, 118*(2), e323–e330.

Wang, H. L., Chen, X. T., Yang, B., Ma, F. L., Wang, S., Tang, M. L., … Ruan, D. Y. (2008). Case-control study of blood lead levels and attention deficit hyperactivity disorder in Chinese children. *Environmental Health Perspectives, 116*(10), 1401–1406.

Weiss, B., Clarkson, T. W., & Simon, W. (2002). Silent latency periods in methylmercury poisoning and in neurodegenerative disease. *Environmental Health Perspectives, 110*(Suppl 5), 851–854.

Weisskopf, M. G., Kioumourtzoglou, M. A., & Roberts, A. L. (2015). Air pollution and autism spectrum disorders: Causal or confounded? *Current Environmental Health Reports, 2*, 430–439.

White, R. F., Palumbo, C. L., Yurgelun-Todd, D. A., Heaton, K. J., Weihe, P., Debes, F., & Grandjean, P. (2011). Functional MRI approach to developmental methylmercury and polychlorinated biphenyl neurotoxicity. *Neurotoxicology, 32*(6), 975–980.

Woodruff, T. J., Zota, A. R., & Schwartz, J. M. (2011). Environmental chemicals in pregnant women in the United States. *Environmental Health Perspectives, 119*(6), 878–885.

Wright, J. P., Dietrich, K. N., Ris, M. D., Hornung, R. W., Wessel, S. D., Lanphear, B. P., … Rae, M. N. (2008). Association of prenatal and childhood blood lead concentrations with criminal arrests in early adulthood. *PLoS Medicine, 5*(5), e101.

Yorifuji, T., Kato, T., Ohta, H., Bellinger, D. C., Matsuoka, K., & Grandjean, P. (2016). Neurological and neuropsychological functions in adults with a history of developmental arsenic poisoning from contaminated milk powder. *Neurotoxicology and Teratology, 53*, 75–80.

Yuan, W., Holland, S. K., Cecil, K. M., Dietrich, K. N., Wessel, S. D., Altaye, M., … Lanphear, B. P. (2006). The impact of early childhood lead exposure on brain organization: A functional magnetic resonance imaging study of language function. *Pediatrics, 118*(3), 971–977.

Zheng, W., Aschner, M., & Ghersi-Egea, J. F. (2003). Brain barrier systems: A new frontier in metal neurotoxicological research. *Toxicology and Applied Pharmacology, 192*(1), 1–11.

Part VI

Assessment

21

The Role of Intelligence Tests in the Assessment of Intellectual Disabilities

Mark R. McGowan and Jenna M. Hennessey

An intellectual disability (ID) is a developmental disorder that impacts functioning over the course of an individual's life span and is marked by limitations in both intellectual and adaptive domains. The onset of these limitations must manifest prior to the age of 18. While both genetic and environmental factors contribute to this condition, the most common causes of the disorder are due to chromosomal abnormalities (e.g., fragile X syndrome or Down syndrome) and fetal exposure to drugs or toxins (Walker & Johnson, 2006). ID impacts approximately 1% of the general population (American Psychiatric Association [APA], 2013). Intellectual deficits may impact an individual's reasoning, problem solving, planning, abstract thinking, judgment, or capacity for learning. Additionally, an individual must demonstrate an inability to meet normative expectations for independence and self-sufficiency in either conceptual, social, or practical adaptive skills (Schalock et al., 2010). According to the Diagnostic and Statistical Manual of Mental Disorders, fifth edition (APA, 2013), intellectual deficits are substantiated through the use of "both clinical assessment and individualized, standardized intelligence testing" (p. 33). Although the diagnostic criteria have not changed substantially over the past 50 years, the manner in which intelligence estimates are used in diagnosing ID has changed (Brown, 2007; Schalock, Luckasson, & Shogren, 2007).

Historically, determinations concerning the diagnosis and severity of impairment (e.g., mild, moderate, severe, profound) were based upon the magnitude of deviation of an individual's IQ score from the mean. For example, individuals with IQ scores ranging from 50–55 to 70 were classified as mildly impaired while individuals with IQ scores ranging from 35–40 to 50–55 were moderately impaired. More recently, the diagnostic criteria for ID was changed to emphasize the use of adaptive functioning deficits to determine the level of impairment (Tassé, Luckasson, & Schalock, 2016). This shift in emphasis was based in part on lingering concerns regarding the validity of estimates observed at the lower end of the IQ range (Armstrong, Hangauer, & Nadeau, 2012; Flanagan & Alfonso, 1995). The clinical implications of this change mean that, while intelligence tests continue to play an important role in determining that the criteria necessary for diagnosing ID have been met, clinical determinations concerning the degree of impairment are based on more readily observable behaviors rather than the latent construct of intelligence. In this chapter, we will focus on the role intelligence tests have played in our understanding of ID as a diagnostic construct, the instruments currently used to assess it, and the clinical considerations for their use.

M. R. McGowan (✉) · J. M. Hennessey
Department of Educational and School Psychology,
Indiana University of Pennsylvania,
Indiana, PA, USA
e-mail: mmcgowan@iup.edu

J. L. Matson (ed.), *Handbook of Intellectual Disabilities*, Autism and Child Psychopathology Series, https://doi.org/10.1007/978-3-030-20843-1_21

Intellectual Assessment

Intelligence testing has evolved over the course of more than a century. While consensus on a definition of intelligence has been elusive, the history of intelligence testing has, in many respects, developed in parallel with theoretical understandings and contemporary definitions of this construct (see Wasserman, 2012). As we will discuss, a reciprocal relationship exists between the conceptualization of intelligence and how it is measured. This relationship holds great bearing upon how practitioners conceptualize intellectual disabilities (ID). Interestingly, many of the questions that were central to the study and assessment of intelligence at its beginning continue to exist today. Terman's (1916) observation concerning balancing the need for an agreed upon theoretical definition of intelligence and the task of measuring the construct continues to hold relevance.

> To demand, as critics of the Binet method have sometimes done, that one who would measure intelligence should first present a complete definition of it, is quite unreasonable. As Stern points out, electrical currents were measured long before their nature was well understood. Similar illustration could be drawn from the processes involved in chemistry, physiology, and other sciences. In the case of intelligence, it may be truthfully said that no adequate definition can possibly be framed which is not based primarily on the symptoms empirically brought to light by the test method (p. 36).

While agreed upon definitions have been difficult to achieve, intelligence testing has traditionally been the primary method for diagnosing ID. In most clinical settings, IQ scores that are two standard deviations or more below the mean have served as the diagnostic marker for ID. Not surprisingly, the evolution of the diagnostic nomenclature for ID can be traced in the historical tapestry that has developed from the study of intelligence over time. Therefore, a brief history of intelligence testing and the forces that have shaped its development will be introduced prior to reviewing contemporary measures used to assess intellectual capacities in children and adolescents.

A Brief History

The origins of intelligence testing date back to the late 1800s. The history of testing often begins with the work of Francis Galton and James Cattell who pioneered the use of anthropometric measurement to identify human characteristics that could be used to understand individual differences, heritability, and vocational potential (Galton, 1890). These early researchers also laid the groundwork for the use of psychometric principles and statistical analyses in the development and interpretation of assessment instruments (Cattell, 1890). While the work of these early researchers provides an important context for understanding the evolution of contemporary intelligence tests, Alfred Binet is generally credited with developing the first working intelligence test in 1905 and with ushering in the wide-ranging use of testing in western culture (Jenkins & Paterson, 1961; Tuddenham, 1962).

The original Binet-Simon Scale (Binet & Simon, 1905/1916) consisted of 30 items designed to assess a variety of abilities in children ages 3–11 years. He employed standardized test administration procedures that introduced items in a developmentally sensitive sequence from easy to hard and interpretation was based on the number of items a child was able to successfully complete. The impetus for developing the Binet-Simon Scale was in direct response to a need for educators to identify and support the learning needs of children with ID in accordance with mandatory public education laws in France at the time (Wolf, 1969). Later versions of the scale were developed in order to broaden its use by extending the age range from 3 years through adulthood. Binet also pioneered other advancements in the field. For example, he introduced the concept of using mental age to summarize an individual's performance on the test. In doing so, Binet established objective criteria for the diagnosis of intellectual disabilities based on their performance on the Binet-Simon Scale. He suggested that, for children under the age of 9, a 2-year delay in mental age be used as the threshold for an intellectual disability and, for children above the age of 9, a 3-year delay should be used.

The scale measured higher order cognitive functions including language, auditory processing, visual processing, learning and memory, judgment, and problem-solving (Binet, 1911/1916). It is important to note that the Binet-Simon Scale was developed using empirical evidence rather than theory. While Binet was convinced that more complex cognitive processes should be the focus of measurement as opposed to simple motor or sensory processes, he defined the construct of intelligence in fairly broad terms suggesting that it "…marks itself by the best possible adaptation of the individual to his environment" (p. 301).

Interestingly, the Binet-Simon Scale was not used widely in the French educational system at the time (Carson, 2007). However, the importance of the instrument for diagnosing intellectual disabilities was noted by an American, Henry H. Goddard (Zenderland, 1998). Goddard promoted the use of both the instrument and Binet's method for diagnosing ID in the United States. While Goddard was instrumental in having Binet's work translated into English, it was Lewis Terman who would later adapt the Binet-Simon Scale for widespread use in the United States. Based upon historical accounts, Lewis Terman, a professor at Stanford University, was granted the rights to publish an American revision of the Binet-Simon Scale for a nominal fee (see Wolf, 1973). Terman's work would eventually lead to the first edition of the Stanford-Binet Intelligence Scale, which introduced a number of noteworthy advancements in intellectual assessment. First, Terman's revised scale incorporated theoretical frameworks for guiding test construction and interpretation by purporting to measure "*general intelligence*" (Terman, 1916; p. 48). The concept of general intelligence was introduced earlier by Spearman (1904) in a paper reporting on his discovery of a mathematically derived factor, or psychometric *g*, that was hypothesized to exist based upon shared variance noted between test items. Second, Terman's revision also differed from the original test in its use of an Intelligence Quotient (IQ) to summarize test performance. The IQ score was calculated by dividing the mental age by the chronological age and multiplying the sum by 100. While the use of this type of ratio IQ score would later fall into disfavor due to psychometric vulnerabilities (see Becker, 2003), the use of IQ scores for diagnosing intellectual disabilities has stood the test of time.

Terman's standardization sample for the Stanford-Binet was comprised of 905 youth between the ages of 5 and 14 years who were drawn from schools in California and Nevada. The adult sample included 200 adolescents and 180 men in California and Oregon. The final version of the Stanford-Binet had 90 items, with approximately 60% of the items being adapted from the original test. Based upon frequency distributions found in the American standardization sample for the Stanford-Binet, children with IQs of 70 and below were classified as intellectually disabled, while children with IQs between 80 and 70 were classified as "Border-line" (Terman, 1916, p. 79). Over time, the Stanford-Binet would become the most frequently used intelligence test in the United States for decades (Louttit & Browne, 1947).

However, external forces continued to shape innovation in the field of intelligence testing that resulted in significant advancements in contemporary assessment practices. As noted previously, the genesis of the Stanford-Binet can be traced back to demands facing the public educational system. The successor, in terms of popularity of use, to the Stanford-Binet traces its roots back to demands facing the United States military. At the start of World War I, Robert M. Yerkes, then president of the American Psychological Association (APA), convened the Committee on the Psychological Examination of Recruits in 1917 for the purpose of leveraging psychology to assist the government in a time of war. The APA committee's focus was on using intelligence testing to identify recruits who were either unfit for duty or who demonstrated potential for leadership roles in the military. The product of this committee's work resulted in the development of the Army Examination Alpha and Beta. Army Alpha was an intelligence test intended for use with literate English-language speakers, while the Army Beta was designed for use with limited English-language proficient or illiterate examinees

(Yoakum & Yerkes, 1920). These intelligence tests were designed to be administered to large groups of examinees by Army examiners who were specially trained at the School for Military Psychology. Over the course of the war, Yerkes (1919) estimated that 1,726,000 servicemen had participated in psychological examinations, with an additional 83,000 enlisted men being given individual examinations.

Among the many Army examiners who contributed to this enterprise was a graduate student by the name of David Wechsler. While his contribution to the field of intelligence testing would not manifest for a number of years following the war, Wechsler drew upon his experience as an Army examiner to develop the Wechsler-Bellevue Scales in 1939 (Wechsler, 1939), which would later become the Wechsler-Bellevue Intelligence Scales (Wechsler, 1946). The Wechsler-Bellevue was comprised entirely of subtests taken from other intelligence tests available at that time. Wechsler's intent was to produce an intelligence test that would provide clinicians with a better option for assessing adults than the Stanford-Binet, which received criticism for its limited adult normative sample (Wechsler, 1939). While the development of the Wechsler-Bellevue was not driven by a particular theoretical model, Wechsler did embrace theoretical notions prevalent at the time in his definition of intelligence. According to Wechsler (1939),

> Intelligence is the aggregate or global capacity of the individual to act purposefully, to think rationally and to deal effectively with his environment. It is global because it characterizes the individual's behavior as a whole; it is an aggregate because it is composed of elements or abilities which, though not entirely independent, are qualitatively differentiable. By measurement of these abilities, we ultimately evaluate intelligence. But intelligence is not identical with the mere sum of these abilities, however inclusive (p. 3).

The Wechsler scales offered a number of advantages over its competitors at the time. First, Wechsler used a deviation IQ, which is based on how an individual's performance deviates from the average IQ for the sample. Unlike the ratio IQ used by the Stanford-Binet, a deviation IQ provides an index of intelligence based on the examinee's age and the statistical distance from the mean. A second advancement offered by the Wechsler scales was the inclusion of both Verbal and Performance (nonverbal) tasks, which was an approach adapted from lessons learned from the development of the Army Alpha and Beta tests (Wasserman, 2012). Subtest raw scores were converted to standard scores with a mean of 10 and a standard deviation of 3, while IQ scores had a mean of 100 and standard deviation of 15. The conversion of raw scores to standard scores also provided clinicians with opportunities to analyze intra-individual performance in the interpretation of the results. This approach to profile analysis based on the interpretation of individual strengths and weaknesses continues to shape practice today. Finally, by drawing together well-known tests from other instruments, Wechsler was able to provide practitioners with a single battery of co-normed tests that were administered and scored in a similar manner to produce a psychometrically sound and valid intelligence battery of tests. While critics of the Wechsler scales have commented on its lack of theoretical orientation (Anastasi, 1942), the empirical support and popularity of the test among practitioners made it the instrument of choice for working with adult populations (Rabin & Guertin, 1951). Over time, Wechsler went on to publish many different intelligence measures until his death in 1981 that include the Wechsler Intelligence Scale for Children (WISC; Wechsler, 1949), the Wechsler Adult Intelligence Scale (WAIS; Wechsler, 1955), and the Wechsler Preschool and Primary Scale of Intelligence (WPPSI; Wechsler, 1967). Over the years, these measures have been through multiple revisions and continue to be very popular among practitioners (Oakland, Douglas, & Kane, 2016).

A Contemporary Theory of Intelligence

A comprehensive overview of this topic is beyond the scope of this chapter. However, an effort will be made to focus our discussion on the theory that appears with the greatest regularity in the intelligence tests reviewed in this chapter. Before doing so, it is important to again reflect on how intelligence is currently being defined. While no universally agreed upon definition exist, our understanding of intelligence has continued to evolve in relation to the methods that are used to measure it. Consequently, some researchers have argued that, in the absence of adequate language to define the construct, structural or statistical models should be used to guide our understanding of intelligence (Gottfredson & Saklofske, 2009). This notion has been the foundation for the evolution of the Cattell-Horn-Carroll (CHC) framework which currently represents that most comprehensive and well-accepted model of intelligence to date (Keith & Reynolds, 2010).

The CHC theoretical framework is a structural model of intelligence that developed from the work of Raymond B. Cattell, John L. Horn, and John B. Carroll. The work of Cattell and Horn focused on developing a multifactor model of intelligence based upon Cattell's Gf-Gc theory (Cattell, 1963; Horn & Cattell, 1966). While the original theory hypothesized that general intelligence or (*g*) was the result of fluid and crystallized intelligence factors working in concert in the service of learning, the evolution of the theory has continued to expand the number of factors included in the theory (see Cattell, 1998). Based upon continued factor analytic research, the Horn-Cattell Extended Gf-Gc theory grew from a two-factor model to one that recognized between 9 or 10 broad ability factors (Horn & Noll, 1997). The relevance of this theory rests not only on its empirical replicability, but also on its utility for explaining patterns of variability observed in these results (Schneider & McGrew, 2012).

Carroll's three-stratum model is the other theoretical contributor to contemporary CHC theory. Drawing upon more than 460 factor analytic studies of intellectual abilities available at that time, Carroll's seminal research introduced a hierarchical model that, as the name suggests, is comprised of three strata or levels. The framework takes on a shape of a pyramid. At its pinnacle (stratum III), the model posits that a general intelligence factor (or *g*) is comprised of the broad and narrow ability factors in the other two levels of the model. Stratum II includes eight board abilities that are recruited based upon the task demands faced by the individual. At stratum I, there are a large number of narrow abilities subsumed under each of the broad abilities in stratum II (Carroll, 1993). The three-stratum model demonstrated considerable overlap with the Gf-Gc theory. More specifically, Carroll's broad abilities at stratum II are similar to those found in the Horn-Cattell Extended Gf-Gc theory including fluid intelligence (Gf), crystallized intelligence (Gc), general memory and learning (Gy), broad visual perception (Gv), broad auditory perception (Ga), broad retrieval ability (Gr), broad cognitive speediness (Gs), and reaction time/decision speed (Gt). However, the three-stratum model expanded the framework of the Gf-Gc model by introducing a taxonomy for understanding the narrow factors that contribute to differences at the broad ability level. The importance of understanding these narrow ability factors was summarized by Kevin McGrew who suggested that focusing on these factors would "go far in helping us all better understand what we are measuring, facilitate better communication between and among professionals and scholars, and increase our ability to compare individual tests across and within intelligence batteries" (1997, p. 171). This line of reasoning eventually gave rise to CHC theory.

The first integration of the Horn-Cattell and Carroll models was articulated in a chapter by McGrew (1997). CHC theory has continued to command the attention of researchers and test publishers since 2000 (McGrew, 2005). Currently, the CHC theoretical framework has arguably had the most influential impact on contemporary intelligence testing of any theory to date (Newton & McGrew, 2010). According to Keith and Reynolds (2010), "although most new and revised tests of intelligence are based, at least in part, on CHC theory, earlier versions generally were not. Our review suggests that whether or not they were based on CHC theory, the factors derived from both new and previous versions of most tests are well explained by the theory" (p. 635). In addition to providing the theoretical foundation for the majority of commonly used intelligence tests, CHC theory has also been used to develop practice guidelines for assessment and interpretation of intelligence tests, e.g., cross-battery approaches (see Flanagan, Ortiz, & Alfonso, 2013).

In practice, the relevance of CHC theory to the assessment and diagnosis of intellectual disabilities is less clear. While the CHC model provides for rare common ground between test developers and theorists, practitioners are only beginning to see specific guidance on how CHC may aid in making differential diagnoses. Preliminary research has largely been limited to the identification of specific learning disabilities (Flanagan, Fiorello, & Ortiz, 2010; Niileksela & Reynolds, 2014; Proctor, 2012). As continued efforts are made to identify how CHC ability factors relate to learning outcomes for children (McGrew & Wendling, 2010), the benefits for utilizing CHC theory to guide intervention planning for individuals with ID may prove to be beneficial. Regardless, practitioners must be knowledgeable about the constructs that are being measured in order to make informed decisions concerning the selection of appropriate assessment batteries and to ensure that the interpretations based upon those findings are valid. The remainder of this chapter will review instruments frequently used to measure intelligence in child and adolescent populations. Diagnostic and clinical implications associated with selecting and interpreting these instruments for use with the ID population will also be explored.

Stanford-Binet Intelligence Scales, Fifth Edition (SB5)

The Stanford-Binet Intelligence Scales, Fifth Edition (SB5; Roid, 2003a) is the most current version of the Stanford-Binet Scales. There were significant modifications made to the previous version, Stanford-Binet Intelligence Scales, fourth edition (SB4; Thorndike, Hagen, & Sattler, 1986), when developing the SB5. These changes included an additional factor to the test framework, improved content under the nonverbal index, extended age range, new test items to enhance the measurement of very low functioning individuals, and reintroduced manipulatives from earlier editions of this instrument with the intention of increasing examinees' engagement (Roid, 2003b).

Test Description. The SB5 can be administered to children as young as 2-years of age, which allows for early detection of developmental delay. This instrument also grants examiners the ability to assess intelligence throughout adulthood with norms spanning to age 85+ (Roid, 2003b). Administration times vary based upon the battery of tests administered. These administration times range from 15–20 min for the Abbreviated Battery to 45–75 min for the Full-Scale IQ Battery (Roid, 2003c). The SB-5 was normed on 4800 individuals between the ages of 2 and 85+. Furthermore, approximately 77% of the sample population ranged in age between 2 and 20. "The sample was nationally representative to percentages of the stratification variables identified in the U.S. Census data in 2001" (Roid, 2003c, p.48). The norming sample includes individuals with ID and other developmental disabilities (Roid, 2003c).

Theoretical Orientation and Test Structure. The SB5 exclusively uses the CHC theory to support its test framework. The SB5 includes five broad cognitive abilities highlighted in the CHC model (Roid, 2003b). According to Roid (2003c), "the five factors included in this instrument have the highest factor loading on general intellectual ability (*g*)" (p.9). The average factor loadings on all of the broad cognitive abilities across all age groups for both verbal and nonverbal domains are as follows. The factor loading on *g* for the verbal domain of the five broad abilities are Fluid Reasoning (0.77), Knowledge (0.75), Quantitative Reasoning (0.83), Visual-Spatial Processing (0.82), and Working Memory (0.76). The factor loading on *g* for the nonverbal domains of the five broad abilities are Fluid Reasoning (0.70), Knowledge (0.80), Quantitative Reasoning (0.82), Visual-Spatial Processing (0.74), and Working Memory (0.74). See the *Stanford- Binet Technical Manual* for additional information regarding the factor loadings for this instrument (Roid, 2003c).

Psychometric Properties. The SB-5 is well known for its respectable reliability and validity (Sattler, 2007). According to the *Technical Manual* (Roid, 2003c), the internal reliability is exceptional for the Full-Scale IQ with alpha coefficients ranging from 0.97 to 0.98 across age groupings. The five broad cognitive abilities factors discussed above also yield reputable internal consistency ($r = 0.84–0.96$) across all age groupings. The author of this assessment tool also indicated excellent stability in terms of the Full-Scale IQ and across all five-factor indexes. In regards to validity, high correlation ($r = 0.90$) was noted between the Full IQ Scale of the SB5 to its predecessor's, SB4, Composite Scale. In addition, the SB5s Full IQ Scale also highly correlated ($r = 0.78–0.84$) with other well-known standardized intelligence assessments (Roid, 2003c).

Wechsler Intelligence Scale for Children, Fifth Edition (WISC-V)

The Wechsler Intelligence Scale for Children, Fifth Edition (WISC-V; Wechsler, 2014) is the most current children's version of the original Wechsler Intelligence Scale for Children (WISC; Wechsler, 1949). The WISC-V aimed to improve the content through adding three subtests to the core battery as well as enhance the factor structure through increasing the number of factors by one to better measure isolated domains of intellectual ability, such as visual-spatial ability. Ancillary and complementary diagnostic index scales were also added to provide additional information regarding a child or adolescents' overall cognitive functioning in the areas of reasoning ability, nonverbal reasoning, auditory working memory, and memory retrieval (Wechsler, Raiford, & Holdnack, 2014).

Test Description. The age range for the WISC-V includes children between the ages 6 through the age of 16. The *WISC-V Technical and Interpretive Manual* claims that the administration of the core subtests can be completed in approximately 60 min (Wechsler et al., 2014); however, this assessment is perceived by practitioners to often take upwards of 120 min to administer. Normative data were collected on 2200 children spanning the entire age range of the assessment. The sample is representative of "demographic variables, such as (i.e., age, sex, race/ethnicity, parent education level, geographic region) using October 2012 U.S. census data" (Wechsler et al., 2014, p. 31). Furthermore, the WISC-V norms include participants from various disability classifications including ID and developmental disabilities (Wechsler et al., 2014).

Theoretical Orientation and Test Structure. Dating back to the inception of the WISC, there has not been a single theoretical framework guiding the development of this assessment. The WISC-V encompasses an eclectic theoretical orientation rooted in neurocognitive research, working memory models, and CHC theory of intelligence (Wechsler et al., 2014). The factor structure for the WISC-V includes five primary indexes. The average factor loadings on g for these primary indexes across all age groups are as follows: Verbal Comprehension Index (0.84), Fluid Reasoning Index (0.99), Visual-Spatial Index (0.90), Working Memory Index (0.83), and Processing Speed Index (0.51). Refer to *WISC-V Technical Interpretive Manual* for additional information concerning the factor loadings associated with this assessment tool (Wechsler et al., 2014).

Psychometric Properties. The WISC-V is well known for having sufficient psychometric properties (Sattler, 2007). Internal reliability is very high for Full-Scale IQ at all ages ($r = 96–0.97$). The reliability alpha coefficients across all five primary index scores on all age groupings range between 0.84 and 0.95. The authors of this assessment tool also report excellent stability for the Full-Scale IQ and across all primary indexes. With regard to validity, the WISC-V's Full-Scale IQ highly correlates ($r = 0.86$) with the previous version of this instrument, WISC- IV. Furthermore, this assessment tool is highly correlated ($r = 0.83$ or above) with other Wechsler Scales (Wechsler et al., 2014).

Woodcock-Johnson IV Tests of Cognitive Abilities (WJ IV COG)

Woodcock-Johnson IV Tests of Cognitive Abilities (WJ IV COG; Schrank, McGrew, & Mather, 2014) is the most recent version of this instrument. Noteworthy modifications were made to this assessment tool during test development from the prior version (WJ III COG; Woodcock, McGrew, & Mather, 2001). In addition to renorming, the primary revisions include the addition of new tests as well as new clusters, such as the Gf-Gc Composite, Perceptual Speed, and Number Facility, to improve diagnostic interpretations (McGrew, LaForte, & Schrank, 2014).

Test Description. The WJ IV COG can be administered to children and adults ranging in age between years 2 through 90+. This assessment tool is divided into two batteries. The Standard Battery takes approximately 60 min to administer. The testing duration of the Extended Battery is about 40 min (Mather & Wendling, 2014). The WJ IV COG was normed on 7416 individuals between ages 2 and 90+ years. Over 5000 of these individuals were under the age of 20. The normative sample was collected over 25 months and was representative of U.S. Census data in 2010. Sampling variables included sex, race, ethnicity, community type, parent education, etc. The sample population includes individuals with ID and developmental disabilities (McGrew et al., 2014).

Theoretical Orientation and Test Structure. The WJ IV COG was designed specifically to measure constructs outlined in CHC theory. This instrument incorporates seven broad cognitive abilities encompassed in the CHC theory (Mather & Wendling, 2014; McGrew et al., 2014). The median factor loadings on *g* for these seven broad cognitive abilities across all age groups are as follows: Comprehension-Knowledge (0.83), Fluid Reasoning (1.0), Visual Processing (0.82), Short-term Working Memory (0.76), Long-term Retrieval (0.85), Cognitive Processing Speed (0.57), and Auditory Processing (0.78). See *WJ IV COG Technical Manual* for specific information regarding the factor loadings of this instrument (McGrew et al., 2014).

Psychometric Properties. *WJ IV COG Technical Manual* reports that internal reliability is very high for the Gf-Gc Composite and General Intellectual Ability with alpha coefficients ranging from 0.94 to 0.98. In addition, the broad abilities also have adequate internal consistency of 0.82 or higher on average. The authors of this assessment tool note excellent stability across all seven broad abilities. Adequate criterion-related validity was also reported based upon other commonly used intelligential assessments (McGrew et al., 2014).

Differential Ability Scales, Second Edition (DAS-II)

The Differential Ability Scales, Second Edition (DAS-II; Elliott, 2007a) is the most recent version of this assessment tool. The DAS-II includes an expanded clinical sample of children with a variety of disabilities as well as a Spanish language translation for the nonverbal subtests (Elliott, 2007a).

Test Description. The DAS-II consists of two assessment batteries. The Early Years Battery includes children between the ages of 2 years 6 months through age 6. This battery can also be administered to children ages 7 through 8 with lower cognitive functioning. The School-age Battery includes children from age 7 through 17. Children ages 5 through 6 who display high cognitive functioning can be administered this battery. In regards to administration time, the core battery of the DAS-II takes approximately 45 to 60 min to administer (Elliott, 2007b). If diagnostic subtests are selected by the examiner to acquire additional information on a child's cognitive functioning, administration time needed is approximately 5 min per diagnostics subtest (Sattler, 2007). The normative sample for the DAS-II included 3480 children and was associated with the October 2002 census data across selected demographic variables including age, sex, race/ethnicity, parent education level, and geographic region. As previously mentioned, the revision of this instrument expanded the clinical sample of children among a variety of disabilities including mild to moderate ID and developmental disabilities (Elliott, 2007b).

Theoretical Orientation and Test Structure. The DAS-II was not shaped by a single theory. Rather, this instrument used multiple theories to guide its development including Spearman's theory of general intelligence (Spearman, 1904), Thurstone's (1938) seven primary mental abilities, Wechsler's (1939) overall intelligence, Vernon's (1950) verbal-educational factor and spatial-mechanical factor, and Cattell's and Horn's (Horn & Cattell, 1966) Gf-Gc theory. Based on the above theoretical foundations, the DAS-II yields an overall intellectual score, General Conceptual Ability, along with cluster scores for both the Early Years battery and the School Years battery. For the Early Years battery, the average factor loadings on *g* for clusters across ages 4 through age 5 are as follows: Verbal (0.86), Nonverbal Reasoning (0.99), Spatial (0.90), Visual-Verbal Memory (0.50), and Verbal Short-term Memory (0.65). For the School Years battery, the average factor loadings on *g* for clusters across ages 6 through age 17 are as follows: Verbal (0.84), Nonverbal Reasoning (0.95), Spatial (0.91), Visual-Verbal Memory (0.53), Verbal Short-term Memory (0.84), and Cognitive Speed (0.67). Refer to the *DAS-II Introductory and Technical Handbook* for additional information on factor loadings on *g* that include younger age ranges (Elliott, 2007b).

Psychometric Properties. The DAS-II has reputable reported internal consistency, stability, and criterion-related validity (Elliott, 2012). In particular, the reliability alpha coefficients for the General Conceptual Ability is 0.93 or higher across age groupings. The cluster scores for all age groupings on both the Early Years and School-Age batteries on average range between 0.89 or higher. The DAS-II also displays adequate stability over time. The DAS-II's General Conceptual Ability is highly correlated with the DAS (r = 0.88). Furthermore, this instrument also adequately correlates with other well-known standardized intelligence assessments (Elliott, 2007b).

Kaufman Assessment Battery for Children, Second Edition (KABC-II)

The Kaufman Assessment Battery for Children, Second Edition (KABC; Kaufman & Kaufman, 2004a) was most recently revised in 2004. Consistent with the original version of this assessment tool, the authors claim that this assessment measures intellectual functioning in a culturally fair manner by using universal concepts on test items (Kaufman & Kaufman, 2004b).

Test Description. As previously mentioned, the KABC-II instrument's age range was widened to include children from ages 3 through the age of 18 (Kaufman & Kaufman, 2004b). Administration times vary based on the theoretical model the examiner decides to use. The core battery associated with the Luria model can be administered in approximately 30–55 min. In contrast, the CHC core battery is longer in duration with an estimated administration time between 40 and 70 min (Singer, Lichtenberger, Kaufman, Kaufman, & Kaufman, 2012). The KABC-II's normative sample of 3025 children was intended to accurately represent the 2001 U.S. census across stratification variables such as gender, race, socioeconomic status, and geographical region. Special education classifications incorporated in the sample population include individuals previously diagnosed with ID and developmental disabilities (Kaufman & Kaufman, 2004b).

Theoretical Orientation and Test Structure. The KABC-II was developed to align with two theoretical frameworks, Luria neuropsychological model (Luria, 1966) and CHC model (Horn & Cattell, 1966). The Luria model has a focus on cognitive processing. The four cognitive processes associated with the Luria model that are highlighted in the KABC-II are sequential processing, simultaneous processing, learning ability, and planning ability (Kaufman & Kaufman, 2004b). This model is favored in circumstances such as when a child exhibits bilingual language ability, when a child's cultural background is outside of the mainstream American culture, and when a child presents with a language impairment (Singer et al., 2012). In regards to the CHC model, five broad abilities are utilized in this assessment battery. Of these five broad abilities, the average factor loadings on *g* for age ranges 7 through 12 and 13 through 18 are as follows. The factor loadings on *g* for the age range 7 through 12 are Knowledge (0.85), Simultaneous (0.87), Planning (1.0), Learning (0.77), Sequential (0.58). The factor loadings on *g* for the age range 13 through 18 are Knowledge (0.83), Simultaneous (0.86), Planning (1.0), Learning (0.83), and Sequential (0.66). Refer to the manual for additional information regarding factor loadings on g (Kaufman & Kaufman, 2004b).

Psychometric Properties. The *KABC-II's Manual* (Kaufman & Kaufman, 2004b) reports that the mean internal reliability alpha coefficients' for both Mental Processing Index (MPI), Luria model's Full-scale IQ, and the Fluid-Crystallized Index (FCI), CHC model's Full-scale IQ, across age groups are 0.90 or higher. The alpha coefficients across all broad ability indexes on average range between 0.88 or higher. Performance on this instrument shows suitable stability (Singer et al., 2012). Adequate criterion-related validity was noted when comparing the MPI and FCI to other well-known intelligence assessments for children. It is worth noting that the FCI tends to correlate higher with other assessment instrument's global scales compared to the MPI (Kaufman & Kaufman, 2004b).

Universal Nonverbal Intelligence Test, Second Edition (UNIT-2)

The Universal Nonverbal Intelligence Test (UNIT-2; Bracken & McCallum, 2016a) was developed in 2016. All subtests included in this battery are administered without the use of verbal communication.

Test Description. The UNIT-2 assesses nonverbal intelligence of children and adolescents ranging from age 5 through age 21. Administration time varies depending on the battery given. These administration times range from 5 to 15 min for the Abbreviated Battery to 45–60 min for the Full-Scale IQ Battery (Bracken & McCallum, 2016b). The normative sample includes 1603 individuals. Similar to other intellectual assessments discussed, the chosen sample was representative of the United States population in terms of geographic region, race, ethnicity, gender, parent education, and household income. This instrument contains normative data on the following special populations such as ID and developmental disabilities (Bracken & McCallum, 2016b).

Theoretical Orientation and Test Structure. The UNIT-2 is not based on a single theory. The instrument was developed by drawing upon models of intelligence such as the Jensen's (1980) two-factor model of intelligence (i.e., memory and reasoning) and Carroll's (1993) intelligence model. The UNIT-2 test structure is hierarchical with overall intellectual functioning at the top (i.e., *g*) followed by primary ability scales and then subtests. The primary abilities scales consist of Memory, Reasoning, and Quantitative Reasoning. It is important to note that factor loadings of *g* reported in the *UNIT-2 Examiner's Manual* (Bracken & McCallum, 2016b) used subtests instead of the primary abilities scales. For ages 8 through 10, the average factor loadings on *g* for each subtest is as follows: Symbolic Memory (0.41), Spatial Memory (0.65), Analogic Reasoning (0.77), Cube Design (0.73), Nonsymbolic Quantity (0.60), and Numerical Series (0.72). For ages 11 through 13, the average factor loadings on *g* for each subtest is as follows: Symbolic Memory (0.55), Spatial Memory (0.64), Analogic Reasoning (0.69), Cube Design (0.67), Nonsymbolic Quantity (0.69), and Numerical Series (0.79). For ages 14 through 18, the average factor loadings on *g* for each subtest is as follows: Symbolic Memory (0.46), Spatial Memory (0.68), Analogic Reasoning (0.76), Cube Design (0.76), Nonsymbolic Quantity (0.66), and Numerical Series (0.76). See the technical manual for additional information regarding factor loadings on *g* related to other age ranges.

Psychometric Properties. The UNIT-2 is a psychometrically sound instrument. According to the manual, internal reliability is high across the Full-Scale IQ (r = 0.97–0.99) and all primary abilities scales (r = 0.91–0.99) for all age groupings. Test-retest stability is reportedly adequate for the Full-Scale IQ and all primary ability scales. Related to validity, the UNIT-2 adequately correlates with prior version of this instrument, UNIT (r = 0.96). This measure also highly correlates with well-known intelligence tests (Bracken & McCallum, 2016b).

Leiter International Performance Scale-Third Edition (Leiter-3)

The Leiter International Performance Scale-Third Edition (Leiter-3; Roid, Miller, Pomplun, & Koch, 2013a) is the most recent revision of this instrument. Similar to the UNIT-2, the Leiter-3 is administered nonverbally with pantomimed instructions such as hand gestures, facial expressions, and demonstrations (Roid et al., 2013a).

Test Description. The Leiter-3 assesses nonverbal intellectual functioning of children and adolescents ranging from 3 through 75+. Administration time varies depending on the battery given. The Cognitive Battery can be administered in approximately 45 min and the Attention/Memory Battery can be administered in about 40 min (Roid, Miller, Pomplun, & Koch, 2013b). The Leiter-3 has a normative sample of 1603 and is representative of the U.S. census in 2008 in terms of sex, race, ethnicity, parental educational level, and geographic region. The sample population includes several disability categories including ID and developmental disabilities (Roid et al., 2013b).

Theoretical Orientation and Test Structure. The Leiter-3 is based on the CHC theory that encompasses a hierarchical structure (Roid et al., 2013b). The Leiter-3 includes four cognitive abilities in its test structure. Specifically, these cognitive abilities consist of Fluid Reasoning, Short-term Memory, Visual Processing, and Processing Speed. Both the Cognitive and Attention/Memory batteries contain five subtests that factor into the CHC broad cognitive abilities. The *Leiter-3 Manual* (Roid et al., 2013b) only provides g factor loadings for subtests. For ages 3 through 8, the average factor loadings on g for each subtest are as follows: Figure Grounds (0.66), Form Completion (0.69), Classification/Analogies (0.56), Sequential Order (0.56), and Visual Patterns (0.45). For ages 9 through 15, the average factor loadings on g for each subtest is as follows: Figure Grounds (0.70), Form Completion (0.77), Classification/Analogies (0.49), Sequential Order (0.65), and Visual Patterns (0.57). See the *Leiter-3 Examiner's Manual* (Roid et al., 2013b) for additional information regarding factor loadings on g for older age ranges.

Psychometric Properties. Internal reliability coefficients for the Nonverbal IQ Scale (r = 0.94–0.98) are more than adequate across all age groupings. The authors of this assessment tool also indicated acceptable stability across all subtests on both the Cognitive and Attention/Memory Batteries, except for the Stroop subtest (r = 0.42). Adequate criterion-related validity was noted with commonly used intelligence instruments that include both verbal and nonverbal domains (Roid et al., 2013b).

Diagnostic and Clinical Considerations

A number of factors are important to consider when selecting and interpreting intelligence tests used to evaluate children with ID. While adequate knowledge of an instrument's strengths and weakness (see Table 21.1) is important for selecting an appropriate measure, interpretation of the results requires clinical acumen that carefully considers factors that may limit the validity of the findings. Psychometric considerations that may impact the interpretation include factors related to characteristics of the norming sample. IQ estimates based on norm-referenced tests have been demonstrated to increase in relation to the age of the norming sample. In other words, as the norms get older, the mean IQ increases by several points. This psychometric phenomenon is referred to as the Flynn effect (Flynn, 1984, 1998). This factor is particularly important when selecting a test that has either been recently renormed or one that has not been revised for a number of years. Prior research exploring the

Table 21.1 Clinical implications associated with assessment instruments used to identify intellectual disabilities in children and adolescents

Assessment instrument	Strengths	Limitations
Stanford-Binet Intelligence Scales, Fifth Edition (SB5)	Early identification of developmental risk due to administration age starting at age 2 (Sattler, 2007) Low floors on most subtests allowing examiners to differentiate between subsets of low-functioning ability (Flanagan & Harrison, 2012) Appropriate normative sample of 119 individuals with ID (Roid, 2003c) Verbal and nonverbal formats for each factor measuring intelligence (g) allows for examiners to discern if language, visual, or motor impairment(s) are impacting intellectual functioning (Flanagan & Harrison, 2012)	Many subtests have a steep gradient between test items making it difficult to assess marginal progress, especially for children with low intellectual ability (Flanagan & Harrison, 2012) The SB5 has not been renormed for 15 years, which may result in a potential overestimation of one's intellectual ability related to the Flynn effect (Flynn, 1984)
Wechsler Intelligence Scale for Children, Fifth Edition (WISC-V)	Improved floors from previous edition, WISC-IV, permits examiners to identify various levels of low-functioning ability Appropriate normative sample of children and adolescents with both mild ($n = 74$) and moderate ($n = 37$) ID (Wechsler et al., 2014) Test-rest stability across subtests were reported for Mild and Moderate ID population (Canivez & Watkins, 2001; Wechsler et al., 2014) Nonverbal Index is useful to rule out a ID diagnosis in special populations such as English language learners, language impaired and autism spectrum disorder with language impairment (Mather & Wendling, 2014)	Motor demands are complex on Coding Subtest, which may underestimate a child's ability to process information Limited criterion validity studies on instruments outside of the Wechsler Scales (Sattler, 2007)
Woodcock-Johnson IV Tests of Cognitive Abilities (WJ IV COG)	Test items were added to improve floors and ceilings of this instrument (Reynolds & Niileksela, 2015) Appropriate normative sample of individuals with ID Opportunities for teaching and providing feedback on most subtests, may be useful to ensure understanding of task demands for individuals with ID (McGrew et al., 2014)	A few of the subtests (e.g., Passage Comprehension, Writing Samples, and Word Attack) have steep item gradients for younger children (McGrew et al., 2014) The technical manual does not indicate clearly whether an examiner should use the GIA or Gf-Gc composite when acquiring an estimate of general intellectual functioning. Further research is warranted to inform users of which method would be more appropriate for evaluations of intellectual disabilities (Reynolds & Niileksela, 2015)
Differential Ability Scales, Second Edition (DAS-II)	Appropriate sample population of 74 children and adolescents with mild or moderate intellectual functioning (Elliott, 2007a, 2007b) Extended norms on many of the subtests to better assess children who display low ability (Sattler, 2007) Higher internal consistency was noted for ID population ($r = 0.87–0.99$) (Elliott, 2007a, 2007b) Flexibility in using the Early Year Battery for children ages 7 through 9 with lower cognitive functioning (Flanagan & Harrison, 2012) Nonverbal composite is helpful to assess intellectual functioning in children with hearing and language impairments Test materials and manipulatives assist in retaining attention of examinees (Sattler, 2007)	The DAS-II has not been renormed for over 11 years, which may result in a potential overestimation of one's intellectual ability related to the Flynn effect (Flynn, 1984) Intellectual functioning (GCA) for ages 2 years 6 months through 3 years 5 months should be interpreted with caution since only four subtests comprise the Full-Scale IQ. The DAS-II should instead be used as a screener (Sattler, 2007)

(continued)

Table 21.1 (continued)

Assessment instrument	Strengths	Limitations
Kaufman Assessment Battery for Children, Second Edition (KABC-II)	This instrument was adequately normed on 42 children with a diagnosis of mild intellectual disability Low floors (e.g., as low as a standard score of 40 on scales) to sufficiently measure low levels of intellectual functioning There is flexibility in the response modalities (i.e., signing, writing) provided by examinees to more appropriately portray intellectual ability when individuals present with language impairments The additional nonverbal scale is intended for use to English language learners to rule out intellectual disability (Kaufman & Kaufman, 2004a, 2004b)	Complex directions can lead to difficulties with understanding task demands, especially for children with low intellectual functioning (Flanagan & Harrison, 2012)
Universal Nonverbal Intelligence Test-Second Edition (UNIT-2)	Adequate norming sample of individuals with intellectual disability High internal consistency ($r = 0.96–0.98$) across all composites and age groups of individuals with ID Subtests test items gradually increase in difficulty (Bracken & McCallum, 2016a, 2016b) This instrument is useful to rule out a ID diagnosis in special populations with communication and/or language deficits (Bracken & McCallum, 2016a, 2016b)	This instrument only measures nonverbal reasoning and memory skills Responses are limited to gestures, which can have an influence on children with severe motor impairment (Bracken & McCallum, 2016a, 2016b)
Leiter International Performance Scale-Third Edition (Leiter-3)	Adequate sample population of children and adolescents with low levels of intellectual functioning Manipulatives were redesigned in the Leiter 3 to accommodate individuals with cognitive and physical disabilities The manual discusses, in detail, adaptations for examinees with significant motor and/or communication deficits This instrument assist in measuring intellectual functioning in children with hearing and language impairments(Roid et al., 2013a, 2013b)	Due to the nonverbal nature of this instrument, verbal and communication abilities are not measured on this instrument. Further assessments measuring the above constructs are needed for a comprehensive evaluation of an individual's cognitive ability

impact of the Flynn effect on estimates of children with IQ scores in the deficient range have suggested that using a recently renormed instrument increased the percentage of children who would meet diagnostic criteria for ID by "nearly threefold" (Kanaya, Scullin, & Ceci, 2003, p. 785). These fluctuations in IQ estimates underscore the importance of using confidence intervals when interpreting test scores and considering multiple sources of data when rendering a diagnosis.

Regression to the mean is another psychometric phenomenon related to the norming sample that is important to consider in the diagnosis of ID. Regression to the mean occurs when standard scores occurring at the extreme ends of the normal curve are reassessed and demonstrate a tendency to become less extreme by moving closer to the mean. For example, an IQ score of 50 is significantly below the mean (more than 3 standard deviations) and relatively rare in that it occurs in less than 0.1% in a standard normal distribution. Extreme IQ scores, while relatively rare, are prone to move toward the mean upon reassessment. While IQ scores tend to be relatively stable over time, this phenomenon has been observed in prior research studies with ID children. Spitz (1983) found that the mean IQ scores of a group of children with ID to be 55 at age 13 and 58 at age 15. Clinically, this phenomenon becomes more important when scores are nearer to the threshold for diagnosing ID, e.g., standard scores of 70. Again, practitioners are well advised to consider this psychometric phenomenon when making clinical judgments in their work with this population.

Test construction also presents factors that must be considered in both the selection and interpretation of the results. More specifically, practitioners need to consider factors related to floor effects and test demands. Floor effects occur when an instrument does not have enough test items at the beginning of the subtest in order to yield a valid score. While issues related to floor effects are more likely to occur when evaluating younger children or individuals with lower-functioning ID (Flanagan & Alfonso, 1995), these effects limit the utility and validity of the findings. Similarly, factors related to test demands also play a role in valid interpretation of intelligence test results. For example, practitioners must take into consideration the impact the use of manipulatives (i.e., blocks) during a subtest may have on a child's performance who has motor delays or sensory deficits. In these situations, the findings are likely to underestimate intellectual ability and increase the likelihood of misdiagnosis.

It is also critically important to consider language and cultural differences as a factor that may bias test results. This goes beyond simply ensuring that the instrument has a representative norming sample. It includes considering factors that may influence the meaning assigned to information gathered from the assessment. As such, language and cultural factors are not specific to the assessment of intelligence, rather they pertain to all data collection methods being used as part of the assessment. Competency in this area requires knowledge of how language and cultural factors influence particular aspects of the assessment process in order to reduce the potential for bias. While a comprehensive overview of this topic is beyond the scope of this chapter, we will highlight those aspects most germane to the assessment of intelligence.

Research in this area that has examined the influence of language and cultural factors on psychometric properties of tests has suggested that the reliability of the instruments used are less susceptible to bias than the validity of their findings (Reynolds & Carson, 2005). More specifically, the validity of these findings is influenced most by the developmental language proficiency and the acculturative knowledge acquisition of the examinee (Ortiz, Ochoa, & Dynda, 2012). According to Ortiz (2014), "it is only the degree to which an individual possesses both age- or grade-related proficiency in the language in which the test is being administered, as well as the child's equivalent levels of opportunity for acquiring the culture-specific knowledge contained or expected by the test (as compared with individuals on whom the test was normed), that directly and profoundly affect test performance" (p. 63). While these factors are more apparent among individuals who have recently emigrated from other countries, it is relevant to note that these differences have also been observed in populations that are native to the United States, such as Native Americans (Coutinho & Oswald, 2000; Nakano & Watkins, 2013). Given the potential for obtaining lower IQ estimates due to language or cultural differences, practitioners working with diverse populations need to be cognizant of these factors when selecting and interpreting intelligence tests being used to make a diagnosis of ID.

The last factor that will be discussed relates to score variability. As noted previously, intellectual ability is typically represented by a single score or IQ estimate. In certain instances, unusual variability among the scores that make up the IQ is observed. While some score variability is relatively common in general practice, an unusual difference noted between index or subtest scores makes summarizing intellectual ability as a single score potentially misleading (Donnell, Belanger, & Vanderploeg, 2011; Hale & Fiorello, 2001). The term unusual difference refers to differences that are both statistically significant and occur rarely in the population. As a rule of thumb, differences that are both statistically significant and occur in 10% or less of the standardization sample are considered unusual (Sattler, 2007). When the IQ score is not unitary or "best measure" of intellectual ability due to unusual differences among scores, there is no clear-cut answer as to which composite score should be used in its place. The decision depends on a number of factors, some of which have already been reviewed.

The first consideration should be the instruments factor structure. Using information concerning the test's factor structure as a guide, the practitioner can identify which composite scores account for the most variance in the measurement of general intelligence. For example, in the majority of intelligence tests reviewed, measures of fluid reasoning and crystallized intelligence accounted for the preponderance of variance in the full-scale score. Therefore, examinees who present with low average scores on a particular composite such as in fluid reasoning (e.g., a standard score of 82) may not have an ID even if their full-scale IQ score was 70 due to below average scores on the other composites. Detailed information on the factor structures for the tests reviewed in this chapter has been provided for your reference. Second, prudent evidence-based practice suggests that a theoretical rationale also be used to guide decision-making (Hale & Fiorello, 2001). While the instruments reviewed vary in terms of their theoretical orientation, CHC theory is likely to provide the best option for test interpretation of scores across all of the measures reviewed (see Flanagan et al., 2013). From a theoretical standpoint, a particular pattern of score variably may be explored in order to arrive at a final diagnosis. As an example, practitioners may not consider near average processing speed to be as central to the final diagnosis as near average fluid reasoning.

Conclusions

The advancement of intelligence testing over time has played an instrumental role in shaping how ID is defined and assessed. The use of theoretical models to guide test development has become a more universally accepted practice. Nonetheless, continued research efforts are needed in order to better understand how these models will inform clinical practice and intervention planning. Throughout history, improvements in our knowledge of this construct have continued to enhance the instruments we use to measure intelligence as well as how we interpret their findings.

Selecting, using, and interpreting intellectual assessments is a multifaceted endeavor. As illustrated throughout this chapter, ID evaluations require practitioners to reflect on a number of clinical considerations. These considerations not only assist practitioners in selecting an assessment tool that is likely to yield a valid measure of intellectual functioning, but also ensure that the practitioner is using evidence-based decision-making when interpreting the findings. In closing, it is important to keep in mind that the assessment process should seldom rely solely on measurements of intellectual and adaptive functioning alone. Rather, best practice suggests that multiple methods of assessment including background information, developmental history, behavioral observations, and academic achievement should also be considered. By drawing upon multiple sources of information, the likelihood that the results will contribute to accurate diagnostic conclusions and meaningful intervention planning are increased.

References

American Psychiatric Association. (2013). Diagnostic and statistical manual of mental disorders (5th ed.). Washington, DC: Author.

Anastasi, A. (1942). Review: The measurement of adult intelligence by David Wechsler. *American Journal of Psychology, 55*, 608–609.

Armstrong, K., Hangauer, J., & Nadeau, J. (2012). Use of intelligence tests in the identification of children with intellectual and developmental disabilities. In D. P. Flanagan & P. L. Harrison (Eds.), *Contemporary intellectual assessment: Theories, tests, and issues* (3rd ed., pp. 726–736). New York: The Guilford Press.

Becker, K. A. (2003). *History of the Stanford-Binet Intelligence Scales: Content and psychometrics. (Stanford-Binet Intelligence Scales, Fifth Edition Assessment Service Bulletin No. 1)*. Ithaca, IL: Riverside Publishing.

Binet, A. (1916). New investigations upon the measure of the intellectual level among school children. In H. H. Goddard (Ed.), *The development of intelligence in children (the Binet-Simon Scale)*. (E. S. Kite, Trans; pp. 274-329). Baltimore, MD: Williams & Wilkins. (Original work published in 1911).

Binet, A., & Simon, T. (1916). Application of the new methods to the diagnosis of the intellectual level among normal and subnormal children in institutions and in the primary schools. In H. H. Goddard (Ed.), *The development of intelligence in children (the Binet-Simon Scale)*. (E. S. Kite, Trans; pp. 91–181). Baltimore: Williams & Wilkins. (Original work published in 1905).

Bracken, B. A., & McCallum, R. S. (2016a). *Universal nonverbal intelligence test, second edition*. Austin, TX: Pro-Ed.

Bracken, B. A., & McCallum, R. S. (2016b). *Examiner's manual. Universal nonverbal intelligence test, second edition*. Austin, TX: Pro-Ed.

Brown, I. (2007). What is meant by developmental disability. In I. Brown & M. Percy (Eds.), *A comprehensive guide to intellectual and developmental disabilities* (pp. 3–15). Baltimore, MD: Brookes.

Canivez, G. L., & Watkins, M. W. (2001). Long-term stability of the Wechsler Intelligence Scale for Children-Third Edition among students with disabilities. *School Psychology Review, 30*, 438–453.

Carroll, J. B. (1993). *Human cognitive abilities: A survey of factor-analytic studies*. New York: Cambridge University Press.

Carson, J. (2007). *The measure of merit: Talents, intelligence, and inequality in the French and American republics, 1750–1940*. Princeton, NJ: Princeton University Press.

Cattell, J. M. (1890). *Mental tests and measurements. Mind* (Vol. 15, pp. 373–381).

Cattell, R. B. (1963). Theory of fluid and crystalized intelligence: A critical experiment. *Journal of Educational Psychology, 54*, 1–22.

Cattell, R. B. (1998). Where is intelligence?: Some answers from the triadic theory. In J. J. McArdle & R. W. Woodcock (Eds.), *Human cognitive abilities in theory and practice* (pp. 29–38). Mahwah, NJ: Erlbaum.

Coutinho, M. J., & Oswald, D. P. (2000). Disproportionate representation in special education: A synthesis and recommendations. *Journal of Child and Family Studies, 9*, 135–156.

Donnell, A., Belanger, H., & Vanderploeg, R. (2011). Implications of psychometric measurement for neuropsychological interpretation. *The Clinical Neuropsychologist, 25*(7), 1097–1118.

Elliott, C. D. (2007a). *Differential ability scales, second edition*. San Antonio, TX: Harcourt Assessment, Inc.

Elliott, C. D. (2007b). *Introductory and technical handbook. Differential ability scales, second edition*. San Antonio, TX: Harcourt Assessment, Inc.

Elliott, C. D. (2012). The differential Ability Scales-Second Edition. In D. P. Flanagan & P. L. Harrison (Eds.), *Contemporary intellectual assessment: Theories, tests, and issues* (3rd ed., pp. 336–356). New York: The Guilford Press.

Flanagan, D., & Alfonso, V. (1995). A critical review of the technical characteristics of new and recently revised intelligence tests for preschool children. *Journal of Psychoeducational Assessment, 13*, 66–90.

Flanagan, D. P., Fiorello, C. A., & Ortiz, S. O. (2010). Enhancing practice through application of Cattell-Carroll theory and research: A "third method" approach to specific learning disability identification. *Psychology in the Schools, 47*, 739–760.

Flanagan, D. P., Ortiz, S. O., & Alfonso, V. C. (2013). *Essentials of cross-battery assessment* (3rd ed.). Hoboken, NJ: Wiley.

Flynn, J. R. (1984). The mean IQ of Americans: Massive gains 1932 to 1978. *Psychological Bulletin, 95*, 29–51.

Flynn, J. R. (1998). WAIS-III and WISC-III: IQ gains in the United States from 1972 to 1995: How to compensate for obsolete norms. *Perceptual and Motor Skills, 86*, 1231–1239.

Galton, F. (1890). Why do we measure mankind? *Lippincott's Monthly Magazine, 45*, 236–241.

Gottfredson, L. S., & Saklofske, D. H. (2009). Intelligence: Foundation and issues in assessment. *Canadian Psychology, 50*, 183–195.

Hale, J. B., & Fiorello, C. A. (2001). Beyond the academic rhetoric of g: Intelligence testing guidelines for practitioners. *The School Psychologist, 55*, 113–139.

Horn, J. L., & Cattell, R. B. (1966). Refinement and test of the theory of fluid and crystallized general intelligence. *Journal of Educational Psychology, 57*, 253–270.

Horn, J. L., & Noll, J. (1997). Human cognitive capabilities: Gf-Gc theory. In D. P. Flanagan, J. L. Genshaft, & P. L. Harrison (Eds.), *Contemporary intellectual assessment: Theories, tests, and issues* (pp. 53–91). New York: Guilford Press.

Jenkins, J. J., & Paterson, D. G. (1961). *Studies in individual differences*. New York: Appleton-Century-Crofts.

Jensen, A. R. (1980). *Bias in mental testing*. New York, NY: The Free Press.

Kanaya, T., Scullin, M. H., & Ceci, S. J. (2003). The Flynn effect and U. S. policies: The impact of rising IQ scores on American society via mental retardation diagnoses. *American Psychologist, 58*, 778–790.

Kaufman, A. S., & Kaufman, N. L. (2004a). *Kaufman assessment battery for children* (2nd ed.). Bloomington, MN: Pearson.

Kaufman, A. S., & Kaufman, N. L. (2004b). *Kaufman assessment battery for children (2nd E.), manual*. Bloomington, MN: Pearson.

Keith, T. Z., & Reynolds, M. R. (2010). Cattell-Horn-Carroll abilities and cognitive tests: What we've learned form 20 years of research. *Psychology in the Schools, 47*, 635–650.

Louttit, C. M., & Browne, C. G. (1947). The use of psychometric instruments in psychological clinics. *Journal of Consulting Psychology, 11*, 49–54.

Luria, A. R. (1966). *Human brain and psychological processes*. New York: Harper & Row.

Mather, N., & Wendling, B. J. (2014). Examiner's Manual. In *Woodcock-Johnson IV tests of cognitive abilities*. Rolling Meadows, IL: Riverside.

McGrew, K. S. (1997). Analysis of the major intelligence batteries according to a proposed comprehensive Gf-Gc framework. In D. P. Flanagan, J. L. Genshaft, & P. L. Harrison (Eds.), *Contemporary intellectual assessment* (pp. 151–179). New York: Guilford Press.

McGrew, K. S. (2005). Editorial: CHC theory and the human cognitive abilities project: Standing on the shoulders of the giants of psychometric intelligence research. *Intelligence, 37*, 1–10.

McGrew, K. S., LaForte, E. M., & Schrank, F. A. (2014). Technical manual. In *Woodcock-Johnson IV*. Rolling Meadows, IL: Riverside.

McGrew, K. S., & Wendling, B. J. (2010). Cattell-Horn-Carroll cognitive-achievement relations: What we have learned from the past 20 years of research. *Psychology in the Schools, 47*, 651–675.

Nakano, S., & Watkins, M. W. (2013). Factor structure of the Wechsler Intelligence Scales for Children-Fourth Edition among referred Native American students. *Psychology in the Schools, 50*, 957–968.

Newton, J. H., & McGrew, K. S. (2010). Introduction to the special issue: Current research in Cattell-Horn-Carroll-based assessment. *Psychology in the Schools, 47*, 621–634.

Niileksela, C. R., & Reynolds, M. R. (2014). Global, broad, or specific cognitive differences? Using MIMIC Model to examine differences in CHC abilities in children with learning disabilities. *Journal of Learning Disabilities, 47*, 224–236.

Oakland, T., Douglas, S., & Kane, H. (2016). Top ten standardized tests used internationally with children and youth by school psychologists in 64 countries: A 24-year follow-up study. *Journal of Psychoeducational Assessment, 34*, 166–176.

Ortiz, S. O. (2014). Best practices in nondiscriminatory assessment. In P. L. Harrison & A. Thomas (Eds.), *Best practices in school psychology: Foundations*. Bethesda, MD: NASP.

Ortiz, S. O., Ochoa, H. S., & Dynda, A. M. (2012). Testing with culturally and linguistically diverse populations: Moving beyond the verbal-performance dichotomy into evidence-based practice. In D. P. Flanagan & P. L. Harrison (Eds.), *Contemporary intellectual assessment: Theories, tests, and issues* (3rd ed., pp. 526–552). New York: The Guilford Press.

Proctor, B. (2012). Relationships between Cattell-Horn-Carroll (CHC) cognitive abilities and math achievement within a sample of college students with learning disabilities. *Journal of Learning Disabilities, 45*, 278–287.

Rabin, A. I., & Guertin, W. H. (1951). Research with the Wechsler-Bellevue test: 1945-1950. *Psychological Bulletin, 48*, 211–248.

Reynolds, C. R., & Carson, A. D. (2005). Methods for assessing cultural bias in tests. In C. Frisby & C. R. Reynolds (Eds.), *Comprehensive handbook of multicultural school psychology* (pp. 795–823). Hoboken, NJ: Wiley.

Reynolds, M. R., & Niileksela, C. R. (2015). Test review: Woodcock-Johnson IV test of cognitive abilities. *Journal of Psychoeducational Assessment, 33*, 381–390.

Roid, G. H. (2003a). *Stanford-Binet Intelligence Scales, Fifth Edition*. Itasca, IL: Riverside Publishing.

Roid, G. H. (2003b). *Stanford-Binet Intelligence Scales, Fifth Edition, Examiner's Manual*. Itasca, IL: Riverside Publishing.

Roid, G. H. (2003c). *Stanford-Binet Intelligence Scales, Fifth Edition, Techincal Manual*. Itasca, IL: Riverside Publishing.

Roid, G. H., Miller, L. J., Pomplun, M., & Koch, C. (2013a). *Leiter International Performance Scale-Third Edition*. Wood Dale, IL: Stoelting Company.

Roid, G. H., Miller, L. J., Pomplun, M., & Koch, C. (2013b). *Leiter International Performance Scale-Third Edition, Manual*. Wood Dale, IL: Stoelting Company.

Sattler, J. M. (2007). *Assessment of children: Cognition foundations* (5th ed.). San Diego, CA: Jerone M. Sattler Publisher, Inc.

Schalock, R. L., Borthwick-Duffy, S. A., Bradley, V. J., Buntinx, W. H. E., Coulter, D. L., Craig, E. M., … Yeager, M. H. (2010). *Intellectual disability: Definition, classification, and systems of supports* (11th ed.). Washington, DC: American Association on Intellectual and Developmental Disabilities.

Schalock, R. L., Luckasson, R., & Shogren, K. A. (2007). The renaming of mental retardation: Understanding the change to the term intellectual disability. *Intellectual and Developmental Disabilities, 45*, 116–124.

Schneider, W. J., & McGrew, K. S. (2012). The Cattell-Horn-Carroll Model of Intelligence. In D. P. Flanagan & P. L. Harrison (Eds.), *Contemporary intellectual assessment: Theories, tests, and issues* (3rd ed., pp. 99–144). New York: The Guilford Press.

Schrank, F. A., McGrew, K. S., & Mather, N. (2014). *Woodcock-Johnson IV Tests of Cognitive Abilities*. Rolling Meadows, IL: Riverside.

Singer, J. K., Lichtenberger, E. O., Kaufman, J. C., Kaufman, A. S., & Kaufman, N. L. (2012). The Kaufman Assessment Battery for Children-Second Edition and the Kaufman Test of Educational Achievement-Second Edition. In D. P. Flanagan & P. L. Harrison (Eds.), *Contemporary intellectual assessment: Theories, tests, and issues* (3rd ed., pp. 269–296). New York: The Guilford Press.

Spearman, C. (1904). "General intelligence," objectively determined and measured. *American Journal of Psychology, 15*, 201–293.

Spitz, H. H. (1983). Critique of the developmental position in mental-retardation research. *The Journal of Special Education, 17*, 262–294.

Tassé, M. J., Luckasson, R., & Schalock, R. L. (2016). The relation between intellectual functioning and adaptive behavior in the diagnosis of intellectual disability. *Intellectual and Developmental Disabilities, 54*, 381–390.

Terman, L. M. (1916). *The measurement of intelligence: An explanation of and a complete guide for the use of the Stanford revision and extension of the Binet-Simon Intelligence Scale*. Boston, MA: Houghton Mifflin.

Thorndike, R. L., Hagen, E. P., & Sattler, J. M. (1986). *The Stanford-Binet Intelligence Scale-Fourth edition: Technical manual*. Itasca, IL: Riverside.

Thurstone, L. L. (1938). *Primary mental abilities*. Chicago, IL: University of Chicago Press.

Tuddenham, R. D. (1962). The nature and measurement of intelligence. In L. J. Postman (Ed.), *Psychology in the making* (pp. 469–525). New York: Knopf.

Vernon, P. E. (1950). *The structure of human abilities*. London: Methuen.

Walker, W., & Johnson, C. (2006). Mental retardation: Overview and diagnosis. *Pediatrics Review, 27*, 204–212.

Wasserman, J. D. (2012). A history of intelligence assessment: The unfinished tapestry. In D. P. Flanagan & P. L. Harrison (Eds.), *Contemporary intellectual assessment: Theories, tests, and issues* (3rd ed., pp. 3–55). New York: The Guilford Press.

Wechsler, D. (1939). *The measurement of adult intelligence*. Baltimore, MD: Williams & Wilkins.

Wechsler, D. (1946). *The Wechsler-Bellevue Intelligence Scale, Form II. Manual for administering and scoring the test*. New York: Psychological Corporation.

Wechsler, D. (1949). *The Wechsler Intelligence Scale for Children (WISC)*. New York: Psychological Corporation.

Wechsler, D. (1955). *The Wechsler Adult Intelligence Scale (WAIS)*. New York: Psychological Corporation.

Wechsler, D. (1967). *The Wechsler Preschool and Primary Scale of Intelligence (WPPSI)*. New York: Psychological Corporation.

Wechsler, D. (2014). *Wechsler intelligence scale for children* (5th ed.). Bloomington, MN: Pearson.

Wechsler, D., Raiford, S. E., & Holdnack, J. A. (2014). Technical and Interpretive Manual. In *Wechsler Intelligence Scale for Children, Fifth Edition*. Bloomington, MN: Pearson.

Wolf, T. (1969). The emergence of Binet's conceptions and measurement of intelligence: II. A case history of the creative process. *Journal of the History of the Behavioral Sciences, 5*, 207–237.

Wolf, T. (1973). *Alfred Binet*. Chicago, IL: University of Chicago Press.

Woodcock, R. W., McGrew, K. S., & Mather, N. (2001). *Woodcock-Johnson III tests of cognitive abilities*. Itasca, IL: Riverside Publishing.

Yerkes, R. M. (1919). Report of the Psychology Committee of the National Research Council. *Psychological Review, 26*, 83–149.

Yoakum, C. S., & Yerkes, R. M. (1920). *Army mental tests*. New York: Holt.

Zenderland, L. (1998). *Measuring minds: Henry Herbert Goddard and the origins of American intelligence testing*. New York: Cambridge University Press.

22

Early Assessment and Tests for Adaptive Behavior

Esther Hong and Johnny L. Matson

Introduction

Individuals with intellectual disability (ID) experience deficits in intellectual and adaptive functioning across conceptual, social, and practical domains. Standardized measures of developmental and adaptive behavior have been designed to assess an individual's level of functioning across various skill domains. These measures screen for specific skill deficits and aid in the diagnostic process of ID or other developmental disabilities. The results from developmental and adaptive tests may assist parents, practitioners, and educators in making decisions regarding eligibility of services, types of services required, and level of support a child requires to function across various settings. Additionally, developmental and adaptive behavior scales can be used to monitor treatment progress and measure intervention outcomes. This chapter discusses the implications for early childhood assessment and reviews several commonly used developmental and adaptive behavior scales.

Intellectual Disability

Intellectual disability (ID) is a disorder characterized by deficits in both intellectual functioning and adaptive behavior (American Psychiatric Association [APA], 2013; American Association on Intellectual and Developmental Disabilities Association [AAIDD], 2010). Intellectual functioning is typically assessed using standardized intelligence tests, with an Intelligence Quotient (IQ) below 70 indicating impairment in intellectual functioning. However, in recent years, the construct of "intelligence" has shifted from the measurement of an individual's IQ to as a measure of an individual's adaptive functioning. As a result, the focus of a diagnosis of ID is on an individual's level of impairment in adaptive functioning rather than intellectual functioning.

Adaptive behavior is defined as "the collection of conceptual, social, and practical skills that have been learned by people in order to function in their everyday lives" (Schalock et al., 2010). Individuals with impairment in adaptive functioning demonstrate deficits across all three skill domains. Conceptual skills include skills related to academics, language, numbers, time, money, executive functioning, memory, and abstract thinking. Skills that fall under the social domain include social communication, interpersonal skills, emotion regulation, social cues, and social problem solving. The practical domain includes

E. Hong (✉) · J. L. Matson
Department of Psychology, Louisiana State University, Baton Rouge, LA, USA
e-mail: ehong1@lsu.edu

J. L. Matson (ed.), *Handbook of Intellectual Disabilities*, Autism and Child Psychopathology Series, https://doi.org/10.1007/978-3-030-20843-1_22

activities of daily living (e.g., eating, dressing, toileting, hygiene), occupational skills, recreational skills, healthcare, safety, and vocational skills. Standardized tests of adaptive behavior are used to assess deficits in adaptive functioning.

In accordance with the Diagnostic and Statistical Manual of Mental Disorders (DSM-5; APA, 2013), the severity level of ID is determined based on the individual's overall adaptive functioning, not IQ. The severity level (i.e., mild, moderate, severe, or profound) corresponds to the level of supports that an individual requires to adequately function in daily activities, across environments. By definition, individuals with ID fail to meet the "developmental and socio-cultural standards for personal independence and social responsibility." As a result, examiners should consider cultural differences as well as the individual's community, school, and work environments during the diagnostic process.

The prevalence of ID is approximately 1% (Maulik, Mascarenhas, Mathers, Dua, & Saxena, 2011), with higher rates of ID found in low and middle-income populations. Of the individuals diagnosed with ID, the majority experience mild ID (85%), followed by moderate ID (10%), severe ID (4%), and profound ID (2%; King, Toth, Hodapp, & Dykens, 2009). The high prevalence of ID is associated with high costs, as healthcare providers, schools, and public entities provide resources and support services to individuals with ID across the lifespan. However, these costs can be mitigated by early assessment and early intervention of ID and developmental disabilities.

Early Childhood Assessment

To meet the diagnostic criteria for ID, the DSM-5 specifies that the onset of deficits in intellectual and adaptive functioning occur during the developmental period. Individuals with ID exhibit intellectual and adaptive impairments at an early age, and these impairments persist throughout their lifespan. Given the early onset of ID, the early detection and early intervention of adaptive skills is critical in improving intervention outcomes and quality of life.

Several studies have demonstrated that early intervention is effective in improving outcomes of toddlers and young children with ID and other developmental disabilities (Lovaas, 1987; Sharav & Shlomo, 1986; Smith, Groen, & Wynn, 2000). While there is heterogeneity in treatment outcomes (due to child-specific and treatment-specific factors), there is a general consensus that early intervention is effective in remediating developmental delays and reducing the developmental gap between children with ID and their typically developing peers (Guralnick, 2005). When children learn adaptive skills during the developmental period, their outcomes improve (Maulik et al., 2011).

Given the evidence-based research on early intervention and mandated early intervention services for preschool aged children (Individuals with Disabilities Education Improvement Act of 2004 [IDEIA]), there has been an increased research focus on the development and evaluation of early assessment tools to measure developmental and adaptive functioning. There is a need for standardized, valid behavior scales that accurately assess and diagnose young children with ID and other developmental disabilities. Further, the assessment measures should be sensitive enough to correctly categorized children into the different severity levels of ID.

These screening tools and behavior scales are crucial to the diagnostic process because they are objective measures of how infants, toddlers, and young children are meeting their developmental milestones. The scales capture the child's distinct abilities (i.e., strengths and weaknesses) across various skill domains, which help guide decisions regarding eligibility for special education services, classroom accommodations, support services, and intervention planning. Normative data gathered from developmental and adaptive scales also provide insight into how the child is developing in relation to their typically peers.

Developmental Scales

The assessment of developmental functioning provides valuable information on the child's current level of functioning across various developmental domains, such as language, communication, social, motor, learning ability, feeding, dressing, and toileting. The developmental scales that are discussed in this chapter have been developed in accordance with the existing literature on child development. Rather than focusing a specific child development theory, these developmental measures were designed based on the broad theoretical foundation of child development (Albers & Grieve, 2007). While these measures are used during the initial diagnostic process, they should also be administered periodically, in order to monitor treatment progress and determine if adjustments to the intervention program need to be made.

Bayley Scales of Infant and Toddler Development, Third Edition (Bayley-III)

The *Bayley Sales of Infant and Toddler Development* (*Bayley-III*; Bayley, 2006) is a comprehensive measure designed to assess the developmental functioning of infants and toddlers between the ages of 1 and 42 months. Test administration ranges from approximately 50–90 min, with testing time increasing for older children. The Bayley-III comprises five scales: Cognitive, Language (Receptive and Expressive), Motor (Fine and Gross), Social-Emotional, and Adaptive Behavior (Conceptual, Social, and Practical).

For the Cognitive, Language, and Motor domains, the examiner directly administers the items to the child. Based on the child's inability or ability to complete the tasks, the items are scored as "0" (no credit) or "1" (credit), respectively. The Cognitive scale assess for aspects of cognitive processing such as sensorimotor development, memory, object permanence, concept formation, and object relatedness. The Language scale is composed of the Receptive and Expressive Communication subtests. The Receptive Communication subscale assesses the child's ability to understand and respond to verbal stimuli and identify pictures and objects. The Expressive Communication subscale assesses for skills such as babbling, gesturing, turn taking, and naming objects and pictures. The Motor scale consists of Fine and Gross motor subscales. The Fine Motor subscale assesses for perceptual-motor integration, motor planning, motor speed, visual tracking, grasping, and functional hand skills, while the Gross Motor subscale assesses for body positioning, dynamic movement (e.g., walking, crawling, jumping, running), balance, motor planning, and perceptual-motor integration.

The evaluations for the Social-Emotional and Adaptive Behavior scales are completed by the child's primary caregiver, using parent questionnaires. The Social-Emotional scale consists of 35 items, which measure the child's social-emotional development and functioning based on his or her current age. Items are rated on a 6-point Likert scale: "0"—can't tell, "1"—none of the time, "2"—some of the time, "3"—half of the time, "4"—most of the time, or "5"—all of the time. The Adaptive Behavior scale is the broadest of the five scales and assess for the child's level of functioning across several skill areas: communication, community use, health and safety, leisure, self-care, self-direction, functional pre-academics, home living, social skills, and motor skills. Items are rated on a 4-point Likert scale: "0"—is not able, "1"—never when needed, "2"—sometimes when needed, or "3"—always when needed.

The Bayley-III has been found to have high internal consistency reliability coefficients across scales and subscales, ranging from 0.86 to 0.93. The validity of the Bayley-III was evaluated through several special group studies, which included children with Down syndrome, pervasive developmental disorders, cerebral palsy, language impairment, risk for developmental delays, prenatal alcohol exposure, and premature or low birth weight.

Bayley Scales of Infant and Toddler Development, Third Edition (Bayley-III Screening Test)

The *Bayley Scales of Infant and Toddler Development, Third Edition (Bayley-III Screening Test*; Bayley, 2006) is a screening tool that was designed to quickly assess the developmental functioning of infants and toddlers between the age of 1 and 42 months. The Bayley-III Screening Test can be administered in approximately 15–25 min, depending on the age of the child. The Bayley-III Screening test consists of three scales: Cognitive, Language (Receptive and Expressive Communication), and Motor (Fine and Gross Motor).

The test items were selected from the Bayley-III, to include the main components of the Bayley-III. As with the Bayley-III, the examiner directly administers the items to the child, and based on the child's response, items are scored as "0"—no credit or "1"—credit. The scores are calculated to assesses the level of risk the child has for developing a developmental delay. The risk categories include: At Risk, Emerging, or Competent. Scores falling within the "At Risk" range indicate that the child is at a high risk for a developmental delay, scores in the "Emerging" range indicate that the child has some risk for a developmental delay, and scores in the "Competent" range indicate that the child is at low risk for a developmental delay.

Mullen Scales of Early Learning: AGS Edition (MSEL:AGS)

The *Mullen Scales of Early Learning: AGS Edition* (Mullen, 1995) assesses for cognitive functioning in infants and young children from birth to 68 months. Test administration ranges from 15 min for children around 1 year old, 25–35 min for children around 3 years old, and 40–60 min for children around 5 years old. The MSEL:AGS comprises four cognitive scales (i.e., Fine Motor, Visual Reception, Expressive Language, and Receptive Language) and one Gross Motor scale.

The Fine Motor scale assesses the child's ability to manipulate small objects and demonstrate fine motor control and coordination. The Visual Reception scale assesses for visual processing, visual discrimination, and visual memory skills. The Expressive Language scale assesses the child's verbal response to items, verbal utterances, and concept formation (Dumont, Cruse, Alfonso, & Levine, 2000). The Receptive Language scale assesses the child's ability to respond to verbal input. Prerequisite skills are required for the child to complete to the Receptive Language scale items, as some items require the child to point or respond using one to three-word verbalizations. The Gross Motor subscale assesses a range of gross motor skills (e.g., walking, walking up the stairs) in children between the ages of 0 and 33 months. Most items on the MSEL:AGS are scored based on the examiner's observation of how the child performs a task. The point scale varies per item, from 0–1, 0–2, 0–3, 0–4, and 0–5. The scores on the four cognitive scales are summed together to provide the Early Learning Composite (ELC), which is a measure of general intelligence.

The purpose of the MSEL:AGS is to identify the strengths and weaknesses of the child and assess for early intellectual development, with percentile ranks and age equivalents provided for each of the scales. The MSEL:AGS was designed for educational purposes and can help assess a child's level of school readiness and determine the child's eligibility for intervention and/or special education services. Further, the MSEL:AGS may assist service providers and clinicians with individualized program planning (Dumont et al., 2000).

The internal reliability of the MSEL:AGS is adequate, with internal consistency reliability coefficients ranging from 0.83 to 0.95. However, the validity of the measure is questionable, as children from certain ethnic groups (i.e., African Americans, Hispanics) and socioeconomic statuses (i.e., rural communities, parental occupation) were underrepresented in the standardization sample. Further, the standardization data were collected across two different time periods. As such, the results from the MSEL:AGS should be interpreted with caution (Dumont et al., 2000). The test manual recommends that the MSEL be used in conjunction with other measures of social-emotional functioning (i.e., Vineland Social-Emotional Early Childhood Scales) in order to complete a comprehensive early childhood evaluation.

Battelle Developmental Inventory, Second Edition (BDI-2)

The *Battelle Developmental Inventory, Second Edition (BDI-2;* Newborg, 2005*)* is a comprehensive, early childhood assessment for children from birth to 7 years, 11 months of age. The BDI-2 comprises 450 items and takes 60–90 min to administer, depending on the age of the child. The BDI-2 assesses a child's strengths and weaknesses across five global developmental domains, including Personal-Social, Adaptive, Motor, Communication, and Cognitive functioning. Each domain comprises several subdomains. The Personal-Social domain includes adult interaction, peer interaction, and self-concept and social role subdomains. The Adaptive domain includes the self-care and personal responsibility subdomains. The Motor domain includes gross motor, fine motor, and perceptual motor subdomains. The Communication domain includes receptive and expressive communication subdomains. The Cognitive domain consists of attention and memory, reasoning and academic skills, and perception and concepts subdomains. Not all subdomains are administered to each child, as the subdomains are administered according to the child's current age.

Multiple methods of assessment may be used, including structured play-based activities with manipulatives, direct behavioral observations of the child, and parent/caregiver interviews. The test items are scored as either: "0"—no ability, "1"—emerging ability, or "2"—ability present. The subdomain scores are summed to provide a total domain score, and the five domain scores are calculated to provide the BDI-2 total score. The results of the BDI-2 can help determine school readiness and supplemental services.

The BDI-2 has excellent internal consistency, with reliability coefficients for BDI-2 total score ranging from 0.98 to 0.99 (Bliss, 2007). The reliability coefficients for the domain scores were also excellent; however, the Adaptive domain reliability coefficient was the lowest, at 0.80.

The BDI-2 has been found to be valid in distinguishing children with various diagnoses, such as autism spectrum disorders (ASD), developmental delays, motor delays, and speech and language delays (Bliss, 2007; Sipes, Matson, & Turygin, 2011).

Battelle Developmental Inventory Screening Test

The *Battelle Developmental Inventory Screening Test* (Newborg, 2005) is an abbreviated version of the BDI-2 and takes 10–30 min to administer. The BDI Screening Test is used to screen for deficits in developmental functioning and determine if further comprehensive assessments are needed. The BDI Screening Test comprises the same developmental domains and subdomains assessed by the BDI-2. The BDI Screening Test includes 96 items that had the highest correlation with the BDI-2 total score, with factor loading values of 0.70 and above (Bliss, 2007). As with the BDI-2, multiple methods of assessment are used, such as direct administration of the items to the child, behavioral observations of the child, or parent/caregiver interviews. Items of the BDI Screening Test are rated on the same 3-point Likert scale as the BDI-2 items. The results of the BDI Screening Test can be used to determine children who are at risk for developmental disabilities and provide an overview of the child's current level of developmental functioning.

Ages and Stages Questionnaires: A Parent-Completed Child Monitoring System, Third Edition (ASQ-3)

The *Ages and Stages Questionnaires: A Parent-Completed Child Monitoring System, Third Edition (ASQ-3;* Squires et al., 2009*)* is a screening tool that is completed by the primary caregiver. The ASQ-3 was designed to screen for developmental delays in young children ages 1–66 months, across five skill domains: Communication, Gross Motor, Fine Motor, Problem Solving, and Personal-Social. The ASQ-3 comprises 21 questionnaires, and the questionnaires are administered at designated age intervals. Each questionnaire consists of 30 items, 6 items per each of the five developmental domains. The primary caregiver rates whether their child is: able to complete the task, sometimes able to complete the task, or not yet able to complete the task.

Based on the caregiver's responses, the examiner calculates the total score for each domain. The examiner then compares the child's scores to the cutoff scores. Scores that are above the cutoff score indicate typical development; scores at or close to the cutoff score indicate that the child is functioning at 1 or 2 standard deviations below the mean and may be at risk for developmental disabilities; scores that are below the cutoff score indicate that the child is performing at 2 or more standard deviations below the mean. Children that are below the cutoff score may need further comprehensive assessment.

Reliability of the ASQ-3 was found to be adequate, with internal consistency ranging from 0.51 to 0.87 and test-retest reliability ranging from 75 to 0.82. Test validity has also been established, with high sensitivity (82.5–89.2%) and specificity (77.9–92.1%). The ASQ-3 is an evidence-based screening tool that can be used to gather comprehensive information regarding developmental functioning as well as monitor intervention progress.

Developmental Assessment of Young Children, Second Edition (DAYC-2)

The *Developmental Assessment of Young Children-Second Edition (DAYC-2;* Voress & Maddox, 2013) is a developmental measure designed to assess for developmental delays in infants and toddlers from birth to 5 years, 11 months. The DAYC-2 was originally developed in accordance with the guidelines of as specified in the federal legislation for children with disabilities (i.e., IDEIA). General developmental functioning of the following five domains are measured: Cognition, Communication, Social-Emotional Development, Physical Development, and Adaptive Behavior. If a specific skill area needs to be assessed, the domains may also be assessed independently. Administration time takes 10–20 min for each domain.

The Cognition domain assesses skills related to memory, planning, decision-making, and discrimination. The Communication domain includes Receptive Language and Expressive Language subdomains. The Social-Emotional Development domain assesses for skills related to social development, emotion regulation, and social interaction. The Physical Development domain includes Gross Motor and Fine Motor subdomains. The Adaptive Behavior domain assesses for daily living skills, self-help skills, and level of independence.

The DAYC-2 can be administered using multiple methods of assessment, including behavioral observation, structured caregiver interview, and/or direct assessment of skills. Observations of the child's behavior may be conducted in the home and/or school environment. The examiner rates each item as either "0" (i.e., the child does not exhibit the behavior; the child exhibits the behavior inconsistently) or "1" (i.e., the child exhibits the behavior most of the time; the child exhibited the behavior when he/she was younger but has outgrown the behavior). Item ratings are calculated to produce the raw scores, which are converted to standard scores, age equivalent scores, and percentile ranks. The composite score from all five domains produce the General Development Index (GDI), which represents the child's overall general development.

The DAYC-2 was found to have good reliability, with alpha coefficient values of 0.90 or greater for all five domains (Swartzmiller, 2014). The correlation coefficient for GDI was excellent, ranging from 0.98 to 0.99. The test sensitivity, specificity, and classification accuracy were found to be above 0.70 for all five domains in children with ASD, the Physical Development domain in children with physical disabilities, and the Communication domain in children with language delays. These findings suggest that for special populations, additional behavioral measures may need to be used in conjunction with the DAYC-2 in order to conduct a comprehensive assessment of developmental disabilities.

Developmental Assessment for the Severely Handicapped, Third Edition (DASH-3)

The *Developmental Assessment for the Severely Handicapped, Third Edition (DASH-3*; Dykes & Mruzek, 2012) is a criterion-referenced test that assesses a range of skills in individuals (i.e., 6 months to adulthood). The DASH-3 was designed for use with individuals with intellectual, developmental (e.g., ASD), physical, and sensory disabilities, across various impairment levels (e.g., mild, moderate, severe). The DASH-3 can be used to determine if an examinee is demonstrating the skills relevant to his or her developmental stage.

The DASH-3 comprises five scales: Sensory-Motor, Language, Social-Emotional, Activities of Daily Living, and Academics. The Sensory-Motor scale measures gross motor, sensory, reflexes, and hand skills. The Language scale assesses the individual's ability to understand and use nonsymbolic communication, expressive language, and receptive language. The Social-Emotional scale assesses social skills and measures the awareness and understanding the individual has have the self and others. The Activities of Daily Living Scales measures skills rated to independence and self-sufficiency, such as feeding, dressing, toileting, home routines, and travel and safety. The Academics scale measures the individual's ability to learn and use information related to pre-academic and academic skills (e.g., number skills, reading skills, concept formation).

The measure can be administered using multiple methods of assessment. The DASH-3 may be completed by an examiner based on a 2 to 3-h direct observation of the examinee. A time-efficient option is to administer the test by interviewing a primary caregiver, teacher, or other adult who is able to report on the examinee's abilities. Individuals may also complete the DASH-3 independently, using self-report. Items are rated on a 7-point Likert scale, with a rating of "1" indicating that the individual cannot complete the task and a score of "7" indicating that the individual can complete the task without assistance. The scale scores are summed together to estimate the examinee's overall developmental age. Examiners may also use the Intervention Planning Worksheet, to identify the developmental skill areas that require treatment, guide intervention planning, and track treatment progress over time.

A major strength of the DASH-3 is that the test was specifically designed to assess development in children with significant intellectual and developmental delays. More specifically, this measure was designed to measure discrete, fine-grained skills among individuals with lower developmental age and severely impaired functioning. This test design allows examiners to detect small behavior changes that other developmental assessment scales are not sensitive enough to capture.

Tests for Adaptive Behavior

Developmental measures are often used in conjunction with adaptive behavior measures in order to capture a comprehensive picture of the child's overall level of functioning. There are two broad types of adaptive behavior scales in the existing research: normative measures and criterion-based measures. Normative measures of adaptive functioning identify the broad skill areas in which the individual demonstrates behavioral deficits. Criterion-based measures provide a more fine-grained assessment of adaptive functioning, as they identify the specific behavioral deficits within a particular skill domain.

Even though a measure of adaptive behavior is a crucial component of the ID diagnosis, there is a dearth of research evaluating whether standardized adaptive behavior scales accurately identify individuals with ID vs. individuals without ID (Balboni et al., 2014). Out of the hundreds of adaptive behavior scales available (Schalock, 1999), only four have been standardized using a representative sample of the general population. These four behavior scales (i.e., Vineland Adaptive Behavior Scales, Third Edition, Adaptive Behavior Assessment System, Third Edition, Scales of Independent Behavior-Revised, and Adaptive Behavior Scale- School Version) are reviewed below. Despite this limitation, all of the adaptive behavior scales reviewed in this chapter were found to be psychometrically sound and were developed in accordance with the definition of ID as defined by the AAIDD, DSM-5, and federal mandates for special education and disability classification (i.e., IDEIA).

Vineland Adaptive Behavior Scales, Third Edition (Vineland-3)

The *Vineland Adaptive Behavior Scales, Third Edition (Vineland-3;* Sparrow, Cicchetti, & Saulnier, 2016) is the most commonly used measure to aid in the diagnosis of intellectual and developmental disabilities. The Vineland-3 measures adaptive functioning across the lifespan, from birth to 90 years of age. The measure comprises three, broad adaptive behavior domains: Communication, Daily Living Skills, and Socialization. The measure also includes optional Motor Skills and Maladaptive Behavior domains. The Communication domain consists of Receptive, Expressive, and Written subdomains. The Daily Living Skills domain includes Personal, Domestic, and Community subdomains. The Socialization domain includes Interpersonal Relationships, Play and Leisure, and Coping Skills subdomains. The Motor domain consists of Fine and Gross Motor subdomains. The Maladaptive Behavior domain includes Internalizing and Externalizing Behavior subdomains.

The test administration time ranges from 10 min to 60 min, depending on the type of form used. There are two forms: Comprehensive Form and Domain-Level Form. The Comprehensive Form is the full-length version that provides a more in-depth evaluation of adaptive functioning. This form produces the Adaptive Behavior Composite (ABC) score, domain scores, subdomain scores, and item scores. The Domain-Level Form is the abbreviated version of the Vineland-3 that can be used to reduce administration time. This version produces the ABC score, domain scores, and item scores. Additionally, there are multiple rater forms for both the Comprehensive and Domain-Level Forms: Interview Form, Parent/Caregiver Form, and Teacher Form.

The Interview Form is completed by the examiner using a semi-structured interview method of assessment, and the examiner rates the measure items based on the caregiver's responses. In the Parent/Caregiver Form, the primary caregiver rates each item independently. The Teacher Form is used for school-aged children between the ages of 3 and 21 years. As with the Parent/Caregiver Form, the child's teacher rates the measure items. Items are rated on a 3-point Likert scale, depending on the individual's ability to complete a task. A rating of "0" indicates that the individual is never able to perform the task, a rating of "1" indicates that the individual is sometimes able to perform the task, and a rating of "2" indicates that the individual is usually able to perform the task. Some items are rated as "yes" or "no."

Scores from the Vineland-3 meet the AAIDD and DSM-5 requirements for the diagnosis of ID. The previous version of the Vineland-3, the Vineland-II (Sparrow et al., 2005), has been found to accurately classify 71–100% children (i.e., 6–18 years old) and 97–100% of adults (19–86 years old) into the different severity levels of ID (i.e., mid, moderate, severe, and profound). Results from the Vineland-3 may help determine eligibility for special services and aid in intervention program planning. Further, the Vineland-3 can be used to assess adaptive behavior in individuals with ASD, attention deficit hyperactivity disorder, post-traumatic brain injury, hearing impairment, and dementia or Alzheimer's disease.

Adaptive Behavior Assessment System, Third Edition (ABAS-3)

The *Adaptive Behavior Assessment System, Third Edition* (*ABAS-3;* Harrison & Oakland, 2003) is a behavior rating scale used to assess for adaptive behavior in individuals from birth to 89 years of age and can aid in the diagnosis of various developmental disabilities, learning disorders, and behaviors disorders. The ABAS-3 assesses three adaptive domains: Conceptual, Social, and Practical. The domains include 11 subdomains: communication, community use, functional academics, health and safety, home or school living, leisure, motor, self-care, self-direction, social, and work. There are several rating forms; (1) Infant and Preschool: Parent/Primary Caregiver Form (rated by the primary caregiver, for children 0–5 years); (2) Infant and Preschool: Teacher/Daycare Provider Form (rated by the teacher, for 2–5 years); (3) School: Parent Form (rated by the primary caregiver, for children 5–21 years); (4) School: Teacher Form (rated by the teacher, for children 5–21 years); (5) Adult Form (rated by self or primary caretaker, for individuals 16–89 years).

The ABAS-3 takes approximately 15–20 min to complete. Raters provide a frequency rating and an ability rating based on a 4-point Likert scale. Raw scores are converted to scaled scores. Each subdomain scaled score is then summed to produce the composite score and percentile ranks for each of the three adaptive domains. Finally, the domain composite scores are added together to produce the General Adaptive Composite (GAC) score. There is also an option to compare ratings across different respondents (e.g., between parent and teacher).

The standardization sample for the ABAS-3 included typically developing individuals as well as individuals with mild disabilities, across all ages. The reliability coefficients for the three adaptive domains were above 0.85 and the reliability coefficients for GAC were excellent, ranging from 0.96 to 0.99. In the sample of individuals with disabilities, the reliability coefficient for GAC was 0.99 and the reliability coefficients for the adaptive domains ranged from 0.96 to 0.99.

Scales of Independent Behavior-Revised (SIB-R)

The *Scales of Independent Behavior-Revised (SIB-R;* Bruininks, Woodcock, Weatherman, & Hill, 1996) was designed to measure adaptive functioning and functional independence for individuals 3 months to 80+ years. The SIB-R evaluates four adaptive skill areas: Motor Skills, Social Interaction and Communication, Personal Living Skills, and Community Living Skills. The adaptive domains are further divided into the following 14 subdomains: Gross Motor, Fine Motor, Social Interaction, Language Comprehension, Language Expression, Eating and Meal Preparation, Toileting, Dressing, Personal Self-Care, Domestic Skills, Time and Punctuality, Money and Value, Work Skills, and Home/Community Orientation. An optional Problem Behavior scale may also be administered. The Problem Behavior scale includes 3 maladaptive domains (i.e., Internalized, Externalized, Asocial), which include the following 8 subdomains: Hurtful to Self, Hurtful to Others, Unusual or Repetitive Habits, Withdrawal or Inattentive Behavior, Socially Offensive Behavior, Uncooperative Behavior, Destructive to Property, and Disruptive Behavior.

The SIB-R is administered to the primary caregiver using a semi-structured interview format or as a respondent checklist. There are three different forms available. The Full Scale Form is a comprehensive evaluation that assess all 14 adaptive behavior subdomains. It consists of 259 items and takes approximately 60 min to complete. The Short Form is used as a screening tool and comprises 40 items from the Full Scale SIB-R. The Early Development Form was designed to be used with young children or individuals with a developmental age under 8 years. The Short Form and Early Development Form take approximately 15–20 min to complete.

The Adaptive Behavior items are rated on a 4-point Likert scale, based on the degree to which the individual completes a task without help or supervision. A rating of “0” indicates that the individual never or rarely is able to do the task, even if asked; “1” indicates that the individual does not perform the task well and may need to be asked to do the task 25% of the time; "2" indicates that the individual does the task fairly well and does the task 75% of the time when asked; "3" indicates that the individual does the task very well always or almost always, without being asked. The Problem Behavior scale items are rated on frequency and severity of the behavior. Frequency of the behavior is rated as: "1"—occurs less than once a month; "2"—occurs 1–3 times per month; "3"—occurs 1–6 times per week; "4"—occurs 1–10 times per day; "5"—occurs 1 or more times per hour. Severity of the behavior is rated as: "0"—not serious, not a problem; "1"—slightly serious, mild problem; "2"—moderately serious, moderate problem; "3"—very serious, severe problem; "4"—extremely serious, critical problem. The ratings are calculated to produce a Broad Independence Score, Relative Mastery Indexes (RMI), Support Score, age equivalents, and percentile ranks. The Support Score estimates the level of support score an individual will require (i.e., pervasive to infrequent support, no support) based on individual's current adaptive functioning and frequency and severity of problem behaviors. Higher scores indicate that an individual is functioning with more independence.

The SIB-R has demonstrated adequate test sensitivity and specificity, as 76% of the standardization sample were correctly classified into their respective ID severity groups (i.e., mild, moderate, non-ID). Specifically, 51% of individuals with mild ID were accurately identified, 74% of individuals with moderate ID were accurately identified, and 82% of individuals without ID were accurately identified.

AAMD Adaptive Behavior Scales (AAMD ABS)

The *AAMD Adaptive Behavior Scales (AAMD ABS)* was first published in 1969, following the inclusion of adaptive behavior in the definition of the former "mental retardation" diagnosis (Hopp & Baron, 2011). Since then, the AAMD ABS has undergone several revisions. The AAMD ABS has two versions, the Adaptive Behavior Scales-Residential and Community, second edition (ABS-RC:2; Nihira, Leland, & Lambert, 1993) and the Adaptive Behavior Scale- School, second edition (Lambert, Nihira, and Leland, 1993).

The ABS-RC:2 is used to assess adaptive behavior in residential and community settings, in individuals from birth to 79 years of age. Part One of the ABS-RC:2 comprises 10 domains: Independent Functioning, Physical Development, Economic Activity, Language Development, Numbers and Time, Domestic Activity, Prevocational/Vocational Activity, Self-Direction, Responsibility, and Socialization. Part Two assesses for Social Adjustment (i.e., Social Behavior, Conformity, and Trustworthiness) and Personal Adjustment (i.e., Stereotyped and Hyperactive Behavior, Sexual Behavior, and Self-Abusive Behavior). Each domain includes subdomains that assess for specific skill areas. For the domains included in Part One of the ABS-RC:2, raw scores can be converted to standard scores, percentile ranks, and age equivalents.

The ABS-S:2 is used to assess adaptive behavior in school settings, in children between the ages of 3 and 21 years. Part One comprises nine domains (i.e., Independent Functioning, Physical Development, Economic Activity, Language Development, Numbers and Time, Prevocational/Vocational Activity, Self-Direction, Responsibility, and Socialization) and 18 subdomains. Part Two comprises seven domains: Social Behavior, Conformity, Trustworthiness, Stereotyped and Hyperactive Behavior, Self-Abusive Behavior, Social Engagement, and Disturbing Interpersonal Behavior.

The ABS-RC:2 and ABS-S:2 can be completed by the examiner or by the primary caregiver. Some items are rated on a 3-point Likert scale, with "0" indicating the least amount difficulty with the item and "3" indicating the most amount difficulty with the item. Other items are rated as frequency of behavior, with "N" indicating that the individual never engages in the behavior, "O" indicating that the individual occasionally engages in the behavior, or "F" indicating that the individual frequently engages in the behavior. Some items are rated as "yes" or "no."

The standardization sample for the ABS-RC:2 included individuals with unspecified developmental disabilities in the United States. Individuals included in the standardization sample also had disabilities such as blindness, deafness, learning disability, and speech/language impairments. The reliability coefficient of the ABS-RC:2 was found to range from 0.80 to 0.99. The ABS-S:2 was standardized on individuals with and without ID in the United States. The reliability coefficient of the ABS-C:2 was found to be around or above 0.90. Criterion and content validity were found to be adequate for both versions.

Diagnostic Adaptive Behavior Scale (DABS)

The *Diagnostic Adaptive Behavior Scale (DABS;* Tasse et al., 2012) is a comprehensive standardized assessment of adaptive behavior designed for individuals between the ages of 4 and 21 years. The DABS measures adaptive behavior in the Conceptual, Social, and Practical domains. The Conceptual domain includes skills such as literacy, self-direction, and number concepts, time concepts. The Social domain includes interpersonal skills, social responsibility, self-esteem, following rules, gullibility, and following social rules. The practical skill includes skills related to daily living (i.e., self-care), safety, healthcare, use of money, and occupation skills.

The DABS is administered using an interview format, in which the examiner asks the primary caregiver to rate the individual's ability to perform the task item. Each domain consists of 25 items, and each item is rated on a 4-point Likert scale. A score of "0" indicates that the individual is rarely or never able to do the task and a score of "3" indicates that the individual always or almost always independently does the task. The three domain scores are calculated to produce an Overall Adaptive Behavior Standard Score (OABSS). Scaled scores that are 2 standard deviations below the mean indicate that individuals demonstrate "significant limitations" of adaptive functioning and indicate that a diagnosis of ID or other related developmental disability may be warranted.

The standardization sample for the DABS included individuals with formal diagnoses of ID or developmental delay, individuals without deficits in adaptive behavior, and individuals with disorders not related to ID (e.g., learning disability, ADHD). The DABS was found to accurately identify 81%–98% of individuals with ID and 89%–91% of individuals without ID. Further, the DABS demonstrated good to excellent accuracy in differentiating among the different severity levels of ID (i.e., mild, moderate, severe and profound). A limitation of the DABS is that it does not provide a detailed assessment of specific skill areas. As such, the test authors recommend that the DABS should not be used for treatment planning or to monitor specific intervention areas (Balboni et al., 2014).

Adaptive Behavior Evaluation Scale, Third Edition (ABES-3)

The *Adaptive Behavior Evaluation Scale-Revised (ABES-R;* McCarney & House, 1995) is an adaptive behavior rating scale that was designed to screen for adaptive functioning in educational and residential settings. The ABES-R is used to screen for deficits in specific skill areas, identify areas that require support, and aid in the diagnosis of ID, learning disabilities, behavior disabilities, and other developmental delays. In accordance with the AAIDD definition of adaptive behavior, the ABES-R assesses adaptive skills across the Conceptual, Social, and Practical domains. These three domains include 10 subdomains: Communication, Self-Care, Home Living, Social, Community Use, Self-Direction, Health and Safety, Functional Academics, Leisure, and Work.

There are two versions of the ABES-R: the home version (HV), which is completed by the parent or primary caregiver and school version (SV), which is completed by the teacher. Raters rate each item based on direct observation of the individual's behaviors. The ABES-R is further divided by age group. The ABES-3:4–12 HV and ABES-3:4–12 SV are used to assess children between 4 and 12 years old. The ABES-3:13–18 HV and ABES-3:13–18 SV are used to assess adolescents between 13 and 18 years old. All versions of the ABES-R comprise 104 items and take 15–20 min to complete. Raters rate each item based on a 6-point Likert scale: "0" indicates that the skill is not developmentally appropriate for the individual's age, "1" indicates that the individual does not demonstrate the skill, "2" indicates that the individual is developing the skill, "3" indicates that the individual demonstrates the skill inconsistently, "4" indicates that the individual demonstrates the skill most of the time, and "5" indicates that the individual consistently demonstrates the skill at all times. If the rater has not had an opportunity to observe a particular skill, they may consult with another caretaker or educator who is able to rate the item. The ratings are calculated to produce an Adaptive Skills Quotient (ASQ), which represents a general measure of the individual's adaptive functioning.

Adaptive Behavior Inventory (ABI)

The *Adaptive Behavior Inventory (ABI;* Brown & Leigh, 1986*)* was designed to assess daily living skills in school-aged children between 6 and 18 years old. This measure can be used as a diagnostic tool to identify children with intellectual, developmental, or other emotional disabilities. The ABI is composed of 5 subscales: Self-Care Skills, Communication Skills, Social Skills, Academic Skills, and Occupational Skills. The Occupational Skills domain may be omitted for very young children or children with severe skill impairments.

The ABI was designed to be completed by the child's classroom teacher or other professional staff who has relevant knowledge of the child adaptive abilities. The ABI takes about 30 min to complete and comprises 150 items, with approximately 30 items per subscale. A Short Form is also available for screening or research purposes. The ABI-Short Form comprises 50 items and takes 5–10 min to complete. Test items are rated on a 4-point Likert scale: "0" indicates that the child does not perform the skill, "1" indicates that the skill is emerging, "2" indicates that child performs the skill most of the time, and "3" indicates that the child has mastered the skill. Subscale items are calculated to produce an Adaptive Behavior Quotient and percentile ranks.

The ABI was standardized using a sample of students with and without intellectual disabilities. However, the test has not been revised since its publication in 1986, and the ABI may need to be re-normed. Nevertheless, test reliability of the ABI has been demonstrated, with subtest-by-age reliability coefficients ranging from 0.86 to 97. The reliability coefficient of the Adaptive Behavior Quotient was found to exceed 0.90 for all age groups.

Checklist of Adaptive Living Skills (CALS)

The Checklist of Adaptive Living Skills (CALS; Bruininks & Morreau, 2004) is a criterion-referenced behavior scale that was designed to assess adaptive living skills in individuals with or without ID, from infancy to adulthood. The CALS is compose of 800 specific behavior items, which are categorized into four broad adaptive domains: Personal Living Skills, Home Living Skills, Community Living Skills, and Employment Skills. These domains are further divided into 24 subdomains.

Total administration time of the CALS is 60 min; however, if there are concerns regarding specific areas of adaptive functioning, the CALS can be administered at the domain level. Caregivers rate each item based on the individual's ability to complete the task independently. Additionally, items are rated according to the type of prompting the individual requires to complete the task (i.e., gestural, verbal, physical). Based on the informant's response, a prompting score of "independent performance,"

"performance with prompt," or "nonperformance" are produced.

The CALS was designed to be used in conjunction with the Adaptive Living Skills Curriculum (ALSC; Bruininks, Morreau, Gilman, & Anderson, 2004), to integrate assessment and treatment of adaptive skills. Each item on the CALS is categorized by domain, subdomain, and item number, which corresponds with a specific instructional item on the ALSC. This integrated approach to assessment helps guide the intervention planning process by identifying the specific treatment targets that are associated with the individual's skill deficits. Further, the CALS and ALCS can be used to identify treatment goals and determine mastery of targeted behaviors.

Children's Adaptive Behavior Scale, Revised (CABS-R)

The *Children's Adaptive Behavior Scale, Revised* (*CABS-R*; Kicklighter & Richmond, 1983) is an adaptive behavior measure that was specifically designed to assess adaptive functioning of children (5–11 years old) in the school setting. The CABS-R evaluates five areas of adaptive functioning: Language Development, Independent Functioning, Family Role Performance, Economic-Vocational Activity, and Socialization. Each domain is composed of 16 to 30 items.

The CABS-R differs from the other measures of adaptive behavior described in this chapter because it is administered directly to the child, in the educational setting. The total test administration time is approximately 45 min, as the child is required to respond to all test items. This unique method of assessment inevitably limits the range of children that can be evaluated using the CABS-R. The CABS-R is likely not appropriate for administration with children with limited nonverbal and/or verbal skills, certain medical conditions, cognitive impairments, and developmental delays. In response to these limitations, the test authors recommend that examiners corroborate the child's responses with another individual who is familiar with the child's level of adaptive functioning. Additionally, the test authors suggest that other measures of adaptive behavior be used to supplement the CABS-R and get a comprehensive picture of the child's overall adaptive functioning. However, these recommendations challenge the uniqueness of the CABS-R and have implications for inter-rater reliability of responding between the child and other adult.

Survey of Functional Adaptive Behaviors

The *Survey of Functional Adaptive Behaviors (SFAB*; Dial, Mezger, Massey, Carter, & McCarron, 1986) is a rating scale that was designed to assess adaptive skills in individuals 16 years old and older. The SFAB is composed of 135 test items, which are categorized into four adaptive skill areas: Residential Living Skills, Daily Living Skills, Academic Skills, Vocational Skills. The Residential Living Skills domain evaluates self-care and home management skills. The Daily Living Skills assesses skills related to money management, travel, and community resources. The Academic Skills domain assesses skills related to language, literacy, writing, and number concepts. The Vocational Skills domain evaluates the individual's physical abilities and skill related to vocational tasks.

The SFAB can be completed using multiple methods of administration, including case history review, interview, and/or direct behavioral observation. Test items are rated as: "0"—task not performed, "1"—task performed inadequately or requires assistance, prompting, or cueing; "2"—task performed independently. The scores are calculated to produce a Total Adaptive Behavior score. However, the results should be interpreted with caution, as there is a lack of research evaluating the interpretability of the test findings. As such, it would be beneficial to use the SFAB as a screening tool rather than as a diagnostic tool.

Conclusions

With increased recognition that early intervention is crucial in remediating developmental delays in young children, there has been an increase in research evaluating the efficacy of developmental behavior scales to accurately assess developmental delays in young children. Broadly, these developmental skill areas include language, communication, motor, social, and adaptive skills. Adaptive behavior scales are often used in conjunction with developmental scales to provide a more fine-grained assessment of a child's adaptive skills within conceptual, social, and practical domains. This chapter discussed several developmental and adaptive behaviors scales that have been designed to screen for developmental delays and aid in the early diagnosis of ID or other developmental disabilities. Further, many assessment measures that were reviewed in this chapter have been found to reliably measure developmental delays across the lifespan, from infancy to adulthood. Additionally, these measures offer multiple methods of assessment, so test administration can be modified according to the availability of time and resources. Administration of developmental and adaptive behavior scales is a critical first step in the early diagnosis of ID and developmental disabilities. These tools should also be administered periodically to guide intervention planning, determine the types and level of support services required, and monitor treatment progress, in order to optimize treatment efficacy and improve learning outcomes.

References

Albers, C. A., & Grieve, A. J. (2007). Test Review: Bayley, N. (2006). Bayley Scales of Infant and Toddler Development– Third Edition. San Antonio, TX: Harcourt Assessment. *Journal of Psychoeducational Assessment, 25*(2), 180–190. https://doi.org/10.1177/0734282906297199

American Association on Intellectual and Developmental Disabilities. (2010). *Intellectual disability: Definition, classification, and systems of support* (11th ed.). Washington, DC: American Association on Intellectual and Developmental Disabilities.

American Psychiatric Association. (2013). *Diagnostic and statistical manual of mental disorders* (5th ed.). Arlington, VA: American Psychiatric Association.

Balboni, G., Tassé, M. J., Schalock, R. L., Borthwick-Duffy, S. A., Spreat, S., Thissen, D., … Navas, P. (2014). The diagnostic adaptive behavior scale: Evaluating its diagnostic sensitivity and specificity. *Research in Developmental Disabilities, 35*(11), 2884–2893. https://doi.org/10.1016/j.ridd.2014.07.032

Bayley, N. (2006). *Bayley Scales of Infant and Toddler Development– Third Edition*. San Antonio, TX: Harcourt Assessment.

Bliss, S. L. (2007). Test Reviews: Newborg, J. (2005). Battelle Developmental Inventory−Second Edition. Itasca, IL: Riverside. *Journal of Psychoeducational Assessment, 25*(4), 409–415. https://doi.org/10.1177/0734282907300382

Brown, L., & Leigh, J. E. (1986). *Adaptive Behavior Inventory (ABI)*. Austin, TX: Pro-Ed.

Bruininks, R. H., & Morreau, L. (2004). *Checklist of adaptive living skills (CALS)*. Chicago, IL: Riverside Publishing Company.

Bruininks, R. H., Morreau, L., Gilman, C. J., & Anderson, J. L. (2004). *Adaptive living skills curriculum (ALSC)*. Chicago, IL: Riverside Publishing Company.

Bruininks, R. H., Woodcock, R., Weatherman, R., & Hill, B. K. (1996). *Scales of independent behaviour-revised*. Park Allen, TX: DLM Teaching Resources.

Dial, J. G., Mezger, C., Massey, T., Carter, S., & McCarron, L. T. (1986). *Survey of functional adaptive behaviors*. Dallas, TX: McCarron-Dial Systems, Inc.

Dumont, R., Cruse, C. L., Alfonso, V., & Levine, C. (2000). Book review: Mullen scales of early learning: AGS edition. *Journal of Psychoeducational Assessment, 18*, 381–389.

Dykes, M. K., & Mruzek, D. W. (2012). *Developmental assessment for the severely handicapped, third edition (DASH-3)*. Austin, TX: PRO-ED.

Guralnick, M. J. (2005). Early intervention for children with intellectual disabilities: Current knowledge and future prospects∗. *Journal of Applied Research in Intellectual Disabilities, 18*(4), 313–324. https://doi.org/10.1111/j.1468-3148.2005.00270.x

Harrison, P. L., & Oakland, T. (2003). *Adaptive behavior assessment system–second edition*. San Antonio, TX: The Psychological Corporation.

Hopp, C. A., & Baron, I. S. (2011). In J. S. Kreutzer, J. DeLuca, & B. Caplan (Eds.), *Encyclopedia of clinical neuropsychology*. New York, NY: Springer.

Individuals with Disabilities Education Act of 2004, 20 U.S.C. § 1400 (2004).

Kicklighter, R. H., & Richmond, B. O. (1983). *Children's adaptive behavior scale, revised (CABS-R)*. Lake Worth, FL: Green Dragon Publishing.

King, B. H., Toth, K. E., Hodapp, R. M., & Dykens, E. M. (2009). Intellectual disability. In B. J. Sadock, V. A. Sadock, & P. Ruiz (Eds.), *Comprehensive textbook of psychiatry* (9th ed., pp. 3444–3474). Philadelphia, PA: Lippincott Williams & Wilkins.

Lambert, N., Nihira, K., & Leland, H. (1993). *Adaptive Behavior Scale-School, Second Edition*. Austin, TX: Pro-Ed, Inc.

Lovaas, O. I. (1987). Behavioral treatment and normal educational and intellectual functioning in young autistic children. *Journal of Consulting and Clinical Psychology, 55*, 3–9.

Maulik, P. K., Mascarenhas, M. N., Mathers, C. D., Dua, T., & Saxena, S. (2011). Prevalence of intellectual disability: A meta-analysis of population-based studies. *Research in Developmental Disabilities, 32*(2), 419–436. https://doi.org/10.1016/j.ridd.2010.12.018

McCarney, S. B., & House, S. N. (1995). *Adaptive behavior evaluation scale- revised (ABES-3)*. Columbia, MO: Hawthorne Educational Services, Inc.

Mullen, E. M. (1995). *Mullen scales of early learning: AGS edition*. Circle Pines, MN: American Guidance Service.

Newborg, J. (2005). *Battelle Developmental Inventory* (2nd ed.). Itasca, IL: Riverside Publishing.

Nihira, K., Leland, H., & Lambert, N. (1993). *Adaptive Behavior Scales-Residential and Community, 2nd edition* (ABS-RC:2). Austin, TX: ProEd.

Schalock, R. L. (1999). *Adaptive behavior and its measurement: Implications for the field of mental retardation*. Washington, DC: American Association on Mental Retardation.

Schalock, R. L., Borthwick-Duffy, S. A., Bradley, V. J., Buntinx, W. H., Coulter, D. L., Craig, E. M., … Yeager, M. H. (2010). *Intellectual disability: Definition, classification, and systems of supports* (11th ed.). Washington, DC: American Association on Intellectual and Developmental Disabilities.

Sharav, T., & Shlomo, L. (1986). Stimulation of infants with Down syndrome: Long-term effects. *Mental Retardation, 24*, 81–86.

Sipes, M., Matson, J. L., & Turygin, N. (2011). The use of the Battelle Developmental Inventory–Second Edition (BDI-2) as an early screener for autism spectrum disorders. *Developmental Neurorehabilitation, 14*(5), 310–314. https://doi.org/10.3109/17518423.2011.598477

Smith, T., Groen, A. D., & Wynn, J. W. (2000). Randomized trial of intensive early intervention For children with pervasive developmental disorder. *American Journal on Mental Retardation, 105*, 269–285.

Sparrow, S., Cicchetti, D., & Balla, D. (2005). *Vineland Adaptive Behavior Scales (2nd ed.)*. Circle Pines, MN: American Guidance Service.

Sparrow, S. S., Cicchetti, D. V., & Saulnier, C. A. (2016). *Vineland adaptive behavior scales, third edition (Vineland-3)*. Bloomington, MN: Pearson.

Squires, J., Bricker, D., Twombly, E., Nickel, R., Clifford, J., Murphy, K., … Farrell, J. (2009). *Ages & stages questionnaires, 3rd edition (ASQ-3)*. Baltimore, MD: Paul H. Brookes.

Swartzmiller, M. D. (2014). Test review: Developmental assessment of young children-second edition (DAYC-2). *Journal of Psychoeducational Assessment, 32*(6), 577–580.

Tasse, M. J., Schalock, R. L., Balboni, G., Bersani, H., Borthwick-Duffy, S. A., Spreat, S., … Zhang, D. (2012). The construct of adaptive behavior: Its conceptualization, measurement, and use in the field of intellectual disability. *American Journal on Intellectual and Developmental Disabilities, 117*(4), 293–303.

Voress, J. V., & Maddox, T. (2013). *Developmental assessment of young children–second edition (DAYC-2)*. Austin, TX: PRO-ED.

Tests for Dual Diagnosis

23

Carmen Vargas-Vargas, Anna Costa-Vargas, and Dunia Montalvo-Pérez

Introduction

Before 1970, researchers believed that people with intellectual disabilities (ID) were unable to experience emotional and psychopathologic disorders due to insufficient strength of the ego (Deb, Thomas, & Bright, 2001). However, this point of view has been disappearing for the last decades, since researchers have demonstrated that not only can they experience associated psychopathologic disorders, but also have a bigger risk of having them in comparison with the general population (Borthwick-Duffy, 1994). Due to this high prevalence of detected psychopathologic disorders, it has become a much-studied area. A great number of researchers have provided proof that people with ID are more likely to have associated psychopathologic disorders, when compared with the general population (Cherry, Penn, Matson, & Bamburg, 2000; Matson & Bamburg, 1998).

The development of psychometric tests to evaluate the mental health of these people has been a well-established area of investigation. Its growing expansion has allowed great advances in the last four decades, with the emergence of a large number of evaluation instruments for both adults and children, some of them widely studied both in North America and in Europe (Matson, Belva, Hattier, & Matson, 2012).

The identification of mental disorders in these people is frequently complicated, seeing as they often have difficulty communicating the symptoms (Konstantareas & Hewitt, 2001). Some of these complications include expression and comprehension problems, as well as a smaller level of introspection that may influence their description or recognition.

The lack of adequate instruments makes it necessary to use complementary materials (alternative communication systems) and to be flexible (regarding the place of evaluation, its duration, the language used, etc.).

In addition to the previously stated, there is a frequent occurrence of what is commonly known as "diagnostic eclipse" (Reiss, Levitan, & Szyszko, 1982). This is a phenomenon in which the mental health disorder is not diagnosed or is not accurately so, since the tendency is to attribute all behavioral or cognitive change to the condition of the person with ID and not to a possible associated psychopathology.

Fortunately, today, the recognition of the rights of people with ID to adequate medical attention according to the normalization principles, which

---

C. Vargas-Vargas (✉) · D. Montalvo-Pérez
Hermanas Hospitalarias-Benito Menni CASM,
Service for People with Intellectual Disability,
Barcelona, Spain
e-mail: cvargas.hbmenni@hospitalarias.es

A. Costa-Vargas
Department of Psychology, Universitat Autònoma de
Barcelona, Barcelona, Spain

J. L. Matson (ed.), *Handbook of Intellectual Disabilities*, Autism and Child Psychopathology
Series, https://doi.org/10.1007/978-3-030-20843-1_23

ensure that people with disabilities have a right to live in a community and use their resources, and force an improvement of the skills to detect and diagnose mental disorders.

It is necessary to have information from carers, people with direct contact or relatives as the closest key informants. These are important pieces to the diagnosis. A precise diagnosis allows an adequate treatment (Deb et al., 2001).

The interest for instruments designed to assess psychopathology in people with ID is not only growing but has also had repercussions in the way treatment is administered. Despite the creation of several scales to evaluate the presence of co-occurring psychopathologic disorders in these individuals, some of these instruments were barely studied (Matson et al., 2012).

It was the investigations in the 1980s, specifically, that allowed the community to recognize the need to develop mental health scales specific to this population. The first evaluation instrument developed, the Psychopathology Instrument for Mentally Retarded Adults (PIMRA) which evaluates people with severe and profound ID (Kazdin, Matson, & Senatore, 1983), appeared as a psychopathologic tool specific for adults with ID. This scale is still generally used and has been translated to several languages. This was followed by other scales, including the Reiss Screen, as well as by the diagnosis evaluation measures for people with severe disabilities.

The clinical work and the investigation regarding ID and dual diagnosis were mainly based on investigation derived from general population. Several scales of clinical assessment were developed from the general population and were adapted for use in people with ID.

Recognizing the diagnosing problems that the health professionals face when trying to achieve a diagnostic evaluation in this area, the National Association for the Dually Diagnosed (NADD), in association with the American Psychiatric Association (APA), created the *Diagnostic Manual—Intellectual Disability (DM-ID): A Textbook of Diagnosis of Mental Disorders in Persons with Intellectual Disability* (Fletcher, Barnhill, Mccarthy, & Strydom, 2016).

The DM-ID was designed as a complement to the Diagnostic and Statistical Manual of Mental Disorders, Fourth Edition, Text Revision (DSM-IV-TR). This manual serves as a guide for the evaluation and diagnosis of specific disorders in people with ID and offers information on how to recognize the problematic behaviors, as well as on how to differentiate them from psychopathology. Regarding this, it has been observed that mental health symptoms are, originally, not related to environmental factors and that, conversely, challenging behaviors tend to be independent from the psychopathology and are in large measure determined by environmental factors (Matson & Shoemaker, 2011).

People with ID have cognitive and communicational difficulties, and therefore may not be capable of giving direct information regarding their psychopathologic problems. This makes evaluating them difficult and forces the professionals to deeply investigate the diagnosis bases and to be up-to-date on the advances and developments of the instruments in use. Therefore an adequate evaluation and intervention is required, given that mental health problems cause social exclusion. Current laws protect people with ID and require that they are adequately studied, including their mental health, so that the interventions are appropriate. In this context, the psychopathologic tools are useful and necessary to the clinical evaluation, but if they are used without proper validity and psychometric reliability then they have failed in the identification of the psychopathologic disorders and, consequently, have failed to provide an effective treatment.

Although there are several tools available for a general psychopathologic evaluation, their validity for patients with ID is questionable, since many of those tools were standardized for people without ID and make no reference to the way the examiner must evaluate this population (Kishore, Nizamie, & Nizamie, 2010). Currently, there are multiple psychometric scales that evaluate psychopathology in these people; however, the number of adapted and validated tools is scarcer (Matson et al., 2012).

Description of the Instruments

Tools for the psychopathologic evaluation of people with ID have existed for decades, and a large number of standardized tests have been developed so far. Most of these can be completed in 15–30 min, and are designed to allow the detection of multiple psychopathologic disorders (Matson & Shoemaker, 2011). The tools detailed in this chapter are the most commonly used and with a larger scientific follow-up according to the available literature.

We also included others that need further investigation, as well as some that are still expanding. A mention is made regarding some that have marked history, allowing for the evolution and development of other psychometric instruments that still exist.

We have grouped the different instruments into two main types: (1) ***General psychopathologic tools*** (Table 23.1) and (2) ***Specific psychopathologic tools*** (Table 23.2). Within the specific ones there are three different groups: (A) *Anxiety disorders and depression*, (B) *Dementia* and (C) *Autism spectrum disorders*.

The first deal with how to detect possible psychopathologic diagnosis associated with ID, cover a large range of mental disorders and are based essentially on criteria from the current diagnosis manuals (DSM-III-R, DSM-IV-TR), where the factor analysis is the main method of establishing diagnostics categories. Thanks to these criteria there are a good number of well-established evidence. The second group assesses specific mental disorders. The ones described here are the most widely studied, deriving from more current evidence-based investigation that assesses the diagnostic of a concrete psychopathologic disorder in people with ID. In many cases, these presuppose a follow-up beyond the more general evaluation instruments.

General Psychopathologic Tools

We detail the first scales of clinical evaluation used as general psychopathology measures for the dual diagnosis of people with ID. These were the first tools to be developed and then used as a foundation for the latter unfolding of specific scales.

Table 23.1 General psychopathological scales

Abbreviation	Instrument name	Year	Authors
ABCL	Adult behavior checklist	2003	Achenbach et al.
ADD	Assessment of dual diagnosis	1988	Matson et al.
BSI	The brief symptom inventory	1983	Derogais
CBCL	Child behavior checklist	1983	Achenbach et al.
DASH-II	Diagnostic assessment for the severely handicapped	1991	Matson et al.
DBC	Developmental behavior Checklist	1995	Einfeld et al.
N-CBRF	Nisonger child behavior rating form	1996	Aman et al.
PAID	Psychopathology checklists for adults with intellectual disability	2007	Hove et al.
PAS-ADD	Psychiatric assessment Schedules for adults with developmental disabilities	1993	Moss et al.
PIMRA	Psychopathology instrument for mentally retarded adults	1984	Matson
RSMB	Reiss screen for maladaptive behavior	1987	Reiss

Adult Behavior Checklist (ABCL)

Author(s) and year: Achenbach et al., 2003.

Provenance/adaptations: USA. The ABCL follows the Aberrant Behavior Checklist (ABC) (Aman, Singh, Stewart, & Field, 1985), which in turn follows the Adaptive Behavior Scale (ABS) (Nihira, Foster, Hellhaas, & Leland, 1975). The ABS was the first adaptive behavior scale, important for the development of other scales that assess nonadaptive behavior.

Usage: The ABC was not specifically designed for people with ID. It was later used in people with moderate to profound ID and, after that, in a sample of people with an limit- or low-intelligence quotient without apparent loss of the psychomet-

Table 23.2 Specific psychopathological scales

Abbreviation	Instrument name	Year	Authors
Anxiety disorders and depression			
ADAMS	Anxiety, depression, and mood scale	2003	Esbensen et al.
BDI-II	Beck depression inventory	1978	Beck
GAS-ID	Glasgow anxiety scale for people with an intellectual disability	2003	Midham et al.
GDS-LD	Glasgow depression scale for people with a learning disability	2003	Cuthill et al.
SAS	Zung self-rating anxiety scale	1971	Zung
SAED	Scheme for appraisal of emotional development	1990	Dosen
MIPQ	The mood interest & pleasure questionnaire	2003	Ross et al.
Dementia			
ABDQ	The adaptive behavior dementia questionnaire	2004	Prasher et al.
CAMDEX-DS	The Cambridge examination for mental disorders of older people with Down's syndrome and others with intellectual disabilities	2004	Ball et al.
DMR	Dementia questionnaire for persons with mental retardation	1992	Evenhuis
DRS	Dementia rating scale	1976	Mattis et al.
DSQIID	Dementia screening Questionnaire for individuals with intellectual disabilities	2007	Deb et al.
G-DSDS	Gedye dementia scale for Down syndrome	1998	Gedye
NTG-EDSD	National task group—Early detection screen for dementia	2013	Esralew et al.
Autism spectrum disorders			
ADI-R	The autism diagnostic interview—Revised	1994	Lord et al.
ACL	Autism checklist	2014	Sappok et al.
ADOS	Autism diagnostic observation schedule	2000	Lord et al.
ASD-DA	Autism spectrum disorders—Diagnosis for intellectually disabled adults	2008	Matson et al.
CARS	Childhood autism rating scale	1980	Schopler et al.
DiBAS-R	Diagnostic behavioral assessment for autism spectrum disorder—Revised	2014	Sappok et al.
PDD-MRS	Pervasive developmental disorder in mentally retarded persons	2005	Kraijer et al.
SCQ	Social communication questionnaire	2003	Rutter Bailey et al.

ric integrity of the scale (Rojahn & Helsel, 1991). There is also a new version (ABC-Community), destined for people inserted in the community. This was considered adequate for the age group of 18–59 years old.

Evaluators: Proxy informant.

Duration of test: Indeterminate.

Objective: To assess the psychopathology in adult population with ID and its severity.

Dimension/score: The ABCL (Achenbach & Rescorla, 2003) contains 118 behavioral issues and is scored in a three-level classification scale: *1—Not certain*; *2—Somewhat or sometimes true*; and *3—Very certain or true*. With the analytical factor methods, it is possible to distinguish eight syndromes according to the subscales: *1—Anxious-depressive*; *2—Withdrawnness*; *3—Somatic complains*; *4—Thought problems*; *5—Attention problems*; *6—Aggressive behavior*; *7—Disruptive behavior*; and *8—Intrusive behavior*. Besides these, two wide-reaching syndrome systems, labeled "Internalization" and "Externalization," were identified following the analysis of second order factors.

Antecedents/description: This is not the first instrument developed for general population used in people with ID, since the vast majority come from use in general population. The scale, complemented with proxy informants rather than being self-informed, is made potentially adequate for the evaluation of psychopathology in adults with ID.

Validity: Its factorial structure has been confirmed by several authors, demonstrating its ability to be sensitive to behavioral changes in most studies that measured the effects of the treatment (Aman et al., 1985). Tenneij's study (Tenneij & Koot, 2007) examined the reliability and validity of the scale with 124 adults with mild ID. Inter-rater reliability was good and DSM-IV-TR criteria for particular disorders correlated well with corresponding disorders on the ABCL.

Assessment of Dual Diagnosis (ADD)

Author(s) and year: Matson et al., 1998.

Provenance: USA.

Usage: Adults with mild and moderate ID.

Evaluators: Proxy informant.

Duration of test: 20 min.

Objective: To detect psychopathologic disorders in people with ID.

Dimension/score: It is composed of 79 items and is a screening instrument. The items are related to the DSM-IV criteria. It contains 13 subscales: *1—Mania*; *2—Depression*; *3—Anxiety*; *4—Post-traumatic stress disorder*; *5—Substance abuse*; *6—Somatoform disorder*; *7—Dementia*; *8—Behavioral disorder*; *9—Generalized developmental disorder*; *10—Schizophrenia*; *11—Personality disorder*; *12—Eating disorder*; and *13—Sexual disorders*. It evaluates the frequency, duration and severity of the symptoms, with an emphasis on those detected in the month previous to the evaluation.

Antecedents/description: The ADD has developed according to reported symptoms by the clinical investigation of people with mild or moderate ID. The items that make up the scale derive from the DSM-IV, as well as from published studies with this population (Aman et al., 1985). Through this scale it was possible to find a larger rate of psychopathology in people with mild or moderate ID, in comparison to people with severe and profound ID (Matson & Bamburg, 1998).

Validity: Convergent validity was obtained for the ADD with other well-established evaluation instruments of psychopathology in the population with ID, including the Psychiatric Assessment Schedules for Adults with Developmental Disabilities—short version (Mini-PAS-ASS), the Diagnostic Assessment for the Severely Handicapped (DASH-II) and the Reiss Screen (Myrbakk & von Tetzchner, 2008). These authors evaluated 17 adults with all levels of ID and realized these scales assessed different aspects of the psychopathology and that, along with behavior disorders, it occurs with high percentages. There are few recent studies that prove its validity and reliability.

The Brief Symptom Inventory (BSI)

Author(s) and year: Derogatis, 1983.

Provenance: USA.

Usage: Hospitalized psychiatric patients or outpatients over the age of 13.

Evaluators: Proxy informant.

Objective: To provide a brief description of the symptoms and their intensity in patients with psychologic and psychiatric treatment. It can also provide information in different moments of the treatment.

Dimension/score: The scale is a general screening tool that has been widely used in general psychiatry, and has 53 items that encompass nine symptomatic dimensions: *1—Somatization*; *2—Obsessive-compulsive*; *3—Interpersonal sensitivity*; *4—Depression*; *5—Anxiety*; *6—Hostility*; *7—Phobic anxiety*; *8—Paranoid ideation*; and *9—Psychoticism*. It has a self-informed version.

Antecedents/description: Several studies showed that the BSI had an adequate internal consistency for general population, with alphas from 0.71 to 0.85, and four of the nine scales had an alpha score of 0.80. The BSI also had an adequate validity in its construction, with a nine-factor structure (Derogatis, 1983).

Validity: The study led by Kellet (Kellett, Beail, & Newman, 2003), the first to use this instrument for people with mild ID, evaluated the brief use of this instrument where each element is classified in a 5-points Likert scale. The results suggest that the BSI has a practical use and adequate validity and internal consistency (Cronbach's alpha between 0.63 and 0.7). The use of the normal varimax method of rotation obtained a derivation in eight interpretable factors and a high level of overlapping with the existent factorial structure. Another study, by Wieland (Wieland, Wardenaar, Fontein, & Zitman, 2012), applied to outpatients with ID, obtained an adequate internal consistency, supporting its usage. Nonetheless, it is one of the instruments with the lowest number of validity and reliability studies (Matson et al., 2012).

Child Behavior Checklist (CBCL)

Author(s) and year: Achenbach et al., 1983.

Provenance: Vermont, USA.

Usage: Children and, later, children with ID.

Evaluators: Proxy informant.

Duration of test: Indeterminate.

Objective: This instrument is heavily used to evaluate psychopathologies in children in general. Given its acceptance, its use in children with ID is not surprising.

Dimension/score: Achenbach and Edelbrook's (1983) CBCL has eight scales: *1—Aggressive behavior*; *2—Anxiety*; *3—Depression*; *4—Attention problems*; *5—Disruptive behavior*; *6—Somatic complaints*; *7—Social problems*; and *8—Alterations of thought*.

After 2001, it also includes a set of scales based on the DSM-IV according to items of the diagnosing criteria for said disorders. It also uses a normative sample to create standard scores. These are set where 50 is the age and gender average of the child, with a standard deviation of 10 points. Any score below percentile 93 is considered normal, scores between percentiles 93–97 are clinically borderline, and any score above percentile 97 is in the clinical range.

Antecedents/description: It is a widely used scale to identify problematic behavior in child and juvenile populations, and has a school-age version divided in groups of 6–10-year-olds and 11–18-year-olds. It has been translated to over 90 languages and includes normative data, including information from multiple societies. Due to the addition of a basic set of items since the 1980s, the scale provides a good measure for mental health and behavioral problems.

Validity: The psychometric properties have been proven in several studies, but the results do not confirm the factors established in previous studies in children without ID. Some authors (Embregts, 2000) point out poor psychometrics. Karen P. de Ruiter study's (de Ruiter, Dekker, Verhulst, & Koot, 2007) major conclusion, using a wide sample (N = 2047 of people without ID and N = 978 with ID), was that there was a higher risk of psychopathology in children with ID (Matson et al., 2012). Koskentausta, Iivanainen, and Almqvist (2004) studied 90 Finnish children aged 6 to 13. They found differences in problem behaviors based on the level of ID. Children with moderate ID had higher internalizing and externalizing scores than children with profound ID. The results regarding the validity of the instrument are limited. Nevertheless, it is one of the most used scales to evaluate psychopathology in children and young people.

Diagnostic Assessment for the Severely Handicapped (DASH-II)

Author(s) and year: Matson et al., 1991.

Provenance: USA.

Usage: Adults with severe and profound ID.

Evaluators: Proxy informant.

Duration of test: 20 min.

Objective: To assess psychopathology in people with severe and profound ID.

Dimension/score: It includes 84 items and assesses 13 mental health disorders. The subscales are: *1—Organic syndromes*; *2—Mood*; *3—Anxiety*; *4—Mania*; *5—Pervasive development disorder (PDD)*; *6—Autism*; *7—Schizophrenia*; *8—Stereotypies*; *9—Self-harming behavior*; *10—Elimination disorders*; *11—Eating disorders*; *12—Sleeping disorders*; and *13—Sexual disorders*. It measures the duration and severity of the symptoms in a 3-point scale. The frequency is classified as: 0—No occurrences; 1—It happened between one and 10 times in the previous 2 weeks; or 2—It happened more than 10 times in the previous 2 weeks. The duration is classified as: 0—Less than 1 month; 1—One to 12 months; or 2—More than 12 months. The severity of the symptoms is: 1—Did not cause any interruption or damage; 2—It caused no damages but it interrupts the activity of others at least once; or 3—Lesions or damages caused at least once. The 13 symptoms scales have cutoff scores that indicate when a diagnosis is considered necessary. For the first eight scales, the adjustment for a cutoff score for the request of a psychopathologic diagnosis is one standard deviation over the average of the standardized sample (Matson, Gardner, Coe, & Sovner, 1991).

Antecedents/description: The scale derives from the DSM-IV-TR and it evaluates the presence of psychiatric disorders in people with severe and profound ID. It has been used as a strong measure to validate other psychopathologic scales. It has also been used to study patters on behavior disorders (Matson et al., 1996).

Validity: The psychometric characteristics have been evaluated in many studies, finding reliability and test-retest values of 0.84, and frequency and duration values of 0.84 and 0.91, respectively (Sevin, Matson, Williams, & Kirkpatrick Sanchez, 1995). It is the most widely studied evaluation scale of psychopathology in people with severe and profound ID. The convergent validity of the ABC was proven by analyzing its high correlation in some of the subscales. Its validation should be studied according to the new DSM-5 criteria.

Developmental Behavior Checklist (DBC)

Author(s) and year: Einfeld et al., 1995.

Provenance: Ohio, USA.

Usage: Children in general and, later, children and adults with ID.

Evaluators: Proxy informant.

Duration of test: Indeterminate.

Objective: To evaluate the behavioral and emotional problems in young people.

Dimension/score: The DBC (Einfeld & Tonge, 1995) is a questionnaire answered by parents, primary carers or teachers. It shares its structure with the CBCL (Achenbach & Edelbrook, 1983) which means that each behavioral description has a value range between 0 and 2 (0—Not true; 1—Sometimes certain; and 2—Very certain). The items are derived from a medical study with 7000 children with ID. There are five versions: the parent/carer version (DBC-P); the teacher version (DBC-T); the adult version (DBC-A); the short version (DBC-P24); and the follow-up table (DBC-M). It has been translated into other languages: Dutch, Finnish, and French. Furthermore, it provides a scoring software that is very useful for registering information.

The DBC-P has 96 items used to evaluate behavioral and emotional problems in young people between 4 and 18 years old with ID and development disorders. The DBC-A is an instrument with 107 items to evaluate the behavioral and emotional problems of adults with ID.

Validity: It has high inter-evaluator reliability between parents and teachers. The reliability and internal consistency test is also high. The validity is high, with the existence of a high correlation with the American Association for Mental Deficiency Adaptive Behavior Scales (AAMD ABS) (Perry & Factor, 1989) and the independent behavior scale Inventory for Client and Agency Planning (ICAP) (Delfín Montero, 1993).

Nisonger Child Behavior Rating Form (N-CBRF)

Author(s) and year: Aman et al., 1996.

Provenance: Ohio, USA.

Usage: Children and adolescents with ID.

Evaluators: Informants (parents and/or teachers).

Duration of test: 15 min.

Objective: To evaluate the behavior of children and adolescents with ID.

Dimension/score: It is an adapted behavior scale, containing 76 items, 10 evaluating social competence and 66 behavioral problems. It is composed of three sections: in the first, the qualifiers can identify unusual circumstances that might have influenced the behavior; in the second, the competency behaviors are classified; and the third consists of a list of behavior problems. The informant rates the child's behavior between 0 and 3 in a Likert scale.

Antecedents/description: The authors (Aman, Tassé, Rojahn, & Hammer, 1996) point out that the N-CBRF has been modified through a change in the instructions and the addition of new items that describe the behavior problems that happen in children with ID. Its subscales are: *1—Irritability, agitation, crying*; *2—Lethargy, social isolation*; *3—Stereotyped behavior*; *4—Hyperactivity/noncompliance*; and 5—

Inappropriate speech. In its factorial analysis, it obtains a six-factor solution. Nisonger's CBRF has a considerable contents overlap with the ABC, but they also differ in other important contents. The ABC was mainly developed in order to monitor the effects of treatment, while the N-CBRF was meant for evaluation purposes.

Validity: Several investigations gathered that the scale is reliable and valid for the assessment of children and adolescent behavior. The authors (Lecavalier, Aman, Hammer, Stoica, & Mathews, 2004) found that the internal consistency was high, the degree of agreement from parents and teacher was satisfactory and the subscales were similar to the ABC. Sarimski (2004) described a German sample and concluded that the N-CBRF was reliable and had a good convergent and divergent validity. Lecavalier's study (Lecavalier, 2006) evaluated 487 people with pervasive development disorders and used conglomerate analysis to define subgroups within this sample, categorized as: no problems, well adapted, hyperactive, anxious, and undifferentiated behavior disorder.

It is an instrument popularly used in measuring emotional problems, where the psychometric evaluation of the scale obtains mixed results. The majority of investigation seems destined to demonstrate and/or justify the effects of some psychotropic drugs and therefore its use in this area is limited (Matson et al., 2012).

Psychopathology Checklists for Adults with Intellectual Disability (PAID)

Author(s) and year: Hove et al., 2008.

Provenance: Norway.

Usage: Adults with ID.

Evaluators: Proxy informant.

Duration of test: Undetermined.

Objective: To evaluate the psychopathology in adults with ID.

Description/dimension: The PAID represents the first attempt at developing control-lists for psychopathologies based on the *Diagnostic criterion for psychiatric disorder for use with adults with learning disabilities/mental retardation* (DC-LD) and, in addition, contains references to the *International Classification of Diseases, 10th Edition* (ICD-10). It contains a total of 280 items and comprises 10 psychiatric diagnosis and 8 types of behavioral problem. 18 lists in total were developed, each indicative of a disorder containing a set number of items: *Dementia* (25 items), *Psychosis* (40 items), *Depression* (37 items), *Mania* (24 items), *Anxiety* (93 items), *Obsessive-compulsive disorder* (5 items) and *Behavioral problems* (61 items).

Validity: It is a fairly recent instrument with a reliability of between 0.63 and 0.88 and an acceptable internal consistency, where the specificity was low. Its factorial analysis supported a four-factor solution (Hove & Havik, 2008). Overall, the psychometric properties of the scale seem consistent, despite some scores being below the recommended level, such as the subgroup of people with severe and profound ID. The scale is promising but it requires additional studies (Matson et al., 2012).

Psychiatric Assessment Schedules for Adults with Developmental Disabilities (PAS-ADD)

Author(s) and year: Moss et al., 1993.

Provenance: London.

Usage: Adults with mild and moderate ID.

Evaluators: Proxy informant and subject (self-informed version).

Duration of test: Indeterminate/depending on the version.

Objective: To evaluate mental health problems in people with ID.

Dimension/score: It collects information regarding different situations that may occur to a person with mental health problems. The person delivering the questionnaire should know the subject for longer than 6 months.

Antecedents/description: It is a semi-structured interview designed to detect mental disorders in people with ID (Moss et al., 1993). It includes the exploration of the current state of the patient followed by an interview with a proxy informant. The assessment allows a diagnosis based solely on the interview to an informant if the patient's language skills are insufficient. The ICD-10 version of the PAS-ADD (PAS-ADD-10) provides a means to extend the scoring principles used in a more general clinical instrument to this specific population. It is based on version 1 of the SCAN and it contains items that cover the following ICD-10 diagnosis: F20.1–9—Schizophrenia; F29—Unspecified non-organic psychosis; F30.0—Hypomania; F32.0–3—Depressive episode; F40.0–2—Phobic anxiety disorders; F41.0–1—Panic and generalized anxiety disorders; and F51.1—Non-organic hypersomnia. The Mini-PAS is a variant of the PAS-ADD with 86 items that cover: *depression*, *anxiety*, *mania*, *obsessive-compulsive disorder*, *psychosis*, *unspecified disorders (with items for dementia and organic symptoms)* and *autism spectrum disorders*.

Validity: The inter-evaluator reliability of the ICD-10 detected an average kappa of 0.65 for individual item scores and a kappa of 0.7 for the agreement in the definition indexes of the SCAN (Schedules for Clinical Assessment in Neuropsychiatry) system, developed by the World Health Organization (Moss, Prosser, & Goldberg, 1996). There is a large amount of high-quality research for the PAS-ADD, and in the United Kingdom it is a well-established scale for measuring the psychopathology in people with ID. As such, it is equivalent to what the DASH-II and the PIMRA have become in North-America (Matson et al., 2012).

Psychopathology Instrument for Mentally Retarded Adults (PIMRA)

Author(s) and year: Matson et al., 1984.

Provenance/adaptations: USA.

Usage: Adolescents and adults with mild and moderate ID. Not appropriate for children.

Evaluators: Proxy informant and self-informed version.

Duration of test: Informant version takes between 6 and 12 min.

Objective: To diagnose psychopathology in people with ID.

Dimension/score: This symptom inventory contains five items that cover a broad psychopathologic spectrum based on the DSM-III-R criteria modified to fit subjects with mental retardation. It provides a score for eight types of disorders or subscales: *1—Schizophrenia*; *2—Affective disorders*; *3—Psychosexual disorders*; *4—Adaptive disorders*; *5—Anxiety disorders*; *6—Somatoform disorders*; *7—Personality disorders*; and *8—Social adaptation disorders* (Matson, Kazdin, & Senatore, 1984).

Its second edition (PIMRA-II) covers more psychopathologic disorder types, containing the original PIMRA's 56 items plus 32 new ones (5 items for autism spectrum disorder [ASD], 7 for attention deficit/hyperactivity disorder [ADHD], 4 for mania, 5 for anxiety, 4 for depression, 3 for behavior, 1 for personality disorder, 1 for sexual disorder, 1 for pica and 1 for hoarding). DSM-5 and ICD-10 guidelines and items from other scales, like the DASH-II and the ADD, were included. Its psychometric properties are comparable with the original PIMRA, with an extensive variety of items, and therefore it is an improved evaluation instrument to detect psychopathologies in people with mild and moderate ID.

Antecedents/description: The scale was developed as a selection instrument to help in the differential diagnosis of people with ID. It was the first standardized instrument designed to evaluate psychopathologies in this group. It has two versions, one for the informant and one for the patient, both with the same number of items.

Validity: It is a frequently studied instrument and it is used as a referent to establish the validity of other psychopathology evaluation instruments in people with ID. The recently obtained validity of this instrument shows good reliability, where the majority of the items obtained coefficients over 0.70, in addition to positive correlations (intraclass correlation coefficient [ICC] = 0.70) with the total ADD score.

Future investigation should examine the validity of the self-informed version of the PIMRA-II, given that internal states often cannot be behaviorally evident. Recent investigation suggest that many individuals with ID are capable of answering the self-informed questions (Douma, Dekker, Verhulst, & Koot, 2006).

Reiss Screen for Maladaptive Behavior (RSMB)

Author(s) and year: Reiss, 1987.

Provenance: Ohio, USA.

Usage: Adolescents and adults with ID.

Evaluators: Proxy informant.

Duration of test: 10 min.

Objective: To evaluate psychiatric symptoms or categories of maladaptive behavior in adolescents and adults with ID.

Dimension/score: The scale has different versions, covers all the psychopathologic range and has 38 symptoms (Reiss, 1987) organized in eight subscales: *1—Aggression scale*; *2—Autism scale*; *3—Psychosis scale*; *4—Paranoia scale*; *5—Depression (behavioral) scale*; *6—Depression (physical) scale*; *7—Dependent personality*; and *8—Avoidant personality disorder*. Each item is classified in a three-point scale regarding the functioning basis of the person during the previous 2 months. It measures the severity index of the problems of the person. The total score is the sum of the 26 points that have available normative data, and a cutoff score of 9 has been proposed to differentiate psychiatric diagnosis.

Antecedents/description: Each item is defined in nontechnical language and includes concrete examples. They measure the severity of the challenging behavior, the psychiatric diagnosis and the less frequent but significant symptoms, such as suicidal behavior.

With the 1.0 version of Reiss and Valenti-Hein (1994), it was established as a 72 item evaluation instrument, classified with a severity index: *1—Not a problem*; *2—Is a problem*; and *3—Is an important problem*. There is also a version for children (4–18 years old), the Reiss Scales for Children's Dual Diagnosis, which contains 60 items that offer, similarly to the adult version, information regarding a vast range of psychiatric disorders, including hyperactivity, autism, low self-image and isolation tendency. It also provides scores for behavioral problems and significant symptoms (episodes of crying, hallucinations, pica, self-harm, sexual problems, and suicidal tendencies).

Validity: It has an estimate Cronbach's alpha of 0.91 for the total score and between 0.63 and 0.86 for the scales. The RSMB constitutes a valuable screening method. It is one of the oldest scales established to measure psychopathologies in people with ID. The studies made with important samples show that the prevalence rates obtained not only coincide with the diagnoses of the ICD-10 but also detect profiles of specific behavior. In the USA and the United Kingdom it has been suggested that, compared to others, the scale is psychometrically solid and contains normative data for these people. This instrument is one of the most studied and is translated to several languages (Matson et al., 2012).

Specific Psychopathologic Tools

Anxiety Disorders and Depression

Anxiety disorders in people with ID have been given important attention with the existence of studies that show its frequent association in this group (Holden & Gitlesen, 2004). Depressive disorders have also been examined, showing that they exist in a lot of people with ID (Cicchetti, 1994). It is known that the prevalence of depression in adults with ID varies between 2.2% and 8.9% (Deb et al., 2001). Next, we will observe the tools most used in these disorders.

Anxiety, Depression, and Mood Scale (ADAMS)

Author(s) and year: Esbensen, Rojahn, Aman, & Ruedrich, 2003.

Provenance: Ohio, USA.

Usage: People with ID over the age of 10.

Evaluators: Proxy informant.

Duration of test: Indeterminate.

Objective: To evaluate symptoms of anxiety, depression, and mania in people with ID.

Dimension/score: The instrument contains a total of 28 items that score in a 3-point scale combining the frequency and the severity of the behavior. The items measure: *anxiety*, *fear*, *compulsion*, *appetite*, *communication*, *concentration*, *depression*, *energy levels*, *mood*, *physical state*, *sleep disorders,* and *social interaction*. Each element is classified between 0 and 3, scoring the frequency and the severity of the behavior or symptom.

Antecedents/description: Initially, Esbensen created a list of 131 items using criteria from the DSM-IV, from previous assessment instruments (Prout-Strohmer Assessment System, DASH-II, PIMRA and Self-Report Depression Questionnaire [SRDQ]) and from clinical evaluation of people in the Nisonger Center.

Validity: Esbensen's study (Esbensen et al., 2003) demonstrated that the ADAMS was a valid instrument for the screening of bipolar disorder, depression, and obsessive-compulsive disorder. Additional studies with a larger sample are required, as well as investigation that assesses the convergent and differentiating validity of the 28 items.

Beck Depression Inventory (BDI-II)

Author(s) and year: Beck, 1996.

Provenance: USA.

Usage: Adults without ID, over 13 years old.

Evaluators: Proxy informant.

Duration of test: Indeterminate.

Objective: To measure the severity of depression.

Dimension: The BDI-II (Beck, Steer, & Brown, 1996) contains 21 multiple answer questions. It is composed by items pertaining to depressive symptoms, like despair and irritability, to feelings of guilt or punishment, as well as physical symptoms related to depression (for example, fatigue, weight loss, and loss of sexual appetite). There are multiple versions and the instrument is used more often in investigation; it has been translated in multiple languages.

Validity: Authors such as Kazdin (Kazdin et al., 1983) have studied previous versions of the BDI in people with ID through a version with modified language in a sample of 110 people with limit to severe ID between the ages of 18 and 71 years old. The scales used (BDI, Minnesota Multiphasic Personality Inventory [MMPI], Thematic Apperception Test [TAT] and PIMRA) correlated significantly with each other and related constantly with the diagnostic of depression.

There are not many studies using the scale specifically for people with ID (Hermans & Evenhuis, 2010); one of the most recent studies is Powell's (Powell, 2003), where positive results for use in people with ID were obtained. The findings suggest that the test has good clinical utility and that depression can be more severe in these people.

Glasgow Anxiety Scale for People with an Intellectual Disability (GAS-ID)

Author(s) and year: Mindham et al., 2003.

Provenance: United Kingdom.

Usage: Adults with mild and moderate ID.

Evaluators: Proxy informant.

Duration of test: Indeterminate.

Objective: Scale for the detection of anxiety disorders in people with ID.

Dimension/score: The GAS-ID is a self-informed questionnaire containing 27 items regarding physiologic symptoms of anxiety and specific worries and fears. The total score varies between 0 and 54 (Mindham & Espie, 2003).

Validity: The authors studied its reliability and validity in 35 adults with ID and found an internal consistency of 0.96, a test-retest reliability of 0.95 and a sensitivity and specificity of 100%.

Hermans' study (Hermans, Wieland, Jelluma, Van der Pas, & Evenhuis, 2013) obtained the reliability and validity of the Dutch translation of the GAS-ID in 195 adults with mild or moderate ID, showing a good reliability, with an internal consistency of 0.86 and a test-retest reliability of 0.89. The validity is moderate, with a cutoff score of 17. Comparing with Mindham and Espie's (2003) results, it is demonstrated that the reliability coefficients are smaller, although still good. In conclusion, the GAS-ID seems like a reliable screening tool both for investigation and for clinical practice.

Glasgow Depression Scale for People with a Learning Disability (GDS-LD)

Author(s) and year: Cuthill et al., 2003.

Provenance: Royal College of Psychiatrists, Prescot Street, United Kingdom.

Usage: Adults with mild and moderate ID.

Objective: To detect depressive symptoms.

Dimension/score: 17 items were selected and four more not superimposed items were extracted from the ICD-10 (World Health Organization, 1992) and from the DSM-IV (American

Psychiatric Association, 1994). Several published depression scales were also reviewed: BDI-II (Beck et al., 1996), Hamilton Depression Rating Scale (Hamilton, 1960) and Zung Self-rating Depression Scale (Zung, 1965). From these, seven additional elements were aggregated to the group, forming 28 items in total.

Antecedents/description: The scale was developed according to a content revision of the existing diagnostics criteria and of the symptom scales for identifying a descriptive group of items. The starting point was the DC-LD depression subscale, developed by the Royal College of Psychiatrists (2001) for people with learning disabilities, which contains elements not included in the diagnostics programs developed for general population (Cuthill, Espie, & Cooper, 2003).

Validity/reliability: The results showed that the GDS-LD differentiated between depression and non-depressive groups correlated with the BDI-II ($r = 0.88$). It obtained a good retest reliability ($r = 0.97$), good internal consistency (Cronbach = 0.90), and a cutoff score of 13 determined a sensitivity of 96% and a specificity of 90%. The scale seems useful for screening, follow-up and contributes to the assessment of the results.

Zung Self-Rating Anxiety Scale (SAS)

Author(s) and year: Zung, 1971.

Provenance: United Kingdom.

Usage: People with normal IQ.

Objective: To evaluate anxiety and the effects of treatment.

Description: The SAS (Zung, 1971) evaluates anxiety and the effects of treatment in a clinical population with normal CI. It consists of 20 questions, to which the interviewee answers using a four-point Likert scale. Each question references some general aspect of nervousness or anxiety.

The adaptation of this scale for using with adults with mild and moderate ID was done by Lindsay (Lindsay & Michie, 1988). The 20 items were reformulated and the answer format was simplified to the presence or absence of symptoms.

Validity: In Ramirez study (Ramirez & Lukenbill, 2008), the authors analyzed the psychometric properties of a SAS adaptation. The internal consistency and the reliability coefficient were 0.80 and the correlation with other anxiety-measuring tools was low to moderate. It was concluded that, even though the present study provides preliminary data of the acceptable psychometric properties of the SAS-ID, more investigation is required to substantiate the findings, especially with samples representing other geographic areas and socioeconomic groups.

There is evidence of good test-retest reliability, with the discriminating validity being particularly necessary. Nonetheless, it is considered potentially useful for investigation and clinical purposes, as well as to assess the effectiveness of the treatment.

Scheme for Appraisal of Emotional Development (SAED)

Author(s) and year: Došen, 1990.

Provenance: Belgium.

Usage: Children, adolescents and adults with ID.

Evaluators: Proxy informant.

Antecedents: Došen's SAED (Došen, 1990) constitutes a change in the development perspective. It is innovative and very useful for the assessment, diagnostic and psychopathologic treatment of people with ID.

Researchers from different countries (Holland, Belgium, Germany and Italy) agree, in their studies, with the practical relevance of the SAED (Sappok et al., 2013), although the tool had not been widely validated until 2012. Then, Došen and a group of Belgian professionals took the initiative of making the SAED more widely available for use in clinical practice and, as a result, the Scale for Emotional Development—Revised (SED-R) was developed. In 2016, Stijin Vandevelde presented a systematic description of the SED-R development as well as the results of a study of the tool's reliability (Vandevelde et al., 2016).

Objective: To evaluate the emotional development of children, adolescents, and adults with ID. It is based on the typical emotional development that happens until the age of 12 and must be considered from the point of view of a continuum, incorporating adaptive and maladaptive behavior and psychiatric disorders.

Dimension/score: It is based on the observation of behavior through a semi-structured interview. Došen's SAED consisted of 10 domains or subscales, each with three items. These domains are described as follows: *1—How the person takes care of their own body*; *2—The interaction with the carer*; *3—The interaction with the peers*; *4–The manipulation of material objects*; *5—Affective component*; *6—Verbal communication*; *7—Anxiety*; *8—Object permanence*; *9—The experience of the self*; and *10—Aggression control*. The emotional development was assessed according to what had been established by Došen: *1—Adaptation (0–6 months)*; *2—Socialization (6–18 months)*; *3—Individuation (18 months-3 years old)*; *4—Identification (3–7 years old)*; and *5—Reality Awareness (7–12 years old)*. Three more domains were included in the SED-R, for a total of 13 subscales. The additions were: Daily activity—play development, Moral development, and Emotional regulation.

Validity: The SED-R reliability results indicate high internal consistency and substantial inter-evaluator reliability, although not for all domains. 10 of the 13 domains obtained moderate to substantial reliability between the evaluators. The advantage of using this tool resides in its focus on the importance of basic needs and motivations. It can be considered an adequate tool for integrating information regarding emotional development in the assessment and support planning processes. More investigation is required regarding its psychometric properties.

The Mood, Interest & Pleasure Questionnaire (MIPQ)

Author(s) and year: Ross et al., 2003.

Provenance: Birmingham, United Kingdom.

Usage: Adults with severe and profound ID.

Evaluators: Proxy informant.

Duration of test: 25 min.

Objective: To evaluate the person's mood and interest-pleasure.

Dimension/score: It is a 25-items Likert questionnaire with two subscales (mood, and interest and pleasure). The scale assesses the mood and the interest-pleasure for an activity in people with severe ID. It must be complemented by a qualified professional who knows the person well, preferably a psychologist. Each item must be answered with what has been observed regarding the subject in the two previous weeks (Ross & Oliver, 2003).

Antecedents/description: The rating of the MIPQ items is designed to match mood and interest-pleasure levels and it is based on the DSM-IV's description of the main depression symptoms (low mood and anhedonia), as well as behaviors regarding interest-pleasure, selected by the close correspondence to the main symptoms of major depression in the DSM-IV.

Validity: The first study's results regarding the psychometric properties, made by Ross (Ross & Oliver, 2003), are encouraging given that they obtained a good rate of validity and reliability. As a measure of emotional correlation, it offers an alternative perspective to the phenomenology of depression and to the assessment of the interventions. Petry's study (Petry, Kuppens, Vos, & Maes, 2010) suggests that the proposed factorial structure did not show an adequate adjustment to the data. A 3-factor factorial analysis (positive mood, negative mood, and interest) obtained a better adjustment. Its validation requires additional investigation, including other measurements of the individual's well-being.

Dementia

The life expectancy in people with ID is increasing, due to the improvement in medicine and in living conditions (Eyman & Call, 1991), and these people should have an equal life expectancy to the general population (Patja, Iivanainen, Vesala, Oksanen, & Ruoppila, 2000). In people with Down syndrome (DS), advanced-stage dementia usually sets in at around 50 years old (Janicki & Dalton, 2000).

The Adaptive Behavior Dementia Questionnaire (ABDQ)

Author(s) and year: Prasher et al., 2004.

Provenance: Birmingham, United Kingdom.

Duration of test: 10 min.

Objective: To identify the presence of dementia in adults with DS.

Description: The decrease in the functioning of adaptive behavior beyond the reference levels is a characteristic for the diagnosis of dementia. The ABDQ (Prasher, Farooq, & Holder, 2004) is a tool that detects changes in the adaptive behavior and is designed specifically to detect dementia in Alzheimer's disease in adults with Down syndrome. The questionnaire has 15 items and is derived from the ABS (Nihira et al., 1975).

Validity: The ABDQ is reliable and valid, and shows an excellent global precision (92%) when identifying dementia in adults with Down syndrome. The informants provide a relevant assessment of a variety of daily life activities that include getting dressed, hygiene, eating, manual dexterity, and geographic orientation.

The Cambridge Examination for Mental Disorders of Older People with Down Syndrome and Others with Intellectual Disabilities (CAMDEX-DS)

Author(s) and year: Ball et al., 2004.

Provenance: United Kingdom.

Usage: People with Down syndrome.

Evaluators: Subject and proxy informant.

Duration of test: 40 min.

Objective: To detect dementia in people with Down syndrome.

Dimension/score: It contains three parts: an interview to the subject, another to the proxy informant, and the third is a neuropsychological battery (Cambridge Cognitive Examination for Older Adults with Down syndrome—CAMCOG-DS).

Description: The CAMDEX-DS (Ball et al., 2004) is a diagnostic tool for the detection of dementia in people with DS and other causes of ID. It was originally developed in 1986 (Roth, Tym, & Mountjoy, 1986) as a standardized tool for diagnosing cognitive deterioration in general population. It is a diagnostic tool divided into two sections, a structured interview with an informant (family member or carer) who provides information regarding the current state of the patient and the patient's medical history and a direct assessment of the patient. The brief structured clinical interview collects information regarding the current mental health status as well as additional information regarding the dementia symptoms the patient presents. It also includes a brief neuropsychological battery (CAMCOG-DS) that allows the assessment of the cognitive functions affected by the dementia. The structured summaries of the CAMDEX, ICD-10, and DSM-IV diagnostics criteria for dementia are presented in list form. The information gathered through the informant interview is used to determine if the criteria for dementia are met and is complemented by the CAMCOG-DS results.

Validity: The sensitivity and the specificity of the CAMDEX-DS for the diagnosis of dementia in English-speaking people with ID are 0.88 and 0.94, respectively.

Dementia Questionnaire for Persons with Mental Retardation (DMR)

Author(s) and year: Evenhuis, 1992.

Provenance: Netherlands.

Objective: To evaluate cognitive deterioration.

Description/dimension: This is the best-known tool for the assessment of dementia in adults with ID (Evenhuis, 1992). It is a screening tool to assess the suspicion of cognitive deterioration that causes the recommendation of an exhaustive study. It is formed by a total of 50 items, grouped into eight scales: *1—Short-term memory*; *2—Long-term memory*; *3—Temporal-spatial orientation*; *4—Speech*; *5—Mobility*; *6—Humor*; *7—Activities and interests*; and *8—Behavioral changes*.

The sum of these scales obtains two indexes, which are calculated separately: a cognitive index (cognitive scores [SCS], sum of the scales for short- and long-term memories and temporal-spatial orientation) and a social index (social scores [SOS], sum of the scales for speech, mobility, activities and interests, and behavioral changes).

There are numerous investigations using the DMR as screening tool for the detection of dementia (Boada-Rovira, Hernandez-Ruiz, Badenas-Homiar, Buendia-Torras, & Tarraga-Mestre, 2005; Prasher, Adams, & Holder, 2003).

Validity: Evenhuis reported a DMR sensitivity of 100% for the identification of dementia and suggested cutoff scores indicative of cognitive deterioration. Prasher's study (Prasher, 1997), where he evaluates 100 people with SD, suggests modifications to the cutoff score in this population according to the level of ID. It is concluded that the DMR, when using the modified criteria, may help screen dementia in the population with Down syndrome.

Dementia Rating Scale (DRS)

Author(s) and year: Mattis, Bellak, & Karasu, 1976.

Provenance: New York, USA.

Usage: General population.

Objective: To evaluate cognition in dementia.

Description and subscales: Mattis' DRS (Mattis et al., 1976) is a neuropsychological battery designed to assess dementia, validated by Marson et al., in 1997 (Marson, Dymek, Duke, & Harrell, 1997), from the Department of Neurology and Alzheimer's Disease Center, University of Alabama, in Birmingham.

It has five rating criteria: *1—Wechsler Attention/Concentration Index*; *2—The Controlled Oral Word Association (CFL)* (Benton, Varney, & Hamsher, 1987); *3—The Wechsler Adult Intelligence Scale's (WAIS) block design subtest* (Wechsler, 1955); *4—The WAIS similarities subtest*; and *5—The DRS MEM subtest*, consisting on short-term recovery tasks.

Validity: The validity of the construct and the sensitivity of each DRS subscale were demonstrated by Marson et al. (1997) in a study of people with Alzheimer's disease. An internal consistency of 0.90 was established for the total scale in a geriatric population hospitalized with neurologic disorders (Gardner, Oliver-Munoz, Fisher, & Empting, 1981).

The studies that test the DRS in people with ID and, especially, DS only included a total of 147 participants (Das, Divis, Alexander, Parrila, & Naglieri, 1995; McDaniel & McLaughlin, 2000). Therefore, to support the findings further, more investigation using this tool is required, particularly including a larger sample (Elliott-King et al., 2016).

Dementia Screening Questionnaire for Individuals with Intellectual Disabilities (DSQIID)

Author(s) and year: Deb et al., 2007.

Provenance: Birmingham, United Kingdom.

Usage: Adults with DS and adults with ID.

Evaluators: Proxy informant.

Duration of test: 15 min.

Objective: To evaluate dementia in people with ID.

Description/score: The DSQIID is a questionnaire evaluated by an observer filled by the carers of people with DS who have known the person for some time (Deb, Hare, Prior, & Bhaumik, 2007). It is divided in three parts. The first asks questions regarding the "best" capacity that the person has or has had. The second contains 43 questions regarding the behavior or symptoms that are generally associated with dementia in adults with DS. Each item is scored in a four-point scale, scoring the changes in the behavior and not just the current behavior. The third part of the DSQIID contains 10 questions, all of which are comparative; for example, "speaks less" and "generally looks more tired." A "yes" answer is scored a 1 and a "no" is scored a 0. The second and third parts' scores are aggregated to get a total score.

The 53 items of the DSQIID cover areas such as memory loss, confusion, loss of skills, social isolation, behavioral changes, psychological symptoms, physical symptoms, sleep changes, and speech abnormalities.

Psychometric characteristics: A cutoff score of 20 allowed for optimal sensitivity and specificity, with 92% and 97% respectively. The DSQIID is quick (10–15 min) and easy to score. The single and fixed cutoff may limit its usefulness in more advanced stages of dementia and in people with different degrees of ID (Clune & O'Caoimh, 2013).

Gedye Dementia Scale for Down Syndrome (G-DSDS)

Author(s) and year: Gedye, 1995.

Provenance: Canada.

Usage: Adults with DS and adults with ID.

Evaluators: The questionnaire is answered by a family member or carer.

Duration of test: 30 min.

Objective: To detect dementia in people with ID.

Description: This scale is a tool based on informants created by Gedye (Gedye, 1995). It was developed in British Columbia (Canada) and standardized in 70 adults with, for the larger part, a severe or profound degree of DS. It was standardized in Ontario, with the psychiatrist from the Rideau Regional Centre Dr. Bruce McCreary, the psychologist Ms. Margie Thake, staff, and adults with severe or profound DS.

Validity: The inter-evaluator reliability kappa coefficient was 0.91 and the convergent validity coefficient was 0.81. In people with DS, the G-DSDS has good specificity but a mediocre sensitivity. An independent study by Deb and Braganza (1999), performed in adults with mild and moderate DS, found a specificity of 0.8 and a sensitivity of 0.85. The G-DSDS obtained a correlation of 0.87 with the DMR (Evenhuis, 1992).

To this date, the tool is used in a large part of the USA, Canada, Europe, Japan, and Brazil.

The National Task Group Section on Early Detection and Screening for Dementia (NTG-EDSD)

Author(s) and year: Esralew et al., 2013.

Provenance: United States. Members of the National Task Group Section on Early Detection and Screening for Dementia.

Antecedents/description: The NTG-EDSD (Esralew et al., 2013) is a classification tool based on the information provided by a third person, aimed at adults with intellectual or developmental disorders suspected of cognitive, behavioral and skill changes. It is a screening tool not meant for clinical assessment. The NTG-DST is an instrument adapted from several sources, including the DSQIID (developed by Deb et al., 2007) and the Dementia Screening Tool (2010; adapted from the DSQIID by the Philadelphia Coordinated Health Care Group). The NTG-DST also uses comorbidity items adapted from the Longitudinal Health and Intellectual Disabilities Survey (LHIDS), developed by Hsieh, Rimmer, and Heller (2012) at the University of Illinois, Chicago.

Autism Spectrum Disorders

ID and autism spectrum disorders (ASD) are frequently associated. In fact, it has been positioned that the most frequently associated disorders with ID are autism spectrum disorders (Wilkins & Matson, 2009). Approximately 40% of people with ID fit the diagnostics criteria for ASD (Matson & Shoemaker, 2009). Its combination with autism leads to phenotypes and more serious decline in comparison with people with only ID. Other studies have reached the conclusion that 70% of people with autism have a diagnosis of ID (Magnússon & Sæmundsen, 2001), which represents a significant proportion in severe and profound ID. Most instruments are derived from children's population, later studied and adapted to adults with ID.

The Autism Diagnostic Interview: Revised (ADI-R)

Author(s) and year: Lord et al., 1994.

Provenance: Illinois, USA.

Usage: Children aged between 4 and 5 years old.

Objective: To diagnose ASD.

Description/dimension: The ADI-R is a diagnosing interview to the parents, widely used to diagnose ASD (Lord, Rutter, & Le Couteur, 1994). It is a standardized, semi-structured interview conducted by doctors to the parents and carers of people possibly affected by ASD. It includes 93 ordinal items corresponding to different functioning domains: communication, reciprocal social interactions, and restrictive, repetitive and stereotyped behavioral patterns, as well as other behavioral aspects. Specialized training is required for the administration of the scale.

Validity: In Sappok's study (Sappok et al., 2013), conducted in a sample of adults with ID, the results for the ADI-R indicate good sensitivity (87.5%) and specificity (80%), and its psychometric properties are more balanced than the Autism Diagnostic Observation Schedule (ADOS). On the other hand it has been suggested that the ADI-R is too inclusive for people who are nonverbal or with a severe cognitive decline (Lord et al., 1994). De Bildt's (De Bildt et al., 2004) work has informed that age and level of functioning do not seem to have significant effects in the validity of the tool. ADI-R's diagnostics validity in adults with ID that show characteristics of ASD has not yet been sufficiently assessed.

Autism Checklist (ACL)

Author(s) and year: Sappok et al., 2014.

Provenance: Berlin, Germany.

Duration of test: Approximately 10 min.

Objective and usage: To detect autism during childhood.

Description: It is a screening tool that derives from the ICD-10 investigation criteria for early childhood (before the age of 3 years old), specifically for autism (F84.0) and atypical autism (F84.1x). Its use in adults with ID is recent. In the tool's psychometric study (Sappok, Heinrich, & Diefenbacher, 2014), it obtained a sensitivity of 90.7%, with an acceptable specificity of 67.6%. 11 of the 12 criteria with significant discriminating characteristics derive from the ICD-10.

Dimension/score: The ACL is a scale designed for specialized doctors or psychologists. Each ICD-10 domain (CL1—Social interaction; CL2—Social communication; CL3—Stereotypies and restricted behaviors) is assessed using four ordinal item scales, which ask if a certain symptom is present, partially present or not present. The result of the assessment is obtained if a person scores at least two points in the domain 1 (CL1), at least one point in domain 2 (CL2) and domain 3 (CL3) and at least six points in the three domains.

Validity: Initial evaluation of the diagnostics validity in a sample of adults with ID in Germany showed that the tool was promising, with a sensitivity and specificity of 91% and 68%, respectively (Sappok, Heinrich, & Diefenbacher, 2014). It can be used in people with ID as a brief screening tool.

Autism Diagnostic Observation Schedule (ADOS)

Author(s) and year: Lord et al., 2000.

Provenance: Chicago, USA.

Usage: Children.

Duration of test: 30–60 min.

Objective: To detect autism.

Description/dimension: The ADOS (Lord et al., 2000) is an observation tool designed to assess social and reciprocal interaction, communication, play and imagination. Frequently used to differentiate autism diagnostics from other pervasive developmental disorders not otherwise specified (PDD-NOS), its contribution for a precise diagnosis has been proven in several studies (De Bildt et al., 2004; Lord, Rutter, DiLavore, Risi, & Gotham, 2012). It is a structured observation tool which contains 4 units for the evaluation of social and communication skills in people suspected of having autism. It can be used to plan teaching. The ADOS was originally designed to be used with the ADI and has been considered the gold standard for the assessment of ASD (Filipek et al., 2000). The ADOS and the ADI-R turned out to be valuable tools for the diagnosis of ASD in adults with ID (Sappok, Diefenbacher, et al., 2013). Specialized training is required for the administration of the ADOS.

Autism Spectrum Disorders-Diagnosis for Intellectually Disabled Adults (ASD-DA)

Author(s) and year: Matson, Wilkins, Boisjoli, & Smith, 2008.

Provenance: USA.

Usage: Young people older than 16 years old.

Evaluators: Key-informant.

Duration of test: 10 min.

Objective: To rapidly provide relevant information that allows the differentiation between people with and without ASD in people with ID.

Dimension/score: The items are scored as 0 (not different, without decline) or 1 (different, some decline) through the comparison of the objective person and people of the same age that live in the community. The highest total scores are an indication of a bigger decline.

A cutoff score of ≥19 indicates comorbid ASD, while ≥19 indicates that the adult has ID but no ASD. Scores of 11 over factor I (social decline) and 8 over factor III (restricted behavior) were established as cutoff scores to distinguish between autism and PDD-NOS.

Antecedents/description: It is a structured interview used mostly to differentiate between adults with ID and ASD (autism, pervasive developmental disorder not otherwise specified, and Asperger syndrome) and adults with only ID.

The items are derived from the DSM-IV-TR and ICD-10 criteria for autism, PDD-NOS, and Asperger syndrome. It includes items from other autism diagnosis scales (Childhood Autism-Rating Scale [CARS]) and the autism/PDD-NOS subscale from the DASH-II, as well as from an exhaustive investigation of the literature revisions regarding ASD, diagnostic guidelines and clinical specialists' observations on this population.

Validity: The initial studies established its validity and a factor structure, as well as the cut-off scores to differentiate between ASD and ID, and autism and PDD-NOS (Matson, Boisjoli, González, Smith, & Wilkins, 2007). In the broader field-test conducted on adolescents and adults with ID aged 16 to 88 years old (*N* = 307), using the DSM-IV and ICD-10 criteria, it demonstrated good sensitivity and specificity in comparison with other disorders (Volkmar et al., 1994). The convergent validity of the ASD-DA showed correlation with other symptom evaluation instruments of ASD: a checklist composed of criteria from the DSM-IV-TR and ICD-10 used for diagnosing autism and PDD-NOS (Matson et al., 2008). In a study by Planelles-Fernández et al. (2017), replicating their former one, good test-retest reliability values were obtained through the ASD-DA. The items showed a significant load in the specified factor, with a perfect adjustment to the data.

Childhood Autism Rating Scale (CARS)

Author(s) and year: Schopler, Reichler, DeVellis, & Daly, 1980.

Provenance: USA.

Usage: Children with autism.

Evaluators: Proxy informant.

Description: It is an evaluation tool developed to identify children with autism, compared with other developmental disorders, in order to determine the severity of the symptoms (Schopler et al., 1980). It was designed for children older than 2 years old and it contains 15 four-point scales, where the child's behavior is scored in chronological age. The scores are grouped to categorize autism in mild, moderate and severe.

Validity/reliability: Matson's study (Matson, Nebel-Schwalm, & Matson, 2007) obtained an internal consistency of 0.94 and an acceptable inter-evaluator reliability of 0.71. In 2004, Rellini (Rellini, Tortolani, Trillo, Carbone, & Montecchi, 2004) compared the CARS with the DSM-IV criteria, obtaining a sensitivity of 100% for children with autism.

Diagnostic Behavioral Assessment for Autism Spectrum Disorder: Revised (DiBAS-R)

Author(s) and year: Sappok et al., 2014.

Provenance: Germany.

Objective: To assess autistic behaviors.

Description/dimension: The DiBAS-R is a screening scale with 20 items based on the ICD-10 and the DSM-5. It consists of two DSM-5 domains corresponding to the evaluation of typical autistic behavior over social communication, interaction, and stereotyped and restrictive behaviors, and to the sensorial interests (Sappok et al., 2014).

The tool is a semi-structured interview aimed at the parents. The questions were written in simple language to allow the evaluation by people with no knowledge of ASD.

Validity: Sappok's study (Sappok, Heinrich, & Diefenbacher, 2014), given to a sample of 219 adults with ID, obtained significant average correlations with other scales (ACL, Social Communication Questionnaire [SCQ] and Pervasive Developmental Disorder in Mentally Retarded Persons [PDD-MRS]). This finding supports the diagnostics validity of the DiBAS-R, since all its scales aim to detect behaviors characteristic to ASD. Other recent study (Heinrich, Böhm, & Sappok, 2018) revealed, in its validity analysis, a sensitivity of 0.82 and a specificity of 0.67 in the global sample. The sensitivity (0.79) and the specificity (0.84) are more balanced in people with mild to moderate ID (83.3% agreement), while the specificity was lower in people with severe to profound ID (sensitivity: 0.83; specificity: 0.34). The level of ID, as well as its interaction with ASD, explained a significant proportion of the variation in the DiBAS-R scores. It is considered an adequate screening tool, especially in people with mild to moderate ID.

Pervasive Developmental Disorder in Mentally Retarded Persons (PDD-MRS)

Author(s) and year: Kraijer et al., 2005.

Provenance: Netherlands. The original PDD-MRS version is from 1990; since then, it has been widely used in the Netherlands.

Usage: People with mild to profound ID, between 2 and 55 years old.

Evaluators: Proxy informant.

Duration of test: Between 10 and 20 min.

Objective: The main objective of the scale is to evaluate the entire range of ASD.

Description/dimension: The PDD-MRS, or pervasive developmental disorder scale for people with ID (Kraijer & De Bildt, 2005) and, in its original Dutch version, AVZ-R, covers ASD as well as all PDD (World Health Association: ICD-10, 1992; American Psychiatric Association: DSM-III-R/-IV/-IV-TR, 1987, 1994, 2000). It contains 12 dichotomous items and assigns a higher score when the referenced behavior shows in some degree. The scores vary between 0 and 19, corresponding to the DSM-III-R criteria for autism disorder and PDD. It is a complementary test for other scales, such as the ADOS and the ADI-R. Compared to the PDD-MRS, the ADOS and the ADI-R are preferable in investigation projects.

Validity: The rules of the scale are based on investigation protocols in 1230 Dutch people with ID. Both the sensitivity and the specificity of the scale were found to be 92.4%, which is a high value. Therefore, it must be considered a useful tool in identifying PDD in people with ID.

Social Communication Questionnaire (SCQ)

Author(s) and year: Rutter Bailey & Lord, 2003.

Provenance: Los Angeles, USA.

Usage: The SCQ helps to evaluate the communication skills and the social functioning in children (4–18 year olds).

Evaluators: Proxy informant.

Duration of test: 10 min.

Objective: The early detection of ASD.

Dimension/score: It consists of a categorical scale containing 40 items, answered by the parents. Six of the items are suppressed in case the person answering is nonverbal.

A score of 1 is assigned to the presence of abnormal behavior and a score of 0 if the behavior is not present. A cutoff score of >15 indicates a possible pervasive developmental disorder and a score of 22 or more indicates autism. In one study (De Giacomo, De Giambattista, Balducci, & Craig, 2015) the patients with ID received a higher score. Recently, the use of the simple version for the screening of ASD in adults has been supported by Sappok, Gaul, et al. (Sappok et al., 2015).

Description: The SCQ derives from the ADI-R (Lord et al., 1994). The tool, developed by Michael Rutter and Catherine Lord (Rutter Bailey & Lord, 2003), is a social communication questionnaire based on the DSM-IV and on the contents of the ADI-R.

Current Situation and Future Directions

The development of psychopathologic tools for people with ID has leaped in the last three decades. A large number of tests for adults and children have appeared, some of them extensively studied both in North America and in Europe. Investigation has focused, for the most part, in a single comorbidity diagnosis associated with ID; there is less investigation regarding the presence of two or more axis I disorders co-occurring with ID (Kozlowski, Matson, Sipes, Hattier, & Bamburg, 2011). It is also a necessity to have specific scales for children and adults with mild and moderate ID and other evaluation instruments for severe and profound ID. One of the limitations of the studies made through measuring scales is the origin of the information, since the parameters that define who can and cannot provide it still have not been determined. Adults with milder ID are capable of completing the self-informed ones, whereas the very young, the ones with severe ID, or the person developing dementia or other late-onset cognitive disorders are not capable of completing the task.

Tools such as the CBCL, the DBC, and the N-CBRF are valid psychometric tests, but are only applicable to children with ID. The evaluation of psychopathological disorders in people with severe ID through the DASH-II allows a solid psychometric analysis, but it is limited to adults with severe and profound ID (Matson, Smiroldo, Hamilton, & Baglio, 1997). Among the subpopulation of people with severe and profound ID there is a lack of tools directed towards children (Hermans & Evenhuis, 2010).

The BSI is psychometrically valid but has been applied to people with ID scarcely. The PAID is promising, however it still needs further investigation to establish its psychometric properties. Lastly, the ADD, the RSMB and the PASS-ADD are strong instruments to evaluate the psychopathology in adults with mild and moderate ID, but lack the necessary range to cover the totality of the psychopathologic disorders. It is still necessary to evaluate the capacity of the subscales in the screening of depression, where sensitivity and specificity have not yet been determined (Hermans & Evenhuis, 2010). Scales such as the ABC, the DASH-II, and the MIPQ are considered to have a good level of support regarding the methodological quality for use in people with severe and profound ID. Regarding the foundation of the tools currently available, the DASH-II and the ABC are reliable instruments in the evaluation of mental disorders and the MIPQ has a good reliability measure regarding mood. The tools rarely include a reference to the theoretical process of its development (Flynn et al., 2017).

Despite this, some tools, like PIMRA's second edition (PIMRA-II), have been revised with the inclusion of more items, therefore covering a wide range of psychopathologic disorders. These reasons make the tool significant for the assessment of the psychopathology in people with mild and moderate ID. The scales will soon need to be compared to the DSM-5 (Belva & Matson, 2015).

Tools such as the DBC-P and the PAID have psychometric studies and checks with encouraging results, but still require more investigation to establish its psychometric properties. Validity and reliability tests of these tools must occur before employing them in clinical practice.

The existing tools' psychometric properties should be more studied, instead of developing new ones. It is important to improve methodological and statistical quality as well as to inform regarding useful themes for clinical practice such as application details, evaluation time, etc. (Hermans, van der Pas, & Evenhuis, 2011).

There are few tools for evaluating depression and only three are promising: the ADD, the RSMB and the CDI. However, these tools' ability to measure depression was never completely studied (Hermans & Evenhuis, 2010). In the same way, there are limited tools used for studies regarding anxiety with sufficient psychometric properties; they need to be reliable and valid (Hermans et al., 2011).

The development of tools that evaluate dementia in these people has been growing, especially considering that longevity has increased considerably. The list of tools has been increasing significantly since the most recognized scale appeared, the DMR. It is recommended the use of sets that combine the different methods used (structured interviews with informants and cognitive function tests), since they can offer more information regarding the loss of cognition (Elliott-King et al., 2016). The shorter tools are preferable when the ID is more severe. The linearity of the tests may emphasize any decline by allowing a follow-up of the cognitive functions in years previous to the onset of dementia.

The interest in the study of ASD in people with ID has been relevant in the last decades and clinical criteria that allow a precise diagnosis are necessary. This may be a complex task given that the ID are associated with changes in the three main functioning domains in which autism diagnosis is based, where communication problems, repetitive behaviors and difficulty in social interactions are a common trait (De Bildt et al., 2003). Nonetheless, some authors consider that it is possible to detect ASD co-occurring with ID and that it is possible to make the distinction between the two (Matson, Boisjoli, et al., 2007). It has also been determined that the presence of ASD can be a vulnerability factor (La Malfa et al., 2007), while others have come to the conclusion that approximately 70% of people with autism have a diagnosis of ID. A good number of the scales that have been used to evaluate were, for the most part, originally used in children and only a few tools have been developed for adults. The data regarding their validity are scarce, especially for people with lower functioning (Sappok, Heinrich, & Underwood, 2015).

An additional limitation to the existing investigation is not only the lack of data but also that the published data are often informed by the same people that developed the tools. There is, consequently, a lack of independent evidence from the tools themselves. Furthermore, in many occasions, the populations studied are for the most part in residential and hospital environments, and therefore the generalization of the results cannot be extrapolated.

The current research is focused on the tests that may help the diagnostics of the specific types of psychopathology. We are witnessing the development of these tools, this movement towards specialized scales as a continuation of the general evaluation instruments. It is possible that this continues and that it may be very valuable in order to not only improve the accuracy of the diagnoses but also to allow a better comprehension of the nature of the disorders themselves, where the specificity of the symptoms may turn out useful for the treatment (Matson & Shoemaker, 2011).

There is diversity in the lines of study of comorbidity in ID. North America has followed a distancing from the study of the different forms of psychopathology, turning its focus mainly to autism. Europe seems to have kept a more balanced focus on diverse forms of psychopathology. A positive fact is the emergence of ID investigation in Asia (China and Taiwan).

The current challenge is to be up-to-date in the broadest specialized training in the knowledge of how these disorders manifest themselves, and how they are evaluated and treated in an effective way (Matson & Shoemaker, 2011). The development of specific evaluation instruments will entail the establishment of more specialized interventions.

References

Achenbach, T. M., & Edelbrook, C. (1983). Child behavior checklist (CBCL). *University Associates in Psychiatry, 53*(1), 418–435.

Achenbach, T. M., & Rescorla, L. A. (2003). *Manual for the ASEBA adult forms & profiles*. Burlington, VT: University of Vermont Research Center for Children, Youth and Families.

Aman, M. G., Singh, N. N., Stewart, A. W., & Field, C. J. (1985). The aberrant behavior checklist: A behavior rating scale for the assessment of treatment effects. *American Journal of Mental Deficiency, 89*(5), 485–491. Retrieved from https://www.researchgate.net/publication/19167636

Aman, M. G., Tassé, M. J., Rojahn, J., & Hammer, D. (1996). The Nisonger CBRF: A child behavior rating form for children with developmental disabilities. *Research in Developmental Disabilities, 17*(1), 41–57. https://doi.org/10.1016/0891-4222(95)00039-9

American Psychiatric Association. (1987). *Diagnostic and Statistical Manual of Mental Disorders (DSM-III-R)* (3rd ed., revised). Washington, DC.

American Psychiatric Association. (1994). *Diagnostic and Statistical Manual of Mental Disorders (DSM-IV)* (4th ed.). Washington, DC.

American Psychiatric Association. (2000). *Diagnostic and Statistical Manual of Mental Disorders (DSM-IV-TR)* (4th ed., text rev.). Washington, DC.

Ball, S. L., Holland, A. J., Huppert, F. A., Treppner, P., Watson, P., & Hon, J. (2004). The modified CAMDEX informant interview is a valid and reliable tool for use in the diagnosis of dementia in adults with Down's syndrome. *Journal of Intellectual Disability Research, 48*(6), 611–620. https://doi.org/10.1111/j.1365-2788.2004.00630.x

Beck, A., Steer, R., & Brown, G. (1996). Beck Depression Inventory-II, 12–15. https://doi.org/10.1037/t00742-000

Belva, B. C., & Matson, J. L. (2015). Examining the psychometrics of the Psychopathology Inventory for Mentally Retarded Adults-II for individuals with mild and moderate intellectual disabilities. *Research in Developmental Disabilities, 36*, 291–302. https://doi.org/10.1016/j.ridd.2014.10.017

Benton, A. L., Varney, R., & Hamsher, K. (1987). Visuospatial judgement: A clinical test. *Archives of Neurology, 35*, 363–367.

Boada-Rovira, M., Hernandez-Ruiz, I., Badenas-Homiar, S., Buendia-Torras, M., & Tarraga-Mestre, L. (2005). Clinical-therapeutic study of dementia in people with Down syndrome and the effectiveness of donepezil in this population. *Revista de Neurologia, 41*(3), 129–136.

Borthwick-Duffy, S. A. (1994). Epidemiology and prevalence of psychopathology in people with mental retardation. *Journal of Consulting and Clinical Psychology, 62*(1), 17–27. https://doi.org/10.1037//0022-006X.62.1.17

Cherry, K. E., Penn, D., Matson, J. L., & Bamburg, J. W. (2000). Characteristics of schizophrenia among persons with severe or profound mental retardation. Psychiatric Services, Washington, DC, 51(7), 922–924. doi: https://doi.org/10.1176/appi.ps.51.7.922

Cicchetti, D. V. (1994). Guidelines, criteria, and rules of thumb for evaluating normed and standardized assessment instruments in psychology. *Psychological Assessment, 6*(4), 284–290. https://doi.org/10.1037/1040-3590.6.4.284

Clune, Y., & O'Caoimh, R. (2013). Screening for Alzheimer's disease in downs syndrome. *Journal of Alzheimer's Disease & Parkinsonism, 3*(3), 1–6. https://doi.org/10.4172/2161-0460.S7-001

Cuthill, F. M., Espie, C. A., & Cooper, S. A. (2003). Development and psychometric properties of the Glasgow Depression Scale for people with a learning disability: Individual and carer supplement versions. *British Journal of Psychiatry, 182*(APR), 347–353. https://doi.org/10.1192/bjp.182.4.347

Das, J. P., Divis, B., Alexander, J., Parrila, R. K., & Naglieri, J. A. (1995). Cognitive decline due to aging among persons with down syndrome. *Research in Developmental Disabilities, 16*(6), 461–478. https://doi.org/10.1016/0891-4222(95)00030-5

De Bildt, A., Sytema, S., Ketelaars, C., Kraijer, D., Mulder, E., Volkmar, F., & Minderaa, R. (2004). Interrelationship between Autism Diagnostic Observation Schedule-Generic (ADOS-G), Autism Diagnostic Interview-Revised (ADI-R), and the Diagnostic and Statistical Manual of Mental Disorders (DSM-IV-TR) classification in children and adolescents with menta. *Journal of Autism and Developmental Disorders, 34*(2), 129–137. https://doi.org/10.1023/B:JADD.0000022604.22374.5f

De Bildt, A., Sytema, S., Ketelaars, C., Kraijer, D., Volkmar, F., & Minderaa, R. (2003). Measuring pervasive developmental disorders in children and adolescents with mental retardation: A comparison of two screening instruments used in a study of the total mentally retarded population from a designated area. *Journal of Autism and Developmental Disorders, 33*(6), 595–605. https://doi.org/10.1023/B:JADD.0000005997.92287.a3

De Giacomo, A., De Giambattista, C., Balducci, R., & Craig, F. (2015). SCQ as a tool for screening ASD comorbidities with ADHD. *SCQ Come Strumento Di Screening per ASD in Comorbilita Con ADHD.* https://doi.org/10.1708/1794.19534

de Ruiter, K. P., Dekker, M. C., Verhulst, F. C., & Koot, H. M. (2007). Developmental course of psychopathology in youths with and without intellectual disabilities. *Journal of Child Psychology and Psychiatry and Allied Disciplines, 48*(5), 498–507. https://doi.org/10.1111/j.1469-7610.2006.01712.x

Deb, S., & Braganza, J. (1999). Comparison of rating scales for the diagnosis of dementia in adults with Down's syndrome. *Journal of Intellectual Disability Research, 43*(5), 400–407. https://doi.org/10.1046/j.1365-2788.1999.043005400.x

Deb, S., Hare, M., Prior, L., & Bhaumik, S. (2007). Dementia screening questionnaire for individuals with intellectual disabilities. *The British Journal of Psychiatry, 190*, 440–444. https://doi.org/10.1192/bjp.bp.106.024984

Deb, S., Thomas, M., & Bright, C. (2001). Mental disorder in adults with intellectual disability. 1: Prevalence of functional psychiatric illness among a community-based population aged between 16 and 64 years. *Journal of Intellectual Disability Research, 45*, 495–505.

Delfín Montero, C. (1993). *Evaluación de la conducta adaptativa en personas con discapacidades: Adaptación y validación del ICAP*. Mensajero.

Derogatis, L. R. (1983). The brief symptom inventory: An introductory report. *Psychological Medicine, 13*(3), 595–605. https://doi.org/10.1017/S0033291700048017

Došen, A. (1990). *Psychische en gedragsstoornissen bij zwakzinnigen*. Amsterdam: Boom.

Douma, J. C. H., Dekker, M. C., Verhulst, F. C., & Koot, H. M. (2006). Self-reports on mental health problems of youth with moderate to borderline intellectual disabilities. *Journal of the American Academy of Child & Adolescent Psychiatry, 45*(10), 1224–1231. https://doi.org/10.1097/01.chi.0000233158.21925.95

Einfeld, S. L., & Tonge, B. J. (1995). The developmental behavior checklist: The development and validation of an instrument to assess behavioral and emotional disturbance in children and adolescents with mental retardation. *Journal of Autism and Developmental Disorders, 25*(2), 81–104. https://doi.org/10.1007/BF02178498

Elliott-King, J., Shaw, S., Bandelow, S., Devshi, R., Kassam, S., & Hogervorst, E. (2016). A critical literature review of the effectiveness of various instruments in the diagnosis of dementia in adults with intellectual disabilities. *Alzheimer's & Dementia: Diagnosis, Assessment & Disease Monitoring, 4*, 126–148. https://doi.org/10.1016/j.dadm.2016.06.002

Embregts, P. J. (2000). Reliability of the Child Behavior Checklist for the assessment of behavioral problems of children and youth with mild mental retardation. *Research in Developmental Disabilities, 21*(1), 31–41.

Esbensen, A. J., Rojahn, J., Aman, M. G., & Ruedrich, S. (2003). Reliability and Validity of an assessment instrument for anxiety, depression, and mood among individuals with mental retardation. *Journal of Autism and Developmental Disorders, 33*(6), 617–629. https://doi.org/10.1023/B:JADD.0000005999.27178.55

Esralew, L., Janicki, M. P., DiSipio, M., Jokinen, N., Keller, S. M., & and Members of the National Task Group Section on Early Detection and Screening. (2013). National Task Group Early Detection Screen for Dementia manual, *Available* (May), 1–15.

Evenhuis, H. M. (1992). Evaluation of a screening instrument for dementia in ageing mentally retarded persons. *Journal of Intellectual Disability Research : JIDR, 36*(Pt 4), 337–347. https://doi.org/10.1111/j.1365-2788.1992.tb00532.x

Eyman, R. K., & Call, T. L. (1991). Life expectancy of persons with Down syndrome. *American Journal of Mental Retardation : AJMR, 95*(6), 603–612.

Filipek, P. A., Accardo, P. J., Ashwal, S., Baranek, G. T., Cook, E. H., Dawson, J., … M. (2000). Practice parameter: Screening and diagnosis of autism Report of the Quality Standards Subcommittee of the American Academy of Neurology and the Child. *Neurology, 55*, 468–479. https://doi.org/10.1212/WNL.55.4.468

Fletcher, R. J., Barnhill, J., Mccarthy, J., & Strydom, A. (2016). From DSM to DM-ID. *Journal of Mental Health Research in Intellectual DisabilitiesOnline) Journal Homepage: André Strydom Journal of Mental Health Research in Intellectual Disabilities, ISSN*, 1931–5864. https://doi.org/10.1080/19315864.2016.1185324

Flynn, S., Vereenooghe, L., Hastings, R. P., Adams, D., Cooper, S. A., Gore, N., … Waite, J. (2017). Measurement tools for mental health problems and mental well-being in people with severe or profound intellectual disabilities: A systematic review. *Clinical Psychology Review, 57*, 32–44. https://doi.org/10.1016/j.cpr.2017.08.006

Gardner, R., Oliver-Munoz, S., Fisher, L., & Empting, L. (1981). Mattis dementia rating scale: Internal reliability study using a diffusely impaired population. *Journal of Clinical Neuropsychology, 3*(3), 271–275. https://doi.org/10.1080/01688638108403130

Gedye, A. (1995). Dementia Scale for Down Syndrome. *Gedye Research & Consulting*. Retrieved from http://www.gedye.ca

Hamilton, M. C. (1960). Hamilton depression rating scale (HAM-D). *Redloc, 23*, 56–62. https://doi.org/10.1111/j.1600-0447.1986.tb10903.x

Heinrich, M., Böhm, J., & Sappok, T. (2018). Diagnosing autism in adults with intellectual disability: Validation of the DiBAS-R in an independent sample. *Journal of Autism and Developmental Disorders, 48*(2), 341–350. https://doi.org/10.1007/s10803-017-3336-5

Hermans, H., & Evenhuis, H. M. (2010). Characteristics of instruments screening for depression in adults with intellectual disabilities: Systematic review. *Research in Developmental Disabilities, 31*(6), 1109–1120. https://doi.org/10.1016/j.ridd.2010.04.023

Hermans, H., van der Pas, F. H., & Evenhuis, H. M. (2011). Instruments assessing anxiety in adults with intellectual disabilities: A systematic review. *Research in Developmental Disabilities, 32*(3), 861–870. https://doi.org/10.1016/j.ridd.2011.01.034

Hermans, H., Wieland, J., Jelluma, N., Van der Pas, F., & Evenhuis, H. (2013). Reliability and validity of the Dutch version of the Glasgow Anxiety Scale for people with an Intellectual Disability (GAS-ID). *Journal of Intellectual Disability Research, 57*(8), 728–736. https://doi.org/10.1111/j.1365-2788.2012.01632.x

Holden, B., & Gitlesen, J. P. (2004). The association between severity of intellectual disability and psychiatric symptomatology. *Journal of Intellectual Disability Research, 48*(6), 556–562. https://doi.org/10.1111/j.1365-2788.2004.00624.x

Hove, O., & Havik, O. E. (2008). Psychometric properties of Psychopathology checklists for Adults with Intellectual Disability (P-AID) on a community sample of adults with intellectual disability. *Research in Developmental Disabilities, 29*(5), 467–482. https://doi.org/10.1016/j.ridd.2007.09.002

Hsieh, K., Rimmer, J., & Heller, T. (2012). Prevalence of falls and risk factors in adults with intellectual disability. *American Journal on Intellectual and Developmental Disabilities, 11*(6), 442–454.

Janicki, M. P., & Dalton, A. J. (2000). Prevalence of dementia and impact on intellectual disability services. *Mental Retardation, 38*(3), 276–288.

Kazdin, A. E., Matson, J. L., & Senatore, V. (1983). Assessment of depression in mentally retarded adults. *The American Journal of Psychiatry, 140*(8), 1040–1043. https://doi.org/10.1176/ajp.140.8.1040

Kellett, S., Beail, N., & Newman, D. W. (2003). Utility of the Brief Symptom Inventory in the assessment of psychological distress. *Journal of Applied Research in Intellectual Disabilities, 16*(2), 127–134. Retrieved from http://onlinelibrary.wiley.com/doi/10.1046/j.1468-3148.2003.00152.x/full%5Cnpapers2://publication/uuid/1176B61A-B741-43D8-9E22-0BE102C3F1DE

Kishore, T., Nizamie, H., & Nizamie, A. (2010). Utility of reiss screen in identifying psychiatric problems in persons with mental retardation. *Indian Journal of Psychological Medicine., 32*(1), 38. https://doi.org/10.4103/0253-7176.70531

Konstantareas, M. M., & Hewitt, T. (2001). Autistic disorder and schizophrenia: Diagnostic overlaps. *Journal of Autism and Developmental Disorders, 31*(1), 19–28. https://doi.org/10.1023/A:1005605528309

Koskentausta, T., Iivanainen, M., & Almqvist, F. (2004). CBCL in the assessment of psychopathology in Finnish children with intellectual disability. *Research in Developmental Disabilities, 25*(4), 341–354. https://doi.org/10.1016/j.ridd.2003.12.001

Kozlowski, A. M., Matson, J. L., Sipes, M., Hattier, M. A., & Bamburg, J. W. (2011). The relationship between psychopathology symptom clusters and the presence of comorbid psychopathology in individuals with severe to profound intellectual disability. *Research in Developmental Disabilities, 32*(5), 1610–1614. https://doi.org/10.1016/j.ridd.2011.02.004

Kraijer, D., & De Bildt, A. (2005). The PDD-MRS: An instrument for identification of autism spectrum disorders in persons with mental retardation. *Journal of Autism and Developmental Disorders, 35*(4), 499–513. https://doi.org/10.1007/s10803-005-5040-0

La Malfa, G., Lassi, S., Salvini, R., Giganti, C., Bertelli, M., & Albertini, G. (2007). The relationship between autism and psychiatric disorders in Intellectually Disabled Adults. *Research in Autism Spectrum Disorders, 1*(3), 218–228. https://doi.org/10.1016/j.rasd.2006.10.004

Lecavalier, L. (2006). Behavioral and emotional problems in young people with pervasive developmental disorders: Relative prevalence, effects of subject characteristics, and empirical classification. *Journal of Autism* *and Developmental Disorders, 36*(8), 1101–1114. https://doi.org/10.1007/s10803-006-0147-5

Lecavalier, L., Aman, M. G., Hammer, D., Stoica, W., & Mathews, G. L. (2004). Factor analysis of the Nisonger Child Behavior Rating Form in children with autism spectrum disorders. *Journal of Autism and Developmental Disorders, 34*(6), 709–721. https://doi.org/10.1007/s10803-004-5291-1

Lindsay, W. R., & Michie, A. M. (1988). Adaptation of the Zung self-rating anxiety scale for people with a mental handicap. *Journal of Intellectual Disability Research, 32*(6), 485–490. https://doi.org/10.1111/j.1365-2788.1988.tb01440.x

Lord, C., Risi, S., Lambrecht, L., Cook, E. H., Leventhal, B. L., DiLavore, P. C., … Rutter, M. (2000). Autism diagnostic observation schedule (ADOS). *Journal of Autism and Developmental Disorders, 30*(3), 205–223. https://doi.org/10.1007/BF02211841

Lord, C., Rutter, M., DiLavore, P., Risi, S., & Gotham, K. (2012). *Autism diagnostic observation schedule, (ADOS-2)*. California: Los Angeles.

Lord, C., Rutter, M., & Le Couteur, A. (1994). Autism Diagnostic Interview-Revised: A revised version of a diagnostic interview for caregivers of individuals with possible pervasive developmental disorders. *Journal Autism Developmental Disorders, 24*(5), 659–685.

Magnússon, P., & Sæmundsen, E. (2001). Prevalence of Autism in Iceland. *Journal of Autism and Developmental Disorders, 31*(2), 153–163. https://doi.org/10.1023/A:1010795014548

Marson, D. C., Dymek, M. P., Duke, L. W., & Harrell, L. E. (1997). Subscale validity of the Mattis Dementia Rating Scale. *Archives of Clinical Neuropsychology, 12*(3), 269–275. https://doi.org/10.1016/S0887-6177(96)00003-0

Matson, J. L., & Bamburg, J. W. (1998). Reliability of the assessment of dual diagnosis (ADD). *Research in Developmental Disabilities, 19*(1), 89–95. https://doi.org/10.1016/S0891-4222(97)00031-0

Matson, J. L., Belva, B. C., Hattier, M. A., & Matson, M. L. (2012). Scaling methods to measure psychopathology in persons with intellectual disabilities. *Research in Developmental Disabilities, 33*, 549–562. https://doi.org/10.1016/j.ridd.2011.10.023

Matson, J. L., Boisjoli, J. A., González, M. L., Smith, K. R., & Wilkins, J. (2007). Norms and cut off scores for the autism spectrum disorders diagnosis for adults (ASD-DA) with intellectual disability. *Research in Autism Spectrum Disorders, 1*(4), 330–338. https://doi.org/10.1016/j.rasd.2007.01.001

Matson, J. L., Gardner, W., Coe, D. A., & Sovner, R. (1991). A scale for evaluating emotional disorders in severely and profoundly mentally retarded persons-Development of the diagnostic assessment for the severely handicapped (DASH) scale. *British Journal of Psychiatry, 159*, 404–409. https://doi.org/10.1192/bjp.159.3.404

Matson, J. L., Kazdin, A. E., & Senatore, V. (1984). Psychometric properties of the psychopathology instrument for mentally retarded adults. *Applied Research in Mental Retardation, 5*(1), 81–89. https://doi.org/10.1016/S0270-3092(84)80021-1

Matson, J. L., Nebel-Schwalm, M., & Matson, M. L. (2007). A review of methodological issues in the differential diagnosis of autism spectrum disorders in children. *Research in Autism Spectrum Disorders, 1*(1), 38–54. https://doi.org/10.1016/j.rasd.2006.07.004

Matson, J. L., Rush, K. S., Hamilton, M., Anderson, S. J., Bamburg, J. W., Baglio, C. S., … Kirkpatrick-Sanchez, S. (1996). Characteristics of depression as assessed by the diagnostic assessment for the severely handicapped-II (DASH-II). *Research in Developmental Disabilities, 20*(4), 305–313. https://doi.org/10.1016/S0891-4222(99)00012-8

Matson, J. L., & Shoemaker, M. (2009). Intellectual disability and its relationship to autism spectrum disorders. *Research in Developmental Disabilities, 30*(6), 1107–1114. https://doi.org/10.1016/j.ridd.2009.06.003

Matson, J. L., & Shoemaker, M. E. (2011). Psychopathology and intellectual disability. [Review]. *Current Opinion in Psychiatry, 24*(5), 367–371. https://doi.org/10.1097/YCO.0b013e3283422424

Matson, J. L., Smiroldo, B. B., Hamilton, M., & Baglio, C. S. (1997). Do anxiety disorders exist in persons with severe and profound mental retardation? *Research in Developmental Disabilities, 18*(1), 39–44. https://doi.org/10.1016/S0891-4222(96)00036-4

Matson, J. L., Wilkins, J., Boisjoli, J. A., & Smith, K. R. (2008). The validity of the autism spectrum disorders-diagnosis for intellectually disabled adults (ASD-DA). *Research in Developmental Disabilities, 29*, 537–546. https://doi.org/10.1016/j.ridd.2007.09.006

Mattis, S., Bellak, L., & Karasu, T. B. (1976). *Mental status examination for organic mental syndrome in the elderly patient. Geriatric Psychiatry*. doi: 3T Library-Converted #54; DLB-Converted #99; MCI-Converted #104; VBMprediction-Converted #71; Used to be #72

McDaniel, W. F., & McLaughlin, T. (2000). Further support for using the Dementia Rating Scale in the assessment of neuro-cognitive functions of individuals with mental retardation. *The Clinical Neuropsychologist, 14*(1), 72–75.

Mindham, J., & Espie, C. A. (2003). Glasgow Anxiety Scale for people with an Intellectual Disability (GAS-ID): Development and psychometric properties of a new measure for use with people with mild intellectual disability. *Journal of Intellectual Disability Research, 47*(1), 22–30. https://doi.org/10.1192/bjp.02.286

Moss, S., Patel, P., Prosser, H., Goldberg, D., Simpson, N., Rowe, S., & Lucchino, R. (1993). Psychiatric morbidity in older people with moderate and severe learning disability. I: Development and reliability of the patient interview (PAS-ADD). *British Journal of Psychiatry, 163*, 471–480. https://doi.org/10.1192/bjp.163.4.471

Moss, S., Prosser, H., & Goldberg, D. (1996). Validity of the schizophrenia diagnosis of the psychiatric assessment schedule for adults with developmental disability (PAS-ADD). *British Journal of Psychiatry, 168*, 359–367. https://doi.org/10.1192/bjp.168.3.359

Myrbakk, E., & von Tetzchner, S. (2008). Screening individuals with intellectual disability for psychiatric disorders: Comparison of four measures. *American Journal of Mental Retardation : AJMR, 113*(1), 54–70. https://doi.org/10.1352/0895-8017(2008)113[54:SIWIDF]2.0.CO;2

Nihira, K., Foster, R., Hellhaas, M., & Leland, H. (1975). AAMD adaptive behavior scale, 63:58–60.

Patja, K., Iivanainen, M., Vesala, H., Oksanen, H., & Ruoppila, I. (2000). Life expectancy of people with intellectual disability: A 35-year follow-up study. *Journal of Intellectual Disability Research : JIDR, 44*(Pt 5), 591–599.

Perry, A., & Factor, D. C. (1989). Psychometric validity and clinical usefulness of the Vineland Adaptive Behavior Scales and the AAMD Adaptive Behavior Scale for an autistic sample. *Journal of Autism and Developmental Disorders, 19*(1), 41–55. https://doi.org/10.1007/BF02212717

Petry, K., Kuppens, S., Vos, P., & Maes, B. (2010). Psychometric evaluation of the Dutch version of the Mood, Interest and Pleasure Questionnaire (MIPQ). *Research in Developmental Disabilities, 31*(6), 1652–1658. https://doi.org/10.1016/j.ridd.2010.04.011

Planelles-Fernández, I., Gallardo-Pujol, D., Labad, A., Godall, D., Cristia, N., Gutiérrez-Zotes, A., & Matson, J. L. (2017). Spanish adaptation of the autism spectrum disorders - diagnosis for adults (ASD-DA) in adults with severe and profound intellectual disability. *Journal of Developmental and Physical Disabilities, 29*(4), 625–642. https://doi.org/10.1007/s10882-017-9541-5

Powell, R. (2003). Psychometric properties of the Beck Depression Inventory and the Zung Self Rating Depression Scale in adults with mental retardation. *Mental Retardation, 41*(2), 88–95. https://doi.org/10.1352/0047-6765(2003)041<0088:PPOTBD>2.0.CO;2

Prasher, V., Farooq, A., & Holder, R. (2004). The adaptive behaviour dementia questionnaire (ABDQ): Screening questionnaire for dementia in Alzheimer's disease in adults with Down syndrome. *Research in Developmental Disabilities, 25*(4), 385–397. https://doi.org/10.1016/j.ridd.2003.12.002

Prasher, V. P. (1997). Dementia questionnaire for persons with mental retardation (DMR): Modified criteria for adults with Down's syndrome. *Journal of Applied Research in Intellectual Disabilities, 10*(1), 54–60. https://doi.org/10.1111/j.1468-3148.1997.tb00006.x

Prasher, V. P., Adams, C., & Holder, R. (2003). Long term safety and efficacy of donepezil in the treatment of dementia in Alzheimer's disease in adults with Down syndrome: Open label study. *International Journal of Geriatric Psychiatry, 18*(6), 549–551. https://doi.org/10.1002/gps.859

Ramirez, S. Z., & Lukenbill, J. (2008). Psychometric properties of the zung self-rating anxiety scale for adults with intellectual disabilities (SAS-ID). *Journal of Developmental and Physical Disabilities, 20*(6), 573–580. https://doi.org/10.1007/s10882-008-9120-x

Reiss, S. (1987). *Reiss screen for maladaptive behavior*. Orlando Park, IL: IDS.

Reiss, S., Levitan, G. W., & Szyszko, J. (1982). Emotional disturbance and mental retardation: Diagnostic overshadowing. *American Journal of Mental Deficiency*. 86(6):567-574

Reiss, S., & Valenti-Hein, D. (1994). Development of a Psychopathology Rating Scale for Children With Mental Retardation. *Journal of Consulting and Clinical Psychology, 62*(1), 28–33. https://doi.org/10.1037/0022-006X.62.1.28

Rellini, E., Tortolani, D., Trillo, S., Carbone, S., & Montecchi, F. (2004). Childhood autism rating scale (CARS) and autism behavior checklist (ABC) correspondence and conflicts with DSM-IV criteria in diagnosis of autism. *Journal of Autism and Developmental Disorders, 34*(6), 703–708. https://doi.org/10.1007/s10803-004-5290-2

Rojahn, J., & Helsel, W. J. (1991). The aberrant behavior checklist with children and adolescents with dual diagnosis. *Journal of Autism and Developmental Disorders, 21*(1), 17–28. https://doi.org/10.1007/BF02206994

Ross, E., & Oliver, C. (2003). Preliminary analysis of the psychometric properties of the mood, interest & pleasure questionnaire (MIPQ) for adults with severe and profound learning disabilities. *The British Journal of Clinical Psychology/the British Psychological Society, 42*(Pt 1), 81–93. https://doi.org/10.1348/014466503762842039

Roth, M., Tym, E., & Mountjoy, C. Q. (1986). CAMDEX. A standardised instrument for the diagnosis of mental disorder in the elderly with special reference to the early detection of dementia. *British Journal of Psychiatry, 149*, 698–709. https://doi.org/10.1192/bjp.149.6.698

Rutter Bailey, A., & Lord, C. M. (2003). *Social communication questionnaire*. Los Angeles, CA: Western Psychological Services.

Sappok, T., Budczies, J., Bölte, S., Dziobek, I., Dosen, A., & Diefenbacher, A. (2013). Emotional development in adults with autism and intellectual disabilities: A retrospective, clinical analysis. *PLoS One, 8*(9), e74036. https://doi.org/10.1371/journal.pone.0074036

Sappok, T., Diefenbacher, A., Budczies, J., Schade, C., Grubich, C., Bergmann, T., … Dziobek, I. (2013). Diagnosing autism in a clinical sample of adults with intellectual disabilities: How useful are the ADOS and the ADI-R? *Research in Developmental Disabilities, 34*(5), 1642–1655. https://doi.org/10.1016/j.ridd.2013.01.028

Sappok, T., Gaul, I., Bergmann, T., Dziobek, I., Bölte, S., Diefenbacher, A., & Heinrich, M. (2014). The Diagnostic Behavioral Assessment for autism spectrum disorder - Revised: A screening instrument for adults with intellectual disability suspected of autism spectrum disorders. *Research in Autism Spectrum Disorders, 8*(4), 362–375. https://doi.org/10.1016/j.rasd.2013.12.016

Sappok, T., Gaul, I., Dziobek, I., Bolte, S., Diefenbacher, A., & Bergmann, T. (2015). The Diagnostic Behavioural Assessment for Autism Spectrum Disorders (DiBAS): A screening instrument for adults with intellectual disability suspected of autism spectrum disorders. *Der Diagnostische Beobachtungsbogen Fur Autismus Spektrumstorungen (DiBAS): Ein Screeninginstrument Fur Erwachsene Mit Intelligenzminderung Bei Autismusverdacht.* https://doi.org/10.1055/s-0033-1359970

Sappok, T., Heinrich, M., & Diefenbacher, A. (2014). Psychometrische Eigenschaften der Autismus-Checkliste (ACL) für erwachsene Menschen mit Intelligenzminderung. *Psychiatrische Praxis, 41*(1), 37–44. https://doi.org/10.1055/s-0033-1343193

Sappok, T., Heinrich, M., & Underwood, L. (2015). Screening tools for autism spectrum disorders. *Advances in Autism, 1*(1), 12–29. https://doi.org/10.1108/AIA-03-2015-0001

Sarimski, K. (2004). Assessment of behavioral problems in children with intellectual disability: The Nisonger Child Behavior Rating Form. *Praxis der Kinderpsychologie und Kinderpsychiatrie, 53*(5), 319–332.

Schopler, E., Reichler, R. J., DeVellis, R. F., & Daly, K. (1980). Toward objective classification of childhood autism: Childhood Autism Rating Scale (CARS). *Journal of Autism and Developmental Disorders, 10*(1), 91–103. https://doi.org/10.1007/BF02408436

Sevin, J. A., Matson, J. L., Williams, D., & Kirkpatrick Sanchez, S. (1995). Reliability of emotional problems with the diagnostic assessment for the severely handicapped (DASH). *British Journal of Clinical Psychology, 34*(1), 93–94. https://doi.org/10.1111/j.2044-8260.1995.tb01442.x

Tenneij, N. H., & Koot, H. M. (2007). A preliminary investigation into the utility of the adult behavior checklist in the assessment of psychopathology in people with low IQ. *Journal of Applied Research in Intellectual Disabilities, 20*(5), 391–400. https://doi.org/10.1111/j.1468-3148.2007.00383.x

Vandevelde, S., Morisse, F., Došen, A., Poppe, L., Jonckheere, B., Hove, G. V., … Claes, C. (2016). The scale for emotional development-revised (SED-R) for persons with intellectual disabilities and mental health problems: Development, description, and reliability. *International Journal of Developmental Disabilities, 62*(1), 11–23. https://doi.org/10.1179/2047387714Y.0000000062

Volkmar, F. R., Klin, A., Siegel, B., Szatmari, P., Lord, C., Campbell, M., et al. (1994). Field trial for autistic disorder in DSM-FV. *American Journal of Psychiatry, 151*(9), 1361–1367. Retrieved from http://www.scopus.com/inward/record.url?eid=2-s2.0-0027998520&partnerID=tZOtx3y1

Wechsler, D. (1955). *Manual for the Wechsler Adult Intelligence Scale. New York Psychological Corporation*. doi: Thesis_references-Converted #317

Wieland, J., Wardenaar, K. J., Fontein, E., & Zitman, F. G. (2012). Utility of the Brief Symptom Inventory (BSI) in psychiatric outpatients with intellectual disabilities. *Journal of Intellectual Disability Research, 56*(9), 843–853. https://doi.org/10.1111/j.1365-2788.2011.01440.x

Wilkins, J., & Matson, J. L. (2009). A comparison of social skills profiles in intellectually disabled adults with and without ASD. *Behavior Modification, 33*(2), 143–155. https://doi.org/10.1177/0145445508321880

World Health Organization. (1992). *International Statistical Classification of Diseases and Related Health Problems. 10th Revision. Occupational Health* (Vol. 41). Retrieved from http://www.who.int/classifications/icd/ICD-10_2nd_ed_volume2.pdf

Zung, W. W. (1965). A self rating depression scale. *Archives of General Psychiatry, 12*, 63–70.

Zung, W. W. K. (1971). A rating instrument for anxiety disorders. *Psychosomatics, 12*(6), 371–379. https://doi.org/10.1016/S0033-3182(71)71479-0

24 Assessing Challenging Behaviors

Renee O. Hawkins, Tai A. Collins,
and J. Meredith Murphy

Children and adolescents diagnosed with intellectual disabilities (ID) display challenging behavior at about three times higher the rate than typically developing peers (Baker & Blacher, 2015). Unfortunately, without appropriate intervention, these problem behaviors can persist though adulthood (Carr et al., 2002; Lloyd & Kennedy, 2014). Common challenging behaviors include noncompliance, aggression (e.g., hitting, scratching, biting, verbal aggression), property destruction, and self-injurious behavior (Emerson et al., 2001; Wadsworth, Hansen, & Wills, 2015). In addition, deficits in social skills, adaptive functioning, and communication can further exacerbate challenging behaviors in those with ID (Matson et al., 2011).

Individuals with ID are among the most disadvantaged and socially excluded in society (Kozma, Mansell, & Beadle-Brown, 2009). Individuals with ID who also emit challenging behaviors are more likely than others to live in out-of-home care, be excluded from community services, placed in secure living facilities, be subjected to abuse and restrictive practices, and experience poor outcomes from community services (Beadle-Brown, Murphy, & DiTerlizzi, 2009; Beadle-Brown, Murphy, & Wing, 2006; Emerson, Malam, Davies, & Spencer, 2005; Felce, Lowe, Beecham, & Hallam, 2000; Kozma et al., 2009; Myrbakk & von Tetzchner, 2008). In the school setting, students with ID may have a more difficult time assimilating to inclusive classrooms due to their cognitive limitations and challenging behaviors that occur throughout the learning process (Smith, Polloway, Patton, & Dowdy, 2001). Students with ID may also exhibit a range of challenging behaviors that cause disruptions in the classroom and interrupt the learning environment for themselves and peers (Bilias-Lolis, Chafouleas, Kehle, & Bray, 2012). There is a clear need for early and effective interventions to positively change behavior (Delgado, Gonzalez-Gordon, Aragon, & Navarro, 2017). To inform the development, implementation, and evaluation of intervention supports, reliable and valid assessment methods must be used to understand the challenging behaviors displayed by individuals with ID.

Relevant professionals must engage in evidence-based practice by appropriately assessing the behaviors of concern at an individual level. Information about the challenging behaviors of concerns, including when do they occur, how often, at what intensity, under what conditions, and why, should be collected through the assessment process in order to establish an

---

R. O. Hawkins (✉)
School Psychology Program, sCECH | School of Human Services, University of Cincinnati, Cincinnati, OH, USA
e-mail: renee.hawkins@uc.edu

T. A. Collins · J. M. Murphy
School of Human Sciences, University of Cincinnati, Cincinnati, OH, USA

J. L. Matson (ed.), *Handbook of Intellectual Disabilities*, Autism and Child Psychopathology Series, https://doi.org/10.1007/978-3-030-20843-1_24

understanding of behavior. The assessment of maintaining variables for challenging behavior, with a goal of developing prosocial intervention procedures, is a prominent topic in the literature on ID (Matson & Wilkins, 2009). There is a strong evidence-base supporting the use of functional behavioral assessment (FBA) methods to understand the contextual variables supporting challenging behavior (Hanley, Iwata, & McCord, 2003). Results from FBAs identifying the environmental variables linked to problem behavior are used to develop effective intervention plans that are customized to meet individual needs (Matson & Wilkins, 2009). In addition, once interventions are implemented, ongoing data-based progress monitoring is essential to systematic problem-solving. Data are used to evaluate the effectiveness of intervention plans and make changes as needed (Pluymert, 2014).

This chapter presents an overview of a variety of methods that can be used to assess challenging behavior. The first section reviews general methods for assessing behavior, including interviews, systematic direct observation, rating scales, and direct behavior ratings. This section of the chapter also provides a description of can't do/won't do assessment methods. The second section of the chapter is focused on FBA methods. Descriptions of general methods and procedures are provided as well as information on published assessment tools.

General Assessment Methods

Given the prevalence of problem behaviors in individuals with ID, it is important that assessments methods accurately and consistently capture various dimensions of behaviors for the purposes of data-based decision-making (e.g., target behavior selection, screening, progress monitoring). A later section of this chapter will focus on function-based assessment, which includes a variety of methods assessing the variables maintaining problem behaviors; however, the current section will discuss general assessment procedures and their application to assessing challenging behaviors in individuals with ID. Behavioral assessment measures can be used to identify target behaviors, determine the contingencies acting on behaviors, monitor intervention effectiveness, as well as a host of other uses (Cooper, Heron, & Heward, 2007). According to Mayer, Sulzer-Azaroff, and Wallace (2014), strong measurement systems are (1) sensitive, in that they detect small changes in behavior; (2) objective, as they must measure observable behaviors with little inference or bias; (3) reliable, consistently measuring behaviors; and (4) valid, in that they measure what they are intended to measure. As such, a number of indirect and direct assessment methods may be used to measure target behaviors in individuals with ID, including interviews, direct observation, rating scales, direct behavior ratings, permanent products, and can't do/won't do assessment.

Interviews

Interviews are assessment methods in which stakeholders provide information with regard to target behaviors (e.g., severity, chronicity, topography, maintaining variables), as well as characteristics of the target individual and environment. Interviews are often the initial step in consultation to address problem behaviors with individuals with ID, and the data obtained via interviews are often verified via direct observation (Cooper et al., 2007). Interviews can be useful to obtain information from caregivers, parents, and others who interact with the target individual (Kelley, LaRue, Roane, & Gadaire, 2011), as well as the target individual themselves (Cooper et al., 2007).

Interviews are typically categorized as structured or semi-structured. Structured interviews include a series of questions asked in a specific order (e.g., the interview form of the Vineland Adaptive Behavior Scales—Third Edition; Vineland-3; Sparrow, Cicchetti, & Saulnier, 2016). Structured interviews may be useful to ensure that all parties interviewed are asked the same questions in the same order; however, they are often considered inflexible (Kelley et al., 2011). Semi-structured interviews (e.g., problem identification interview, typical day interview) allow for responsiveness to the interviewee's answers, as they include a framework of themes and potential questions that do not have to be asked in a specific order. When utilizing a semi-structured interview, follow-up questions may be asked to clarify any of the interviewee's responses (Kelley et al., 2011). Although helpful in the early stages of case conceptualization, interviews should not be relied on exclusively due to the indirect nature of the assessment and the reliance on stakeholders' recollections and perceptions (Cooper et al., 2007).

Direct Observation

Direct observation is often the preferred method of behavioral assessment for behavior analysts because data are taken at the time and place of the target behavior (Cooper et al., 2007). Rather than relying on reports from stakeholders, direct observation requires an observer to be present during the session to gather a direct account of the behavior and aspects of the environment. Two types of direct observation methods are typically used, including non-systematic/informal direct observation and systematic direct observation (SDO). Non-systematic or informal direct observation includes methods such as ABC recording (described later in the chapter) and narrative recording, wherein observers record behaviors, social interactions, setting characteristics, antecedents, and consequences of behaviors without predetermined definitions or coding schemes (Cooper et al., 2007). Non-systematic direct observation methods can be used to identify patterns in behavior and maintaining variables, as well as to develop operational definitions of target behaviors; however, qualitative data obtained with these methods may be difficult or impossible to quantify or compare (Thompson & Borrero, 2011). With SDO, observers utilize specific operational definitions and structured coding schemes to collect data, which leads to quantifiable and comparable data (Thompson & Borrero, 2011).

When collecting SDO data, commonly used methods include event recording, duration recording, and time sampling. With event recording, the number of occurrences of the target behavior is tallied. This is a relatively easy method of data collection in schools and other educational settings, as teachers can collect tally data while engaging in instruction (Cooper et al., 2007). Event recording methods are best for data with discrete beginning and ending points (Mayer et al., 2014). They are not appropriate for long-duration behaviors (e.g., on-task behavior) or behaviors that occur too frequently to be recorded accurately and reliably (e.g., rapid self-injurious behavior; Cooper et al., 2007). Event recording methods include count (i.e., number of occurrences of problem behavior during a session); rate/frequency (i.e., count per unit time); celebration (i.e., change in rate over time); trials to criterion (i.e., the number of opportunities necessary to meet a predetermined performance level); and percentage (i.e., ratio of responses to opportunities; Cooper et al., 2007; Mayer et al., 2014).

Duration recording requires observers to measure the amount of time behaviors occur or the time between behaviors and other events. When measuring duration, observers often utilize a stopwatch or other instrument to measure when behaviors start and stop, thereby calculating a total amount of time per behavior and within sessions (Cooper et al., 2007). Response latency is a measure of the amount of time between a behavior and some other stimulus or event, such as between a request or demand and the corresponding behavior. Inter-response time is the amount of time between two consecutive behaviors (e.g., smoking cigarettes). When using duration recording to measure duration, latency, and inter-response time, precision is required when measuring starting and ending points of behaviors relative to other behaviors and events (Cooper et al., 2007).

In time sampling methods, SDO sessions are divided into intervals and estimates of the overall duration or rate of behaviors are obtained (Cooper et al., 2007). The four types of time sampling methods include whole interval, partial interval, momentary time sampling, and planned activity check. With whole interval time sampling, an interval is coded if the target behavior occurs throughout the entire interval. Whole interval recording is best for long-duration behaviors, as well as behaviors that occur so frequently that they may be indistinguishable from each other (Cooper et al., 2007). Whole interval methods may underestimate the actual duration of a behavior because the behavior may occur during portions of intervals but not the entire interval (Thompson & Borrero, 2011). This underestimation becomes more problematic when longer intervals are used (Cooper et al., 2007). With partial interval time sampling, intervals are coded if the behavior occurs at any time during the interval, or for prespecified portions of the interval. Partial interval recording may overestimate the duration of behaviors because entire intervals are counted even if behaviors occur during a brief portion of the interval (Thompson & Borrero, 2011); however, partial interval time sampling may also underestimate the rate of frequently occurring behaviors because intervals are coded once no matter how many times they occur during the interval (Cooper et al., 2007). With momentary time sampling, the observer codes whether behaviors occur at the end of each interval. This may be a more feasible method of time sampling data collection, as observation is not required throughout intervals; however, this method is not appropriate for behaviors that occur infrequently or for short durations (Cooper et al., 2007). Momentary time sampling may over- or underestimate problem behavior, but results are more similar to the actual duration of problem behaviors when intervals are short (e.g., Powell, Martindale, Kulp, Martindale, & Bauman, 1977). Finally, planned activity check (PLACHECK; Cooper et al., 2007) is a measure of group behavior whereby the number of individuals in a group engaged in a target behavior at the end of each interval is recorded.

Baer, Wolf, and Risley (1968) described SDO, or “the direct observation and recording of a subject’s target behaviors by an observer under the stimulus control of a written behavior code” (p. 316) as the gold standard of behavioral assessment in applied behavior analysis, which has been repeatedly echoed throughout the SDO literature (e.g., Stichter & Riley-Tillman, 2014). Advantages of SDO include that it can be used to assess various dimensions of problem behaviors (Gresham, 2015), interobserver agreement can be calculated to assess for reliability of data collection, it requires little interference relative to other behavioral assessment methods (e.g., interviews, rating scales), and it has high treatment validity (i.e., results can be used to directly inform intervention efforts).

Although SDO is the preferred method of data collection in many fields, a number of disadvantages should also be considered. Reactivity, or the extent to which individuals behave differently when they are being observed, can affect the validity of SDO (Mayer et al., 2014); however, the effects of reactivity are often temporary and can be overcome by utilizing unobtrusive observers (Cooper et al., 2007; Gresham, 2015). SDO is often time-consuming and utilizes considerable resources, as it requires a trained observer to be present at the time and place of the behavior, but technology such as video recording may reduce the labor required for SDO (Gresham, 2015). Because SDO is a sampling of behavior at a particular time and place, it may not represent a general outcome measure of an individual’s behavior that is generalizable across time and settings (Gresham, 2015). Additionally, multiple observations may be necessary to ensure reliable and useful data (Chafouleas, 2011; Gresham, 2015). For example, Hintze and Matthews (2004) did not establish adequate, dependable SDO measurement when observing twice a day, indicating that four observations per day for a minimum of 4 weeks would be required. In contrast, Briesch, Chafouleas, and Riley-Tillman (2010) indicated that 3–5 observations were required to ensure a dependable estimation of academic engagement. As such, it is unlikely that one isolated SDO session is adequate when assessing target behaviors. Finally, observer biases such as the halo effect (i.e., data affected by the observer’s judgments of the person or situation) and expectancy effect (i.e., data influenced by the observer’s knowledge of the situation) may impact the validity and reliability of systematic direct observation (Cooper et al., 2007).

Behavior Rating Scales

Recent developments of psychometric sound instruments, such as behavior rating scales and checklists, can be used to identify variables in the environment that maintain challenging behaviors (Matson, Tureck, & Rieske, 2012). Behavior rating scales and checklists require stakeholders (e.g., target individual, parents, teachers, caregivers) to respond to descriptions of behaviors, often in a Likert format with a fixed choice of ratings (Kelley et al., 2011). Rating scales are particularly useful at the beginning of consultation to identify potential behaviors to be targeted by treatment and intervention efforts (Cooper et al., 2007), as they allow practitioners to better understand the type, frequency, and severity of challenging behaviors to inform treatment planning (Rojahn, Matson, Lott, Esbensen, & Smalls, 2001). When completing rating scales, raters are asked to respond based on their recollections of the target individual's behavior over some length of time (Campbell & Hammond, 2014). A number of broad-band rating scales can be useful to assess a variety of behaviors, including externalizing and internalizing problem behaviors, as well as school and family problems (e.g., Achenbach System of Empirically Based Assessment [ASEBA]; Achenbach & Rescorla, 2001), Behavior Assessment System for Children—Third Edition [BASC-3]; Reynolds & Kamphaus, 2015; Conners—Third Edition [Conners 3]; Conners, 2008). Additionally, narrow-band rating scales assess a more limited number of symptoms, but can be particularly helpful to pinpoint more prescribed dimensions of problem behaviors to target (e.g., Beck Depression Inventory [BDI-II]; Beck, Steer, & Brown, 1996; Revised Children's Manifest Anxiety Scale, second Edition [RCMAS-2]; Reynolds & Richmond, 2008; Social Skills Improvement System Rating Scales [SSIS]; Gresham & Elliott, 2008).

Rating scales have been developed specifically to assess problem behaviors in individuals with ID, including the Behavior Problems Inventory (BPI-01; Rojahn et al., 2001) and the Developmental Behavior Checklist (DBC; Einfeld & Tonge, 2002). The Behavior Problems Inventory is a 52-item rating scale assessing disruptive and aggressive behavior, stereotypic behavior, and self-injurious behavior in individuals with ID (Rojahn et al., 2001). The rating scale was developed for various functions, such as clinical assessment for individuals at risk for behavior problems, as a treatment outcome measure, and for administrative decision-making (Rojahn et al., 2001). Several studies have been completed to provide evidence for convergent, divergent, and concurrent validity of the measure (e.g., Gonzalez et al., 2008; Hill, Powlitch, & Furniss, 2008; Rojahn, Aman, Matson, & Mayville, 2003). Researchers have developed and established some psychometric properties of the Behavior Problems Inventory-Short Form (BPI-S), a more feasible 30-item measure (Mascitelli et al., 2015; Rojahn et al., 2012). The Developmental Behavior Checklist (DBC) is a suite of measures including a 93-item teacher form (DBC-T) and 96-item parent/caregiver form (DBC-P). These assessments, originally developed in Australia for the purposes of assessing problem behaviors in individuals with ID, include subscales for social behaviors, disruptive behaviors, anxiety, and communicative behaviors (Dekker, Nunn, Einfeld, Tonge, & Koot, 2002). As such, a variety of measures are available for practitioners treating individuals with ID, including assessments normed on general populations as well as measures specifically developed for individuals with ID.

Behavior rating scales are associated with a number of advantages that make them useful when assessing problem behaviors. Because many rating scales assess a broad range of behaviors or symptoms, they can yield a comprehensive assessment of the target individual's functioning (Gresham, 2015). They are also typically efficient, in that they often require fewer than 20 min to produce a large amount of data (Campbell & Hammond, 2014). Rating scales often have alternate forms that can be completed by multiple raters, allowing for cross-informant agreement to be analyzed. In addition, rating scales produce quantifiable data that can be compared to normative samples for overall populations, as well as populations of interest (Campbell & Hammond, 2014; Kelley et al., 2011). Quantitative information from rating scales can be more easily compared across settings, raters, and assessment locations than qualitative information gathered from interviews, and reliability and validity data can be calculated and interpreted with less bias (Kelley et al., 2011).

Although they are commonly used in many settings, some disadvantages are also associated with behavior rating scales. The most consequential disadvantage of behavior rating scales is the indirect nature of the assessment, as stakeholders' perceptions of individuals' typical rates of behavior, rather than actual direct rates of behavior, are obtained (Gresham, 2015). Additionally, although cross-informant agreement is considered a strength, it can also be a weakness of behavior rating scales when there is low agreement among raters (Gresham, 2015). Variance between raters can be problematic for interpretation and may be due to rater biases (e.g., biases to more severely or leniently rate behaviors, the central tendency effect, or halo effects and reputational biases) or setting variance (i.e., that expectations and demands vary between settings and may result in differences in stakeholders' perceptions of rates of problem behaviors; Campbell & Hammond, 2014). Omnibus rating scales may also lack sensitivity for small changes in problem behavior, which, along with administration times and number of items, may limit the ability of rating scales to be used for the purpose of progress monitoring. Each of these disadvantages should be considered when utilizing behavior rating scales to assess rates of behaviors in individuals with ID.

Direct Behavior Ratings

Direct Behavior Ratings (DBR) are evaluative judgments typically completed immediately after an observation period (Chafouleas, 2011). Common examples include teachers, school or clinic staff, parents, and the target individual providing a rating of behaviors after a session or classroom period (e.g., rating on a percentage scale or Likert scale the amount of time a target student was on task during a class). DBRs were designed as a hybrid technique to capitalize on the advantages of SDO and behavior rating scales (Chafouleas, 2011; Christ, Riley-Tillman, & Chafouleas, 2009). Specifically, like SDO, DBRs are typically completed at the time and place of the behavior, require little inference, and can be administered repeatedly for progress monitoring purposes, while, also capitalizing on the efficiency of behavior rating scales (Chafouleas, Riley-Tillman, & Christ, 2009). A strong literature base supports the technical adequacy of DBR, as moderate correlations were found between teacher-completed DBR and observers' SDO data (Chafouleas, McDougal, Riley-Tillman, Panahon, & Hilt, 2005; Riley-Tillman, Chafouleas, Sassu, Chanese, & Glazer, 2008).

Many advantages make DBRs appropriate for use in a variety of settings. First, because DBRs are brief, they are often considered a feasible and acceptable measure of behavior (Chafouleas, Riley-Tillman, & Sassu, 2006). As such, DBR-Single Item Scales (DBR-SIS; Chafouleas, 2011) have been developed with a single item to improve the efficiency of measurement. Also, DBRs may not need to be completed immediately after the observation session, as Dart et al. (2017) found in an analog study that latency of completing a DBR did not affect the accuracy of obtained data. Finally, DBRs are flexible, in that they can be personalized to the target individual, and the rater and frequency of rating occasions can be varied (Gresham, 2015). The personalization of DBRs may also be a disadvantage, in that the technical adequacy of the resulting instrument may be unverified.

Permanent Products

Permanent products are byproducts or outcomes produced by behaviors that last long enough to be measured and recorded (e.g., homework assignments, number of correct math problems; Cooper et al., 2007). Permanent products may also be contrived, such as with audio or video recordings of sessions. An advantage of permanent products is that observers are not required to be present during the session of interest, as some behaviors occur during times and in places that are not able to be observed (Cooper et al., 2007). Also, permanent products allow for the utilization of more complex coding schemes and measurement of multiple behaviors since they are analyzed after the observation session, and interobserver agreement may be calculated (Cooper et al., 2007). With regard to disadvantages, some behaviors may not produce long-lasting byproducts that can be collected as permanent products (e.g., on-task behavior), and, if the observation session is not recorded, important aspects of the environment may not be assessed and it may not be clear whether the target individual or some other person produced the permanent product (Cooper et al., 2007). As such, permanent products are best utilized when real-time data are not needed for decision-making, for behaviors that reliably produce measurable outcomes, and when measurement by permanent product would be unobtrusive to the natural environment (Cooper et al., 2007).

Can’t Do/Won’t Do Assessment

Can’t do/won’t do assessment refers to a variety of procedures used to determine whether problem behaviors are acquisition or performance deficits. Acquisition deficits are behaviors that are not in the individual’s repertoire (i.e., the individual cannot currently do them), while performance deficits are in the individual’s repertoire, but they are not performed adequately or in the appropriate situations (i.e., the individual will not do them; Gresham & Elliott, 2008). With regard to academic problems, can’t do/won’t do assessment is typically conducted by offering the individual a preferred reward for exceeding a previously obtained score and determining whether the next score is altered by the presentation of a reinforcer (e.g., VanDerHeyden, 2014). In the realm of social skills, the SSIS-RS (Gresham & Elliott, 2008) is a behavior rating scale that indicates whether problem behaviors are performance or acquisition deficits. Regardless of the methods, can’t do/won’t do assessments are useful in treatment planning, as direct instruction with performance feedback is appropriate for acquisition deficits, while performance deficits require behavioral interventions including differential reinforcement, prompting, and feedback (Gresham & Elliott, 2008).

Functional Behavioral Assessment

Functional behavioral assessment (FBA) refers broadly to methods for identifying environmental factors contributing to challenging behavior (Cooper et al., 2007; Steege & Watson, 2009). FBA relies on the assessment techniques already described in this chapter, including interviews, systematic direct observation, and rating scales, to describe and analyze problem behavior in the context in which it occurs. The goal of FBA is to determine the antecedents and consequences that trigger and maintain challenging behavior. Antecedents are events, activities, and stimuli that occur before problem behavior presents whereas consequences are events, activities, and stimuli that occur after the challenging behavior. Antecedents may include discriminative stimuli, which are stimuli that signal that reinforcement is available for a particular response. For example, a box of cereal on the kitchen table may serve as a discriminative stimulus for asking to eat breakfast. As another antecedent event, motivating operations change how valuable a reinforcer is to an individual. For example, the day after Halloween, a child might not be as motivated by a candy as a reinforcer as they were previously, when candy was not as readily available. Consequences include positive reinforcement (social attention, activities, tangibles), negative reinforcement (escape or avoidance of social attention, settings, task demands, or activities), and automatic reinforcement (a change in sensory stimulation). Antecedents and consequences each affect the likelihood that a problem behavior will occur. By identifying functional relationships between environmental variables (i.e., antecedents and consequences) and problem behavior, FBA aims to determine why a problem behavior is occurring (i.e., the function of the behavior). This information about function can then be used to develop intervention plans that manipulate the environment to effectively decrease challenging behavior and promote positive behavior (Cooper et al., 2007; Steege & Watson, 2009).

There is a strong evidence base supporting the use of FBA. Over the last four decades, researchers have repeatedly demonstrated that FBA is a valid approach for assessing problem behaviors and there are several books dedicated to the topic (e.g., Cipani, 2018; Crone, Hawken, & Horner, 2015; Steege & Watson, 2009). Research has been conducted using FBA to examine a variety of challenging behaviors including aggression, noncompliance, property destruction, stereotypy, off-task, self-injury, tantrums, elopement, inappropriate vocalizations (Anderson, Rodriguez, & Campbell, 2015; Hanley et al., 2003); across clinical, school, and home settings (Hanley et al., 2003); for typically developing children and adults (Hanley et al., 2003); and for children and adults with disabilities, including ID (Wadsworth et al., 2015), autism spectrum disorder (Larkin, Hawkins, & Collins, 2016), emotional and behavior disorders (Gage, Lewis, & Stichter, 2012), and attention-deficit/hyperactivity disorder (Miller & Lee, 2013). This extensive literature base has highlighted continuously the importance of identifying the function of problem behavior for effective intervention development (Didden, Korzilius, Van Oorsouw, & Sturmey, 2006; Harvey, Boer, Meyer, & Evans, 2009). In fact, researchers have noted that interventions based on the results of FBA result in better outcomes than interventions that are developed without information regarding function (Ingram, Lewis-Palmer, & Sugai, 2005; Newcomer & Lewis, 2004; Payne, Scott, & Conroy, 2007).

Three approaches to FBA are described in the literature, including (1) indirect FBA, (2) direct descriptive FBA, and (3) functional analysis (Cooper et al., 2007; Steege & Watson, 2009). All three fall under the umbrella term FBA, yet they differ in the intensity of the assessment procedures involved. Based on the strengths and limitations of the assessment methods used within each type of FBA, indirect and direct descriptive FBA lead to clear descriptions of challenging behavior and hypothesized functions of the behavior while functional analysis is used to confirm functional hypotheses by systematically manipulating the environment. To conserve often-limited resources in providing assessment and intervention services to individuals with ID, problem-solving teams should try to match the intensity of the FBA approach selected with the intensity of the challenging behavior.

Indirect FBA

Indirect FBA relies on indirect data sources including interviews, rating scales, and record reviews to generate functional hypotheses (Iwata & Worsdell, 2005). Given the often-questionable reliability and validity of indirect assessments, relying solely on indirect FBA methods is not recommended but rather can be seen as a first step in developing a clear understanding of the challenging behavior, its context, and possible functions. Further, when a problem behavior is relatively minor (e.g., occurs infrequently, does not cause harm to the individual or others), problem-solving teams may decide that it is appropriate to move forward with intervention planning based only on information collected through indirect FBA. In this context, problem-solving teams may determine that resources (i.e., time, personnel) may be better utilized addressing concerns related to more severe challenging behavior rather than expending valuable resources collecting additional information. A team may decide that data collected through an indirect FBA may provide enough to develop an intervention plan with a high likelihood of success.

FBA interviews. To guide FBA interviews and help address concerns with the technical adequacy of clinical interviews in general, several structured FBA interview forms have been developed. Generally, FBA interview forms provide prompts to gather information about the individual displaying the challenging behavior (e.g., demographic information, strengths, likes/dislikes, social skills, communication strategies), descriptions of the problem behaviors, contextual variables that affect the behavior (e.g., routines, sleep patterns, medications, eating habits, triggering events, consequences), and previous intervention strategies that have been implemented. Prompts are also included to generate hypotheses regarding the function of the problem behavior. The major goals of the FBA interview are to operationally define the challenging behavior in observable and measureable terms; to identify the antecedents and consequences that are associated with the problem behavior; to generate a hypothesized function for the behavior; and to identify and clearly define appropriate, desired, alternative behaviors (Gresham, Waston, & Skinner, 2001). Several interview forms have been designed to support the accomplishment of these goals and most are readily available to practitioners. Table 24.1 lists the indirect FBA tools reviewed in this chapter and how to access them.

The Functional Assessment Checklist: Teachers and Staff (FACTS) is a brief semi-structured interview form designed with school-based problem-solving teams in mind (March et al., 2000). A review of research on the FACTS supported its reliability (0.77 test-re-test reliability; 0.50–0.88 inter-rater reliability; 1.00 interobserver agreement) and validity, with 80% of students receiving interventions developed based on information collected with the FACTS showing at least a 50% decrease in the presentation of challenging behavior (McIntosh et al., 2008). Similar to the FACTS, the slightly longer Functional Assessment Interview (FAI) is a semi-structured interview designed for use in school and clinical settings to assess the behaviors of individuals with and without developmental disabilities (O'Neil et al., 1997). Research on the FAI has found high inter-rater reliability (86–88%) for the tool as well as evidence of convergent validity with other FBA methods (e.g., rating scales, direct observations) and treatment utility (Dufrene, Kazmerski, & Labrot, 2016). The Functional Assessment Informant Record-Teacher (FAIR-T) is another semi-structured interview form to collect information on the context of problem behavior (Edwards, 2002). Results from the FAIR-T also have shown convergence with other FBA methods and led to the development of effective intervention plans (Dufrene et al., 2016).

FBA rating scales. Rating scales specifically designed to consider the function of problem behaviors also have been developed to strengthen

Table 24.1 Indirect functional behavioral assessment tools

Tool	Format	Availability
Functional assessment checklist for teachers and staff (FACTS)	Semi-structured interview	Positive Behavioral Interventions and Supports: OSEP Technical Assistance Center https://www.pbis.org/resource/246/functional-assessment-checklist-for-teachers-and-staff-facts
Functional assessment interview (FAI)	Semi-structured interview	The Center on the Social and Emotional Foundations for Early Learning (Vanderbilt University) http://csefel.vanderbilt.edu/modules/module3a/handout5.pdf
Functional assessment informant record for teachers (FAIR-T)	Semi-structured interview	Included in full in: Steege, M.W. & Watson, T.S. (2009). *Conducting school-based functional behavioral assessment (2nd Ed.).* New York: The Guilford Press
Questions about behavior function (QABF)	Rating scale	Available for order from Disability Consultants, LLC http://www.disabilityconsultants.org/Order.php
Functional analysis screening tool (FAST)	Questionnaire	Included in full as Appendix to: *Journal of Applied Behavior Analysis,* Volume 46, Spring 2013
Motivation assessment scale (MAS)	Rating scale	Available for order from Monaco & Associates https://www.monacoassociates.com/products
Contextual assessment inventory for problem behavior (CAI)	Rating scale/ inventory	Included in full as appendix to: McAtee, M., Carr, E. G., & Schulte, C. (2004). A contextual assessment inventory for problem behavior: Initial development. *Journal of Positive Behavior Interventions, 6(3),* 148–165

the FBA process and results (Matson et al., 2012). The information obtained from these rating scales can serve as a foundation for follow-up interviews and observations and can also provide antecedent or consequent event information to be included in a functional analysis (Iwata, DeLeon, & Roscoe, 2013).

The Questions About Behavioral Function (QABF) is the most studied and psychometrically sound FBA rating scale used to achieve an understanding of challenging behavior presented by individuals with ID in a simpler way than experimental functional analysis (Matson et al., 2012; Matson & Vollmer, 1995). The QABF contains 25 items with five items in each of five factors: attention, escape, non-social, physical, and tangible (Matson et al., 2012). Items are scored on the following dimensions: occur, does not occur, and does not apply (Matson et al., 2012). The challenging behaviors are also rated on severity (e.g., rarely, some, and often). The scale can be completed in 20 min (Matson et al., 2012), saving professionals valuable time. Researchers have repeatedly established the reliability and validity of the QABF and documented convergence with functional analysis results (Matson et al., 2012).

The Functional Analysis Screening Tool (FAST) is a 16-item questionnaire about antecedent and consequent events that may be correlated with the occurrence of problem behavior (Iwata et al., 2013; Iwata & DeLeon, 1996). The FAST was developed to prompt dialogue about conditions under which problem behavior might occur and to organize the reports around positive and negative reinforcement (Iwata et al., 2013). The contingencies are then divided based on whether the source of reinforcement is social (delivered by others) or automatic (produced directly by the response), which yields four functional categories: (1) social-positive reinforcement (access to attention or tangible items), (2) social-negative reinforcement (escape from task demands or other types of social interaction), (3) automatic-positive reinforcement (self-stimulatory behavior), and (4) automatic-negative reinforcement (alleviation of pain or discomfort; Iwata et al., 2013). Assessment of the FAST's reliability and validity are considered adequate, in that they compare well with other rating scales, but it lacks appropriateness for the purpose of treatment development (Iwata et al., 2013).

The Motivation Assessment Scale (MAS) produces scores for an indicated challenging behavior related to four potential functions: sensory, escape, attention, and tangible (Durand & Crimmins, 1992). The MAS is frequently used to assess learned function of challenging behavior in people with ID (Koritsas & Iacono, 2013). The MAS consists of 16 questions related to the challenging behavior that are rated on a 6-point Likert scale (0 = never, 1 = almost never, 2 = seldom, 3 = half the time, 4 = usually, 5 = almost always, 6 = always), (Fee, Schieber, Noble, & Valdovinos, 2016). The function is then determined by the subscale with the highest average score. The MAS has been tested with functional analysis results in the past and was found to have statistically significant correlations with functional analysis results (Herzinger & Campbell, 2007); however, other studies have found mixed results regarding MAS convergence with other FBA approaches, including functional analysis (Arnforfer, Miltenberger, Woster, Rortvedt, & Gaffaney, 1994; Crawford, Brockel, Schauss, & Miltenberger, 1992; Paclawskyj, Matson, Rush, Smalls, & Vollmer, 2000).

The Contextual Assessment Inventory for Problem Behavior (CAI) is a rating scale/inventory to identify environmental variables contributing to challenging behavior in individuals with developmental disabilities (McAtee, Carr, & Schulte, 2004). The CAI includes 80 rating scale items and 13 open-ended questions. For rating scale items, raters are asked to indicate the likelihood that the individual will display the problem behavior based on the information provided based on 5-point scale (1 = never, 3 = half the time, 5 = always). Research studies have indicated moderate to strong internal consistently and temporal stability (Dufrene et al., 2016). In addition, researchers found evidence supporting the convergent and predictive validity of the instrument (Carr, Ladd, & Shulte, 2008) and for the effectiveness of interventions based on the results (Moskowitz, Carr, & Durand, 2011).

Direct Descriptive FBA

Direct descriptive FBA typically incorporates the indirect measures used in indirect FBA but adds and heavily relies on the collection of direct observation data, including the use of the SDO methods described previously. Direct observation is used to clearly define and record the occurrence of not only the challenging behavior but also the co-occurring environmental events, providing context for the problem behavior.

In the simplest form, direct observations may include narrative recordings of the antecedents, challenging behavior, and consequences as they occur during the observation session (i.e., A-B-C recording; O'Neil et al., 1997). A-B-C recoding can be modified to include additional relevant information such as the setting, time of day, or delayed consequences. Scatterplot assessments track when the challenging behavior occurs throughout the day for several days, while also noting the co-occurring tasks and activities. Data are visually analyzed to determine correlates of problem behavior and generate hypotheses regarding function (Touchette, MacDonald, & Langer, 1985). Through descriptive assessments, data are systematically collected to quantify not only the occurrence of the problem behavior, but also the occurrence of antecedent and consequences. Based on these data, conditional probabilities can be calculated (e.g., the student avoids task demands following 90% of occurrences of challenging behavior; conditional probability of 0.90) and used to develop functional hypotheses (Dufrene, Doggett, Henington, & Watson, 2007).

While indirect and direct descriptive FBA methods are supported in the research for developing effective, function-based interventions, they are not without limitations. These FBA approaches result in hypotheses regarding the variables supporting challenging behavior but do not actually confirm such functional relationships. Functional analysis procedures, however, are designed to do just that. By systematically manipulating the environment, functional analysis procedures can verify functional relationships and increase the likelihood that subsequent intervention plans based on the results will be effective. Researchers have noted that the results of indirect FBA methods, in particular, do not consistent align with the results of functional analysis, highlighting the importance of experimentally validating functional hypotheses for intervention planning (Dufrene et al., 2016; Fee et al., 2016)

Functional Analysis

Iwata, Dorsey, Slifer, Bauman, and Richman (1982/1994) were the first to describe functional analysis procedures. They examined the effects of four conditions (three test conditions and one control condition) on the self-injurious behavior (SIB) of nine children with developmental disabilities. Each test condition provided a different potential reinforcer contingent on display of the SIB. Conditions were designed to determine the degree to which each of the following potential reinforcers was functionally related to SIB: attention, escape/avoid task demands, and automatic sensory reinforcement. In the social disapproval condition, which tested for attention as the maintaining consequence, the participant was instructed to play with some toys while the experimenter engaged in another task. The experimenter then provided attention in the form of verbal redirections, and statements of disapproval and concern, accompanied by a brief, neutral physical contact contingent upon the display of challenging behavior. In the academic demand condition, which tested for escape/avoidance as the maintaining reinforcer, participants were instructed to engage in academic tasks that had a low probability of occurrence (i.e., the student typically would not complete them). Upon the display of SIB, the experimenter took away the task and turned away from the participant. In the alone condition, which tested for automatic sensory reinforcement, the participant was placed in a room without access to any external stimulation (i.e., toys, other people). In the unstructured play condition, which served as the control condition, no task demands were placed on participants and they had access to toys and social attention contingent upon appropriate behavior. Conditions were administered repeatedly, with session durations of up to 30 min and the total number of sessions ranging from 24–53 across participants. Although there was variability in the occurrence of SIB across and within individuals, for 6 of the participants SIB consistently occurred at higher rates in one test condition as compared to the other conditions. Since this original study, researchers have repeatedly used these and similar procedures to empirically determine functional relationships between the environment and challenging behavior (Hanley et al., 2003).

While functional analysis is still considered the "gold standard" in FBA methods, concerns have been raised regarding the feasibility and validity of procedures. First, the traditional or extended functional analysis procedures described by Iwata et al. (1982/1994) require an extensive time commitment. On average, a traditional functional analysis requires six and a half hours to complete (Iwata et al., 1994; Tincani, Castrogiavanni, & Axelrod, 1999). Each condition is administered repeatedly, often requiring several days to carry out the procedures. In addition, clinicians need significant training and expertise to ensure the procedures are designed and implemented consistent with the research supporting functional analysis (Iwata et al., 2000). Finally, traditional functional analysis has been criticized for the contrived nature of the experimental conditions, with researchers and clinicians that the conditions often do not resemble real-life scenarios (Carr, Yarbrough, & Langdon, 1997). These issues have led researchers to explore modified functional analysis procedures, including brief functional analysis, trial-based functional analysis, and Interview-Informed Synthesized Contingency Analysis.

Brief functional analysis involves the same procedures and conditions typically included in extended functional analysis but the number of sessions is fewer and each session is briefer (Northup et al., 1991). More specifically, while extended functional analysis procedures may involve 30 or more sessions, with session durations of up to 30 min, brief functional analysis procedures typically involve 8–10 sessions of 10–15 min each (Cihak, Alberto, & Fredrick, 2007; Northup et al., 1991; Steege & Watson, 2009). Research indicates that the average amount of time to conduct a brief functional analysis is 90 min (Asmus et al., 2004). Cihak et al. (2007) used brief functional analysis procedures to identify the environmental variables maintaining the challenging behaviors displayed by four high-school students with ID. Initially, three conditions (escape from task demands, attention, and control) were each implemented once, with each session lasting 10 min. To confirm the results, the condition which resulted in the highest levels of problem behavior was then implemented again, followed by the condition resulting in the second highest levels of problem behavior, and finally reimplementation of the condition with the highest levels of problem behavior in the final functional analysis session. Based on the results, the researchers identified a function for the challenging behavior displayed by each participant and developed an effective function-based intervention.

Trial-based functional analysis is another alternative to traditional functional that has emerged in the literature as an effective way to empirically determine contingencies maintaining challenging behavior (Larkin et al., 2016; Sigafoos & Saggers, 1995). With this method, trial-based assessments of different contingency conditions (attention, escape, tangible, and automatic sensory) are embedded throughout the individual's typical daily routine and activities. Trials last only up to 60s each, with 10–20 trials implemented per condition (Chezan, Drasgow, & Martin, 2014; Sigafoos & Saggers, 1995). Each trial consists of a control part, consisting of free access to reinforcers (attention, tangibles, break from demands), and a test part in which the reinforcement is delivered contingent upon the display of the challenging behavior (Kodak, Fisher, Paden, & Dickes, 2013). Data collection simply involves recording whether or not the challenging behavior occurs during each trial. To interpret the results and determine function, data are compared (1) across test and control parts within conditions and (2) across conditions. Chezan et al. (2014) carried out trial-based functional analysis procedures to determine the function of challenging behavior displayed by three adults with ID.

Trials were embedded in relevant activities. For example, trials for attention occurred during one-on-one time and trials for tangibles occurred during snack time. The results of the trial-based functional analysis clearly pointed to a function for each participant, which informed the implementation of successful function-based intervention plans.

Most recently, Interview-Informed Synthesized Contingency Analysis (IISCA) has been introduced as a modified approach to functional analysis that requires significantly less time (Jessel, Hanley, & Ghaemmaghami, 2016). IISCA first relies on interviews to determine hypothesized functions of behavior. Based on the interview, the subsequent functional analysis includes only one test condition and one control condition. Similar to brief functional analysis, conditions are tested during a limited number of brief sessions. In the initial study of IISCA procedures, researcher reported that it required only 23 min to conduct the analysis and the function of behavior was confirmed for the three children with autism (Hanley, Jin, Vanselow, & Hanratty, 2014). In a follow-up study, researchers conducted 30 analyses using the IISCA procedures (Jessel et al., 2016). The average time to complete the analysis was 25 min, range, 15–75 min. Although more research is needed, IISCA show promise as an efficient option for conducting a functional analysis.

FBA in Law

Due to the strong research supporting its use, FBA is mandated for serving students with disabilities who display challenging behavior by the Individuals with Disabilities Improvement Act (2004). IDEA requires that an FBA be conducted when there is a change in a child's educational placement as a disciplinary measure in response to behavioral misconduct that is (a) a manifestation of the child's disability or (b) a result of the school's failure to implement the Individualized Education Program. IDEA also includes language suggesting that schools consider conducting a FBA when a child's behavior interferes with their learning and/or the learning of others. Further, approximately 30 states include reference to FBA methods in their special education laws (Collins & Zirkel, 2017). Given the strong research evidence for FBA methods for effectively serving individuals with challenging behavior and the inclusion of recommendations around the use of FBA in federal and state laws, it is critical that professionals serving individuals with disabilities gain knowledge and skills in FBA to implement best practice.

Conclusion

Unfortunately, there is a large body of research documenting the negative social, emotional, learning, and occupational outcomes for individuals with ID displaying challenging behavior (Coe et al., 1999; Lloyd & Kennedy, 2014). Evidence-based assessment methods can be used to help inform intervention planning and monitor intervention effects to continuously improve and modify plans to improve these outcomes. A variety of methods have been studies for the assessment of challenging behaviors. Indirect methods such as interviews and rating scales provide a starting point for describing problem behavior and understanding why it occurs. Direct methods, including systematic direct observation, can further this understanding by quantifying behavior in the context in which it naturally occurs. Functional behavioral assessment provides a comprehensive assessment framework for assessing challenging behavior, with varying levels of intensity of assessment procedures that can be tailored to match the intensity of the problem behavior and the resources of the professionals serving the individual demonstrating the behavior.

Indirect and direct descriptive FBA approaches offer methods to generate hypotheses about the environmental contingencies maintaining problem behaviors. Many assessment methods used in indirect and direct descriptive FBA require little time and are straightforward in their procedures and purpose. These approaches are an excellent "first line of defense" for understanding challenging behavior but they are limited in that they do not empirically validate hypothesized functions of behavior. To do so, a functional analysis should be conducted. Functional analysis procedures, however, require resources that are not always available in every setting serving individuals with ID. Conducting a functional analysis requires trained professionals to ensure that the analysis is carried out with integrity and a significant time commitment, which can vary based on the approach used. However, the time spent conducting a functional analysis may be time saved implementing ineffective intervention plans as research supports the increased effectiveness of function-based interventions (Ingram et al., 2005; Newcomer & Lewis, 2004; Payne et al., 2007).

School-, clinical-, and community-based problem-solving teams need to weigh the costs and benefits of different FBA approaches, given the context of their system and the severity of the challenging behavior. At a minimum, all challenging behavior should be considered in terms of function through indirect FBA methods. With increasing intensity of the challenging behavior, additional assessments should be conducted to increase the likelihood that intervention plans will efficiently improve behavior. When faced with challenging behavior that significantly interferes with an individual's daily functioning and/or when the results of indirect and direct descriptive FBA are unclear, functional analysis represents the most widely research and well supported approach for the assessment of challenging behavior.

References

Achenbach, T. M., & Rescorla, L. A. (2001). *Manual for the ASEBA school-age forms and profiles: An integrated system of multi-informant assessment*. Burlington, VT: Research Center for Youth and Families.

Anderson, C. M., Rodriguez, B. J., & Campbell, A. (2015). Functional behavior assessment in schools: Current status and future directions. *Journal of Behavioral Education, 24*, 338–371.

Arnforfer, R. E., Miltenberger, R. G., Woster, S. H., Rortvedt, A. K., & Gaffaney, T. (1994). Home-based descriptive and experimental analysis of problem behaviors in children. *Topics in Early Childhood and Special Education, 14*, 64–87.

Asmus, J. M., Ringdahl, J. E., Sellers, J. A., Call, N. A., Andelman, M. S., & Wacker, D. P. (2004). Use of a short-term impatient model to evaluate aberrant behavior outcome data summaries from 1996-2001. *Journal of Applied Behavior Analysis, 37*, 283–304.

Baer, D. M., Wolf, M. M., & Risley, T. R. (1968). Some current dimensions of applied behavior analysis. *Journal of Applied Behavior Analysis, 1*(1), 91–97.

Baker, B. L., & Blacher, J. (2015). Disruptive behavior disorders in adolescents with ASD: Comparisons to youth with intellectual disability or typical cognitive development. *Journal of Mental Health Research in Intellectual Disabilities, 8*, 98–116.

Beadle-Brown, J., Murphy, G., & DiTerlizzi, M. (2009). Quality of life for the Camberwell cohort. *Journal of Applied Research in Intellectual Disabilities, 22*, 380–390.

Beadle-Brown, J., Murphy, G., & Wing, L. (2006). The Camberwell cohort 25 years on: Characteristics and changes in skills over time. *Journal of Applied Research in Intellectual Disabilities, 19*, 317–329.

Beck, A. T., Steer, R. A., & Brown, G. K. (1996). *Manual for the Beck Depression Inventory–II*. San Antonio, TX: Psychological Corporation.

Bilias-Lolis, E., Chafouleas, S. M., Kehle, T. J., & Bray, M. A. (2012). Exploring the utility of self-monitoring in decreasing disruptive behavior in students with intellectual disability. *Psychology in the Schools, 49*(1), 82–92.

Briesch, A. M., Chafouleas, S. M., & Riley-Tillman, T. C. (2010). Generalizability and dependability of behavior assessment methods to estimate academic engagement: A comparison of systematic direct observation and direct behavior rating. *School Psychology Review, 39*, 408–421.

Campbell, J. M., & Hammond, R. K. (2014). Best practices in rating scale assessment of children's behavior. In P. L. Harrison & A. Thomas (Eds.), *Best practices in school psychology: Data-based and collaborative decision making* (pp. 287–304). Bethesda, MD: National Association of School Psychologists.

Carr, E., Yarbrough, S., & Langdon, N. (1997). Effects of idiosyncratic stimulus variables on functional analysis outcomes. *Journal of Applied Behavior Analysis, 30*(4), 673–686.

Carr, E. G., Dunlap, G., Horner, R. H., Koegel, R. L., Turnbull, A. P., Sailor, W., … Fox, L. (2002). Positive behavior support: Evolution of an applied science. *Journal of Positive Behavior Interventions, 4*(1), 4–16.

Carr, E. G., Ladd, M. V., & Shulte, C. F. (2008). Validation of the contextual assessment inventory for problem behavior. *Journal of Positive Behavior Interventions, 10*, 91–104.

Chafouleas, S. M. (2011). Direct behavior rating: A review of the issues and research in its development. *Education and Treatment of Children, 34*(4), 575–591.

Chafouleas, S. M., McDougal, J. L., Riley-Tillman, T. C., Panahon, C. J., & Hilt, A. M. (2005). What do daily behavior report cards (DBRCs) measure? An initial comparison of DBRCs with direct observation for off-task behavior. *Psychology in the Schools, 42*, 669–676.

Chafouleas, S. M., Riley-Tillman, T. C., & Christ, T. J. (2009). Direct behavior rating (DBR): An emerging method for assessing social behavior within a tiered intervention system. *Assessment for Effective Intervention, 34*(4), 195–200.

Chafouleas, S. M., Riley-Tillman, T. C., & Sassu, K. A. (2006). Acceptability and reported use of daily behavior report cards among teachers. *Journal of Positive Behavior Interventions, 8*(3), 174–182.

Chezan, L. C., Drasgow, E., & Martin, C. A. (2014). Discrete-trial functional analysis wand functional communication training with three adults with intellectual disabilities and problem behavior. *Journal of Behavioral Education, 23*, 221–246.

Christ, T. J., Riley-Tillman, T. C., & Chafouleas, S. M. (2009). Foundation for the development and use of direct behavior rating (DBR) to assess and evaluate student behavior. *Assessment for Effective Intervention, 34*(4), 201–213.

Cihak, D., Alberto, P. A., & Fredrick, L. D. (2007). Use of brief functional analysis and intervention evaluation in public settings. *Journal of Positive Behavior Interventions, 9*, 80–93.

Cipani, E. (2018). *Functional behavioral assessment, diagnosis, and treatment* (3rd ed.). New York: Springer.

Coe, D. A., Matson, J. L., Russell, D. W., Slifer, K. J., Capone, G. T., Baglio, C., & Stallings, S. (1999). Behavioral problems of children with Down syndrome and life events. *Journal of Autism and Developmental Disorders, 29*, 149–156.

Collins, L. W., & Zirkel, P. A. (2017). Funcational behavior assessments and behavior intervention plans: Legal requirements and professional recommendations. *Journal of Positive Behavior Interventions, 19*(3), 180–190.

Conners, C. K. (2008). *Conners 3rd Edition (Conners 3)*. North Tonawanda: Multi-Health Systems.

Cooper, J., Heron, T., & Heward, W. (2007). *Applied behavior analysis* (2nd ed.). Columbus: Prentice Hall/Merrill.

Crawford, J., Brockel, B., Schauss, S., & Miltenberger, R. G. (1992). A comparison of methods for the functional assessment of stereotypic behavior. *Journal of the Association for Persons with Severe Handicaps, 17*, 77–86.

Crone, D. A., Hawken, L. S., & Horner, R. H. (2015). *Building positive behavior support systems in schools: Functional behavioral assessment*. New York: Guilford.

Dart, E. H., Radley, K. C., Fischer, A. J., Collins, T. A., Terjesen, M. D, Wright, S. J., McCargo, M., & Hicks, A. J. (2017). Accuracy in direct behavior rating is minimally impacted by completion latency. Psychology in the Schools, 54, 1123–1133.

Dekker, M. C., Nunn, R. J., Einfeld, S. E., Tonge, B. J., & Koot, H. M. (2002). Assessing emotional and behavioral problems in children with intellectual disability: Revisiting the factor structure of the developmental behavior checklist. *Journal of Autism and Developmental Disorders, 32*(6), 601–610.

Delgado, C., Gonzalez-Gordon, R. G., Aragon, E., & Navarro, J. I. (2017). Different methods for long-term systematic assessment of challenging behaviors in people with severe intellectual disability. *Frontiers in Psychology, 8*, 1–9.

Didden, R., Korzilius, H., van Oorsouw, W., & Sturmey, P. (2006). Behavioral treatment of challenging behaviors in individuals with mild mental retardation: Meta-analysis of single-subject research. *American Journal on Mental Retardation, 111*, 290–298.

Dufrene, B. A., Doggett, R. A., Henington, C., & Watson, T. S. (2007). Functional assessment and intervention for disruptive classroom behaviors in preschool and head start classrooms. *Journal of Behavioral Education, 16*, 368–388.

Dufrene, B. A., Kazmerski, J. A., & Labrot, Z. (2016). The current status of indirect functional assessment instruments. *Psychology in the Schools, 54*, 331–350.

Durand, V. M., & Crimmins, D. B. (1992). *The motivation assessment scale (MAS) Administration guide*. Topeka, KS: Monaco and Associates.

Edwards, R. P. (2002). A tutorial for using the functional assessment informant record for teachers. *Proven Practice, 4*, 31–33.

Einfeld, S. L., & Tonge, B. J. (2002). *Manual for the Developmental Behaviour Checklist (second edition), Primary Care Version (DBC-P) & Teacher Version (DBC-T)*. Australia: School of Psychiatry, University of NSW, and Center for Developmental Psychiatry and Psychology, Monash University.

Emerson, E., Kiernan, C., Alborz, A., Reeves, D., Mason, H., Swarbrick, R., … Hatton, C. (2001). The prevalence of challenging behaviors: A total population study. *Research in Developmental Disabilities, 22*, 77–93.

Emerson, E., Malam, S., Davies, I., & Spencer, K. (2005). *Adults with learning disabilities in England, 2003/04*. London: NHS Health and Social Care Information.

Fee, A., Schieber, E., Noble, N., & Valdovinos, M. G. (2016). Agreement between questions about behavior function, the motivation assessment scale, functional assessment interview, and brief functional analysis of children's challenging behaviors. *Behavior Analysis: Research and Practice, 16*(2), 94–102.

Felce, D., Lowe, K., Beecham, J., & Hallam, A. (2000). Exploring the relationships between costs and quality of services for adults with severe intellectual disabilities and the most severe challenging behaviors in Wales: A multivariate regression analysis. *Australia and New Zealand Journal of Developmental Disabilities, 25*, 307–326.

Gage, N. A., Lewis, T. J., & Stichter, J. P. (2012). Functional behavioral assessment-based interventions for students with or at risk for emotional and/

or behavioral disorders in school: A hierarchical lineal modeling meta-analysis. *Behavioral Disorders, 37*, 55–77.

Gonzalez, M. L., Dixon, D. R., Rojahn, J., Esbensen, A. J., Matson, J. L., Terlonge, C., & Smith, K. R. (2008). The behavior problems inventory: Reliability and factor validity in institutionalized adults with intellectual disabilities. *Journal of Applied Research in Intellectual Disabilities, 22*, 223–235.

Gresham, F., Waston, T. S., & Skinner, C. H. (2001). Functional behavioral assessment: Principles, procedures, and future directions. *School Psychology Review, 30*, 156–172.

Gresham, F. M. (2015). *Disruptive behavior disorders: Evidence-based practice for assessment and intervention*. New York: The Guilford Press.

Gresham, F. M., & Elliott, S. N. (2008). *Social skills improvement system – Rating scales*. Minneapolis, MN: Pearson Assessments.

Hanley, G. P., Iwata, B. A., & McCord, B. E. (2003). Functional analysis of problem behavior: A review. *Journal of Applied Behavior Analysis, 36*, 147–185.

Hanley, G. P., Jin, C. S., Vanselow, N. R., & Hanratty, L. A. (2014). Producing meaningful improvements in problem behavior of children with autism via synthesized analyses and treatments. *Journal of Applied Behavior Analysis, 47*, 16–13.

Harvey, S. T., Boer, D., Meyer, L. H., & Evans, I. M. (2009). Updating a meta-analysis of intervention research with challenging behavior: Treatment validity and standards of practice. *Journal of Intellectual and Developmental Disability, 34*, 67–80.

Herzinger, C. V., & Campbell, J. M. (2007). Comparing functional assessment methodologies: A quantitative synthesis. *Journal of Autism and Developmental Disorders, 37*, 1430–1445.

Hill, J., Powlitch, S., & Furniss, F. (2008). Convergent validity of aberrant behavior checklist and behavior problems inventory with people with complex needs. *Research in Developmental Disabilities, 29*, 49–50.

Hintze, J. M., & Matthews, W. J. (2004). The generalizability of systematic direct observations across time and setting: A preliminary investigation of the psychometrics of behavioral observation. *School Psychology Review, 33*, 258–270.

Individuals with Disabilities Education Improvement Act of 2004, P.L. 108–446, 20 U.S.C. § 1400 et seq. (2004)

Ingram, K., Lewis-Palmer, T., & Sugai, G. (2005). Function-based intervention planning: Comparing the effectiveness of FBA function-based and non-function-based intervention plans. *Journal of Positive Behavior Interventions, 7*, 224–236.

Iwata, B., Dorsey, M., Slifer, K., Bauman, K., & Richman, G. (1994). Toward a functional analysis of self-injury. *Journal of Applied Behavior Analysis, 27*, 197–209. (Reprinted from *Analysis and Intervention in Developmental Disabilities, 2,* 3–20, 1982).

Iwata, B. A., Wallace, M. D., Kahng, S., Lindberg, J. S., Roscoe, E. M., Conners, J., & Worsdell, A. S. (2000). Skill acquisition in the implementation of functional analysis methodology. *Journal of Applied Behavior Analysis, 33*, 181–194.

Iwata, B. A., Worsdell, A. S. (2005). Implications of functional analysis methodology for the design of intervention programs. *Exceptionality, 13*, 25–34.

Iwata, B. L., & DeLeon, I. G. (1996). *The functional analysis screening tool (FAST)*. Gainesville, FL: University of Florida.

Iwata, B. L., DeLeon, I. G., & Roscoe, E. M. (2013). Reliability and validity of the functional analysis screening tool. *Journal of Applied Behavior Analysis, 46*, 271–284.

Jessel, J., Hanley, G. P., & Ghaemmaghami, M. (2016). Interview-informed synthesized contingency analyses: Thirty replications and reanalysis. *Journal of Applied Behavior Analysis, 49*, 576–595.

Kelley, M. E., LaRue, R. H., Roane, H. S., & Gadaire, D. M. (2011). Indirect behavioral assessments: Interviews and rating scales. In W. W. Fischer, C. C. Piazza, & H. S. Roane (Eds.), *Handbook of applied behavior analysis* (pp. 182–190). New York: The Guilford Press.

Kodak, T., Fisher, W. W., Paden, A., & Dickes, N. (2013). Evaluation of the utility of a discrete-trial functional analysis in early intervention classrooms. *Journal of Applied Behavior Analysis, 46*, 301–306.

Koritsas, S., & Iacono, T. (2013). Psychometric comparison of the motivation assessment scale (MAS) and the questions about behavioral function (QABF). *Journal of Intellectual Disability Research, 57*(8), 747–757.

Kozma, A., Mansell, J., & Beadle-Brown, J. (2009). Outcomes in different residential settings for people with intellectual disabilities: A systematic review. *American Journal on Intellectual and Developmental Disabilities, 114*, 193–222.

Larkin, W., Hawkins, R. O., & Collins, T. (2016). Using trial-based functional analysis to design effective interventions for students diagnosed with autism spectrum disorder. *School Psychology Quarterly, 31*, 534–547.

Lloyd, B. P., & Kennedy, C. H. (2014). Assessment and treatment of challenging behavior for individuals with intellectual disability: A research review. *Journal of Applied Research in Intellectual Disabilities, 27*, 187–199.

March, R. E., Horner, R. H., Lewis-Palmer, T., Brown, D., Crone, D., Todd, A. W., & Carr, E. (2000). *Functional assessment checklist: Teachers and staff (FACTS)*. Eugene, OR: Educational and Community Supports.

Mascitelli, A. N., Rojahn, J., Nicolaides, V. C., Moore, L., Hastings, R. P., & Christian-Jones, C. (2015). The behavior problems inventory-short form: Reliability and factorial validity in adults with intellectual disabilities. *Journal of Applied Research in Intellectual Disabilities, 28*, 561–571.

Matson, J. L., Kozlowski, A. M., Worley, J. A., Shoemaker, M. E., Sipes, M., & Horovitz, M. (2011). What is the evidence for environmental causes of challenging behavior in persons with intellectual disabilities and autism spectrum disorder. *Research in Developmental Disabilities, 32*, 693–698.

Matson, J. L., Tureck, K., & Rieske, R. (2012). The questions about behavioral function (QABF): Current status as a method of functional assessment. *Research in Developmental Disabilities, 33*, 630–634.

Matson, J. L., & Vollmer, T. R. (1995). User's Guide: Questions About Behavioral Function (QABF). Baton Rouge, LA. Retrieved from www.disabilityconsultants.org

Matson, J. L., & Wilkins, J. (2009). Factors associated with the questions about behavior function for functional assessment of low rate and high rate challenging behaviors in adults with intellectual disability. *Behavior Modification, 33*(2), 207–219.

Mayer, G. R., Sulzer-Azaroff, B., & Wallace, M. (2014). *Behavior analysis for lasting change (third edition)*. Cornwall-on-Hudson, NY: Sloan Publishing.

McAtee, M., Carr, E. G., & Schulte, C. (2004). A contextual assessment inventory for problem behavior: Initial development. *Journal of Positive Behavior Interventions, 6*(3), 148–165.

McIntosh, K., Borgmeier, C., Anderson, C. M., Horner, R. H., Rodriguez, B., & Tobin, T. J. (2008). Technical adequacy of the functional assessment checklist: Teachers and staff (FACTS) FBA interview measure. *Journal of Positive Behavior Interventions, 10*, 33–45.

Miller, F. G., & Lee, D. L. (2013). Do functional behavioral assessments improve intervention effectiveness for students diagnosed with ADHD? A single-subject meta-analysis. *Journal of Behavioral Education, 22*, 253–282.

Moskowitz, L. J., Carr, E. G., & Durand, V. M. (2011). Behavioral intervention for problem behavior in children with fragile x syndrome. *American Association on Intellectual and Developmental Disabilities, 116*, 457–478.

Myrbakk, E., & von Tetzchner, S. (2008). The prevalence of behavior problems among people with intellectual disability living in community settings. *Journal of Mental Health Research in Intellectual Disabilities, 1*, 205–222.

Newcomer, L. L., & Lewis, T. J. (2004). Functional behavioral assessment: An investigation of assessment reliability and effectiveness of function-based interventions. *Journal of Emotional and Behavioral Disorders, 12*, 168–181.

Northup, J., Wacker, D., Sasso, G., Steege, M., Cigrand, K., Cook, J., & DeRaad, A. (1991). A brief functional analysis of aggression and alternative behavior in an outclinic setting. *Journal of Applied Behavior Analysis, 24*(3), 509–522.

O'Neil, R. E., Horner, R. H., Albin, R. W., Sprague, J. R., Storey, K., & Newston, J. S. (1997). *Functional assessment and program development for problem behavior: A practical handbook* (2nd ed.). Pacific Grove, CA: Brooks/Cole.

Paclawskyj, T. R., Matson, J. L., Rush, K. S., Smalls, Y., & Vollmer, T. R. (2000). Assessment of the convergent validity of the questions about behavioral function scale with analogue functional analysis and the motivation assessment scale. *Journal of Intellectual Disability Research, 45*, 484–494.

Payne, L. D., Scott, T. M., & Conroy, M. (2007). A school-based examination of the efficacy of function-based intervention. *Behavioral Disorders, 32*, 158–174.

Pluymert, K. (2014). Problem-solving foundations for school psychological services. In P. L. Harrison & A. Thomas (Eds.), *Best practices in school psychology data-based and collaborative decision making* (pp. 25–39). Bethesda, MD: National Association of School Psychologists.

Powell, J., Martindale, B., Kulp, S., Martindale, A., & Bauman, R. (1977). Taking a closer look: Time sampling and measurement error. *Journal of Applied Behavior Analysis, 10*, 325–332.

Reynolds, C. R., & Kamphaus, R. W. (2015). *BASC-3 behavioral and emotional screening system manual*. Circle Pines, MN: Pearson.

Reynolds, C. R., & Richmond, B. O. (2008). *Revised children's manifest anxiety scale, 2nd edition (RCMAS-2)*. Los Angeles, CA: Western Psychological Services.

Riley-Tillman, T. C., Chafouleas, S. M., Sassu, K. A., Chanese, J. A. M., & Glazer, A. D. (2008). Examining the agreement of direct behavior ratings and systematic direct observation data for on-task and disruptive behavior. *Journal of Positive Behavior Interventions, 10*, 136–143.

Rojahn, J., Aman, M. G., Matson, J. L., & Mayville, E. (2003). The aberrant behavior checklist and the behavior problems inventory: Convergent and divergent validity. *Research in Developmental Disabilities, 24*, 391–404.

Rojahn, J., Matson, J. L., Lott, D., Esbensen, A. J., & Smalls, Y. (2001). The behavior problems inventory: An instrument for the assessment of self-injury, stereotyped behavior, and aggression/destruction in individuals with developmental disabilities. *Journal of Autism and Developmental Disorders, 31*(6), 577–588.

Rojahn, J., Rowe, E. W., Sharber, A. C., Hastings, R. P., Matson, J. L., Didden, R., … Dumont, E. L. M. (2012). The behavior problems inventory-short form (BPI-S) for individuals with intellectual disabilities II: Reliability and validity. *Journal of Intellectual Disability Research, 56*, 546–565.

Sigafoos, J., & Saggers, E. (1995). A discrete-trial approach to the functional analysis of aggressive behaviour in two boys with autism. *Journal of Developmental Disabilities, 20*, 287–297.

Smith, T. E. C., Polloway, E. A., Patton, J. R., & Dowdy, C. A. (2001). *Teaching students with special needs in inclusive settings* (3rd ed.). Boston, MA: Allyn & Bacon.

Sparrow, S., Cicchetti, D., & Saulnier, C. (2016). *Vineland adaptive behavior scale-third edition (Vineland-3)*. San Antonio, TX: Pearson.

Steege, M. W., & Watson, T. S. (2009). *Conducting school-based functional behavioral assessment* (2nd ed.). New York: The Guilford Press.

Stichter, J. P., & Riley-Tillman, C. T. (2014). Considering systematic direct observation after a century of research – Commentary on the special issue. *Behavioral Disorders, 39*(4), 245–247.

Thompson, R. H., & Borrero, J. C. (2011). Direct observation. In W. W. Fischer, C. C. Piazza, & H. S. Roane (Eds.), *Handbook of applied behavior analysis* (pp. 191–205). New York: The Guilford Press.

Tincani, M. J., Castrogiavanni, A., & Axelrod, S. (1999). A comparison of the effectiveness of brief versus traditional functional analyses. *Research in Developmental Disabilities, 20*, 327–338. https://doi.org/10.1016/S0891-4222(99)00014-1

Touchette, P. E., MacDonald, R. F., & Langer, S. N. (1985). A scatter plot for identifying stimulus control for problem behavior. *Journal of Applied Behavior Analysis, 18*, 343–351.

VanDerHeyden, A. M. (2014). Best practices in can't do/won't do assessment. In P. L. Harrison & A. Thomas (Eds.), *Best practices in school psychology: Data-based and collaborative decision making* (pp. 305–316). Bethesda, MD: National Association of School Psychologists.

Wadsworth, J. P., Hansen, B. D., & Wills, S. B. (2015). Increasing compliance in students with intellectual disabilities using functional behavioral assessment and self-monitoring. *Remedial and Special Education, 36*, 195–207.

Vocational Skills and Their Assessment

25

Miriam Heyman, Holly E. Jacobs, and Gary N. Siperstein

Introduction

Almost 30 years ago, the Americans with Disabilities Act (ADA) was enacted. This landmark legislation was hailed as "a pillar of civil rights," as it mandated equal access across virtually every sector of society (Obama, 2013). The ADA prohibited employment discrimination on the basis of disability and required employers to provide "reasonable accommodations" to employees with disabilities to enable access to work. However, almost 30 years after the passage of this monumental civil rights mandate, the vast majority of adults with intellectual and developmental disabilities (IDD) remain unemployed. The rate of competitive employment for people with intellectual disabilities hovers below 20% (Siperstein, Parker, & Drascher, 2013).

The absence of employment for people with IDD who are capable of work is problematic for several reasons. Unemployment blocks access to financial independence and thus poses an unnecessary burden on taxpayers (Schur, 2002). Businesses are missing out on a potential boost to their bottom line because companies that hire people with disabilities, including IDD, benefit from an improved public perception of those companies and therefore enjoy a wider and more loyal customer base (Burge, Ouellette-Kuntz, & Lysaght, 2007; Siperstein, Romano, Mohler, & Parker, 2006). And not to be overlooked is the fact that the lives of people with IDD can be greatly enhanced through work. We know that employment generates positive results for people with IDD, including improved self-esteem and opportunities to develop meaningful social relationships (Kober & Eggleton, 2005; Schur, 2002).

The low employment rate for adults with IDD is somewhat puzzling, because it exists despite the tremendous potential of the ADA. The low employment rate also persists despite tremendous financial investment provided by the federal government to promote competitive employment for all adults with disabilities through federal initiatives including *Employment First*, as implemented by the United States Department of Labor. *Employment First* identifies competitive employment as "the first option for employment services for youth and adults with significant disabilities" (United States Department of Labor, 2014), including IDD, and it provides platforms through which leaders from across the country can work together to enact change. This initiative is intended to advance *competitive* employment: employment opportunities that enable adults

M. Heyman
Ruderman Family Foundation, Boston, MA, USA

H. E. Jacobs (✉) · G. N. Siperstein
Center for Social Development and Education, University of Massachusetts Boston,
Boston, MA, USA
e-mail: holly.jacobs@umb.edu

J. L. Matson (ed.), *Handbook of Intellectual Disabilities*, Autism and Child Psychopathology Series, https://doi.org/10.1007/978-3-030-20843-1_25

with IDD to work alongside co-workers without disabilities and earn at least minimum wage. This is in contrast to sheltered employment, where most workers have disabilities and often do not earn minimum wage. Competitive employment is the focus of current federal policy, and it is also the focus of this chapter.

Given the ADA and federal financial investment, why do so many adults with IDD lack competitive employment? The problem is complex; if there were a simple solution to boost the employment rate, that solution would have been implemented decades ago. One important barrier is that many parents, service providers, educators, employers, and people with IDD themselves hold low expectations for employment. Spreading the mindset that people with IDD can and should be competitively employed will be a key component to the change that we wish to see. The *Employment First* initiative, with the premise that competitive work should be the goal for all adults with IDD, is an important step toward promoting this mindset.

A second key component to this problem of pervasive unemployment is the need to identify the skills that facilitate success in the workplace for people with IDD and the most effective techniques to assess and teach these skills. Skills are critical because if someone with IDD is hired for a job for which they are not qualified, the story will not end well. The employee won’t experience workplace success and the pride and satisfaction that accompany it. Meanwhile, the employer will be hesitant to hire individuals with IDD in the future.

Fortunately, there is a vast amount of knowledge about the skills that facilitate access to jobs and success on the job for individuals with IDD. Researchers have spent decades identifying and untangling the skills that predict positive employment outcomes. A second and related topic of research has focused on assessment, in order to identify methods to assess individuals’ skill levels. This measurement ultimately enables individuals to be hired for the jobs for which they are qualified and supports programs to deliver instruction that is appropriate for each individual learner. Finally, a third strand of research has focused on programs, with the goal of identifying how to most effectively teach the skills that have been identified as the most critical. Considerations include instructional strategies, contexts for instruction (i.e., school-based or on the job), and who should receive this instruction and when.

Throughout the chapter, as we review what we know about skills, assessment, and programs, we take a holistic approach and strive to apply a contextual lens to the accumulated knowledge about skills, assessment, and programs. This contextual lens includes the acknowledgement, for example, that behaviors and skill levels vary within and across environments. Two people within one environment bring different assets, and their behaviors and skills vary accordingly. Equally important is the contention that one person might manifest different skills and behaviors across two different contexts since environmental supports and demands are unique to each place. A holistic approach, which identifies and targets individual skill levels in addition to environmental opportunities and constraints, will lead to new opportunities for employment for individuals with IDD.

Skills

A positive work experience depends on a good “fit” between the employer and the employee. This fit involves a number of factors, including schedules, degree of flexibility, employee interests, location and commute, and more. Also, and perhaps most critically, a person must be qualified for the job, meaning they must possess the skills necessary to succeed on the job. The sections that follow summarize the research on the skills that are related to employment success, including finding and keeping a job, for individuals with IDD. These skills include adaptive skills and self-determination and advocacy skills, which are relevant for virtually any occupation. They also include the finite skills that are only relevant for very specific occupations, such as bagging groceries or working a cash register. Each skill is presented from a contextual lens, such that the role of the environment in eliciting or obstructing a skill is considered.

Adaptive Skills

The construct of adaptive functioning is defined as conceptual, social, and practical skills utilized in everyday life or "the behavioral skills that people typically exhibit when dealing with the environmental demands they confront" (Luckasson et al., 2002; Widaman & McGrew, 1996, p. 97). A critical element of the definition of adaptive functioning is the emphasis on skills *typically* exhibited *within environments*. Demands and resources both differ across environments, and therefore adaptive functioning is highly context-dependent. An individual might receive reminders of socially appropriate behaviors from a job coach at work but lack these reminders at home. Therefore, this person's adaptive functioning, specifically social skills, will differ across work and home. Specific adaptive skills include communication, social skills, and daily living skills, such as dressing, eating, bathing, and navigating transportation systems. By definition, individuals with IDD have limitations in adaptive functioning; these limitations are a necessary condition for diagnosis of intellectual disability (American Association on Intellectual and Developmental Disabilities, 2018a; American Psychiatric Association, 2013).

For individuals with IDD, decades of research make it abundantly clear that adaptive skills, particularly social skills, facilitate workplace success (e.g., Malgady, Barcher, Towner, & Davis, 1979; McDermott, Martin, & Butkus, 1999; Tomaszewski, Fidler, Talapatra, & Riley, 2018; White & Dodder, 2000). These skills have also been indirectly identified by federal policy makers as critical to employment success. In 2014, the Workforce Innovation and Opportunity Act (WIOA) was signed into law, which requires vocational rehabilitation (VR) agencies to provide pre-employment transition services to students with disabilities. These services, moreover, must include workplace readiness training that addresses adaptive skills, including communication, cooperation, good manners, and good hygiene.

Social skills fall under the umbrella of adaptive skills and emerge as salient to success throughout the employment process. Several studies have identified behavior problems and/or lack of social skills as a barrier to obtaining competitive employment (e.g., Lemaire & Mallik, 2008; Mank, Cioffi, & Yovanoff, 1998; Moran, McDermott, & Butkus, 2001; Murphy, 2009; Noel, Oulvey, Drake, & Bond, 2017), and other studies have found that social skills are important for workplace success, and therefore the influence of social skills extends beyond simply getting a job. For example, higher social skills are related to higher supervisor ratings (Butterworth & Strauch, 1994; McConaughy, Stowitschek, Salzberg, & Peatross, 1989) and have been linked with higher levels of on-the-job productivity (Park, Kim, & Kim, 2016) and longer periods of job tenure (Foss & Peterson, 1981).

Social skills are arguably the most critical for workplace success, but other skills under the construct of adaptive functioning, such as hygiene and self-care skills, are relevant to the conversation. Self-care includes abilities to feed, bathe, and dress and is thus closely linked to hygiene, a critical consideration for employers who rely on employees to represent them and their businesses. It is easy to imagine a scenario in which a person who lacks ability in self-care and/or hygiene interviews for a position that involves interaction with clients and does not receive an offer because the interviewer is hesitant to have this person represent the organization. It is therefore not surprising that proficiency in self-care among adults with IDD is associated with the likelihood of getting a job (Carter, Austin, & Trainor, 2011; Carter, Austin, & Trainor, 2012; Poppen, Lindstrom, Unruh, Khurana, & Bullis, 2017). In one study of adolescents with severe disabilities, those who communicated well with others and were independent in their self-care (i.e., were able to feed and dress themselves) were more likely than their peers without these skills to have paid employment (Carter et al., 2011). Another study examined 4443 Oregon high school students with disabilities and found that challenges with self-care reduced the likelihood that students obtained employment (Poppen et al., 2017). Moreover, hygiene is not only important for getting a job—it is also important for keeping a job. For people with IDD, hygiene has been cited as an essential factor in job tenure (Spence, 2004). Skills related to hygiene and self-care might be necessary for positive social interactions in the workplace because clients and co-workers might be more willing to socialize with individuals who have prerequisite skills in these areas. These social interactions, in turn, are critical for learning on the job and thus enhance work success and tenure.

Transportation skills are a third critical element of adaptive functioning and one that receives relatively little attention in the research literature but remains an essential consideration. The first responsibility of an employee is to show up on time—first to the interview and then to the first day on the job. Candidates and employees need modes of transportation that will get them from Point A to Point B, *reliably and on time*. They also need to understand how to access this transportation, which can be difficult given the complexity of many public transportation systems. A few studies have examined the role of transportation in promoting positive employment outcomes (Carter et al., 2011; Moore, Feist-Price, & Alston, 2002; Simonsen & Neubert, 2013). Transportation skills are associated with obtaining competitive employment (Simonsen & Neubert, 2013), as are transportation services (provided under a larger umbrella of vocational rehabilitation services) and the availability of accessible transportation options in the community (Carter et al., 2011; Moore et al., 2002).

Not only do transportation skills facilitate getting a job, expertise in transportation might enable people to get a job *of their choosing*. The more adept a person is at navigating a transportation system, the more potential employers he or she can access, so that people are not restricted to the one or two businesses within walking distance or on a single bus line. While transportation barriers to employment are pervasive (Noel et al., 2017), at least one study has demonstrated the efficacy of an intervention designed to enhance the navigation skills of young adults with IDD (Price, Marsh, & Fisher, 2017), indicating the potential for meaningful progress in this area.

A vast body of research identifies social skills, hygiene, and transportation skills as essential ingredients for successful employment outcomes for adults with IDD. While skills are usually conceived of as “things” that reside within a person, it is important to acknowledge that they also depend on context. Many of the studies cited above that focused on transportation as a component of employment success utilized transportation as an ecological variable—researchers measured and reported the receipt of transportation services within a VR service system (Moore et al., 2002), or the availability of accessible transportation within and across communities (Carter et al., 2011). Since transportation (or lack thereof) is a huge barrier to employment for adults with ID (Noel et al., 2017), policy makers and practitioners should address this issue on both individual and systemic levels. This would likely involve teaching people to utilize public transportation and also expanding the availability of accessible public transportation.

The contextual approach is relevant for social skills and hygiene as well. For example, frustration and/or boredom can elicit behavior problems, and behavioral outbursts are often incompatible with rules for social interaction in the workplace. Therefore, the most impactful way to enhance someone’s social functioning might be to remove or modify the elements of the environment that are frustrating or boring. Similarly, environmental modifications can enhance self-care abilities. For example, employers can add visual aids above bathroom and kitchen sinks, and these aids can serve as reminders to wash hands or brush teeth. These contextual approaches, alongside interventions targeting individual skill levels, have great potential.

Self-Determination and Self-Advocacy Skills

A policy statement from the American Association on Intellectual and Developmental Disabilities asserts that “People with intellectual and/or developmental disabilities have the same right to self-determination as all people. They must have opportunities and experiences that enable them to exert control in their lives and to advocate on their own behalf” (AAIDD, 2018b). Self-determination, moreover, is largely dependent on self-advocacy skills, which depend on awareness of one’s abilities and challenges, knowledge of the types of supports and accommodations that facilitate success, and the abilities to communicate effectively in order to receive these supports and accommodations.

The possession of these skills contributes to positive employment outcomes. Higher levels of self-determination are associated with longer job retention, more job satisfaction, and better job performance (Akkerman, Kef, & Meininger, 2018; Fornes, Rocco, & Rosenberg, 2008). This relationship between self-determination and employment success is intuitive. When people like the work that they do, when they find it meaningful, and when they go to work on their own accord, they are motivated to do well at work. In these ideal circumstances, work is more than a paycheck—it is a source of identity, self-esteem, and social connectedness. When people gain these nonfinancial benefits from work, they devote more energy and passion to the job, and their performance benefits. The field has recognized self-determination and self-advocacy as critical for employment success, and interventions designed to improve employment outcomes target these areas (Green, Cleary, & Cannella-Malone, 2017).

Finite Skills

All of the research on skills reviewed thus far rests on a basic premise that for any given position a person must meet minimum requirements. These requirements might include minimal fine or gross motor skills, or color differentiation abilities for sorting tasks, for example. This chapter began with the acknowledgement that if someone with IDD is hired for a job for which they are not qualified, the experience will not be positive. The employee will not achieve workplace success and the fulfillment that accompanies it. Meanwhile, the employer will not benefit from the experience and will be reluctant to hire adults with IDD in the future.

The skills highlighted thus far, including adaptive and self-advocacy skills, are important for employment generally. The vast majority of positions require the ability to interact with co-workers and clients, and thus social skills are usually if not always relevant. In addition to these universally relevant skills, certain jobs require specific skills that many not be relevant for other businesses or positions. Early research on employment of adults with IDD focused on the identification of specific skills required for specific jobs (Cohen & Williams, 1961; McCartney, 1958; Posner, 1966). For example, one study identified the following activities (and skills required for those activities) as essential for work within food and domestic service positions: making beds, cleaning windows, polishing, mopping, waxing, dusting, sweeping, setting tables, cleaning food utensils, serving food, washing, ironing, and folding clothes (Cohen & Williams, 1961). This study also outlined perquisite skills for employment within the industry and business sectors (Cohen & Williams, 1961). Other early work emphasized manipulative dexterity as a critical skill for the types of jobs that were available to adults with IDD, including packaging and assembling (Tobias & Gorelick, 1960). For adults with IDD, extent of manipulative dexterity was considered indicative of work ability (Neeman, 1986; Tobias & Gorelick, 1960).

From this foundational research, the field has evolved to develop a focus on the more broadly defined skills, like social skills. This new focus on the broader skills should not be interpreted as finite skills lacking importance. More currently, researchers have turned their attention to complex skills, like social skills, which are perhaps more difficult to define and measure but still important.

The identification of critical skills comprises an essential contribution to the field of employment for IDD. A second key contribution is the research on assessing these skills, given the fact that assessment must precede teaching in order to ensure that the skills being taught are relevant for each individual learner. The following section provides an overview of the field of assessment and describes considerations related to assessment spanning the entire employment process—from before a person enters a vocational training program, continuing throughout participation in a program, and as needed after the person has begun employment.

Assessment

Assessment is a key consideration even at the early stage of the employment process, before an individual has entered a vocational training program (Marston, 2015). In order to maximize the likelihood that any given program will be effective for any given individual, it is critical to ensure a good match between program and participant. The skill levels and interests of a potential program participant need to be identified in order to avoid a situation in which a person enters a program that does not address his or her goals and/or provide appropriate level of instruction. Other considerations at the outset include the type of job the person wants and what skills are necessary for that occupation, what types of jobs are available in the area (and again what skills are necessary for these jobs), and what types of supports will be available after the person starts working and how these supports (or lack thereof) will impact what skills are necessary (Marston, 2015). Thus, assessment must occur at the level of a person (i.e., the abilities and interests of a job candidate) and also at the level of the environment (i.e., availability of job opportunities and supports). The sections that follow describe research at both of these levels.

Person-Level Assessment

At the person level, since many key skills fall under the umbrella of adaptive functioning, the research on assessment of adaptive functioning is of central importance. As described in the review of skills, adaptive functioning, defined as "the ability to respond to environmental demands" (Arias, Verdugo, Navas, & Gómez, 2013, p. 156) or "the behavioral skills that people typically exhibit when dealing with the environmental demands they confront" (Widaman & McGrew, 1996, p. 97), is a construct that subsumes many of these considerations, particularly daily living skills and social skills and, relatedly, reaction to supervisors. Adaptive functioning is a multidimensional domain, with constructs including daily living and social skills, among others. There are four scales of adaptive behavior that meet the following criteria: reflect a multidimensional construct which includes multiple distinct domains; have demonstrated adequate reliability and validity; and "have been standardized on people with and without ID" (Arias et al., 2013, p. 157). These four measures include (1) the Vineland Adaptive Behavior Scales Third Edition (Sparrow, Cicchetti, & Saulnier, 2005); (2) the Adaptive Behavior Assessment System Third Edition (Harrison & Oakland, 2015); (3) the Scales of Independent Behavior-Revised (Bruininks, Woodcock, Weatherman, & Hill, 1996); and (4) the Adaptive Behavior Scale-School Version Second Edition (Lambert, Nihira, & Leland, 1993). Clearly, the School Version scale was not designed for implementation in a vocational training program or in the workplace.

Several studies that have identified relevant employment skills utilize one or more subscales of the measures listed above or use items selected from these measures, as their predictors of employment (Siperstein, Heyman, & Stokes, 2014; Tomaszewski et al., 2018; White & Dodder, 2000). For example, performances on the socialization, communication, and daily living subscales of the Vineland Adaptive Behavior Scales were each explored in relation to employment outcomes in one study of adults with Down syndrome (Tomaszewski et al., 2018). Within each subscale of this assessment, there are additional subcategories, providing the opportunity for assessment of more specific facets of adaptive functioning. The communication subscale yields specific scores for receptive, expressive, and written communication skills. The daily living subscale provides scores for personal (i.e., hygiene and self-care), domestic, and community (i.e., transportation) skills. The socialization subscale includes coping skills, interpersonal skills (i.e., social skills), and engagement in play and leisure. Thus, general measures of adaptive functioning also provide assessment data that is specific to several skills that have been identified as relevant for employment success, including hygiene, transportation, and social skills.

In addition to the measures of social skills within larger measures of adaptive functioning, there are also measures that focus on this skill set explicitly. For example, the Social Skills Improvement System (formerly the Social Skills Rating System) provides scores on each of seven social skills (communication, cooperation, assertion, responsibility, empathy, engagement, and self-control) and each of five problem behaviors (externalizing, bullying, hyperactivity/inattention, internalizing, and autism spectrum; Gresham & Elliott, 2008). Items from this measure have been used to generate social skill scores, which are then explored in relation to employment outcomes (Carter et al., 2011). Importantly, the use of any scale of adaptive behavior or social skills should reflect the premise that because adaptive functioning represents behavior in the context of environmental demands, levels of adaptive functioning will differ across context, including time and place. The supports and demands in a workplace differ from the supports and demands at home, and a person's level of functioning will vary accordingly.

Functional behavioral assessment (FBA) is a technique that is designed to "find effective ways of addressing significant challenging behaviors" (Marston, 2015, p. 117), and it is relevant to workplace assessment because challenging behaviors are a critical barrier to employment obtainment and retention for adults with IDD. The underlying premise behind FBA is that behavior is goal-directed. With FBA, the problematic behavior is defined and observed to yield information about the frequency of the behavior and also the antecedents and consequences of that behavior. Information about the antecedents and consequences informs intervention. It might be possible to remove triggers for problematic behaviors once these triggers (antecedents) have been identified. Or, a FBA might demonstrate that a consequence (i.e., being removed from the job site) is actually serving to reinforce the behavior, if the consequence is in fact desired by the person exhibiting the challenging behavior. After the assessment has yielded one or more hypotheses about the function of a behavior, functional behavioral *analysis* involves the systematic manipulation of antecedents and consequences in order to reduce or eliminate the identified challenging behavior.

As the section on skills describes, in addition to adaptive and social skills, self-determination and self-advocacy have been identified as fundamentally important to positive employment outcomes. The ARC's Self-Determination Scale (Wehmeyer, 1996) is a commonly used measure that was specifically designed for individuals with IDD. The scale measures self-determination generally and also subcomponents including autonomy, self-regulation, psychological empowerment, and self-realization. Scores from this assessment are related to positive employment outcomes, including job satisfaction and job retention (Fornes et al., 2008).

Finally, the measures reviewed thus far are assessments of complex and multifaceted skills, adaptive behavior including social skills, and self-determination. While these skills and their assessment are practically universally relevant for employment of adults with IDD, it is also essential to assess the finite and specific skills that are required for specific tasks. Foundational work in this area culminated in assessments of dexterity and fine motor skills, including the Purdue Pegboard Assembly Task and O'Connor Finger Dexterity Test as measures of finger dexterity, the Hand-Tool Dexterity Test, and the Steadiness Test, which measures accuracy of movement (Bennett, 1972; Corlett, Salvendy, & Seymour, 1971; Human Resources Center, 1967; Neeman, 1986; O'Connor, 1965; Tobias & Gorelick, 1960). These measures indicate ability to perform the finite tasks in industrial and clerical areas, such as packaging, assembling, typing, etc.

As the preceding paragraphs demonstrate, the field of assessment has produced instruments that measure and document the skills and behaviors that are crucial for employment success. With the data that we generate from any of these assessment techniques and when measuring progress on any skill, we should not only ask “did this person learn this skill?” but also “does this skill translate into something meaningful?” For example, one vocational training program resulted in a 50% reduction in need for support on the job (Gomes-Machado, Santos, Schoen, & Chiari, 2016). Even when the skills being taught and assessed are finite and specific, the outcome can have meaningful impact in the real world.

Job-Level Assessment

At the job level, assessment must yield an understanding of the skills that are necessary for a person to complete assigned job duties. Decades ago, researchers understood the importance of identifying the “microunits” of a job or the specific tasks and their associated physical and cognitive requirements (Drewes, 1961). This recognition has led to the use of techniques including task analysis, in order to break down skills and ultimately provide relevant and simple instruction.

Job-level assessment also provides an understanding of the work environment including what might be challenging and what might be supportive to the individual. In a sense, then, FBA can be considered an assessment at the job level since its purpose is to identify environmental determinants of behavior. A holistic approach to job-level assessment is an inherent aspect of customized employment and an approach which is increasingly recognized as a promising alternative to more traditional strategies for obtaining employment, such as submitting an application that aligns with a specific job description put forth by an employer (Nord, Luecking, Mank, Kiernan, & Wray, 2013). Customized employment, in contrast to these traditional strategies, involves creating a job that aligns with the unique and individualized skills, interests, talents, challenges, and limitations of an adult with ID. The creation of this job requires an assessment (either formal or informal) of the workplace in order to ensure appropriate alignment. The approach is promising, as research has begun to link customized employment practices with positive employment outcomes that extend beyond obtaining employment and encompass things like higher wages and more hours (Nord et al., 2013).

The following section reviews research that has examined how to most effectively teach the skills that facilitate employment success for adults with IDD, beginning by describing how a holistic approach can be applied to program implementation. Successful program implementation, in turn, will impart necessary skills for employment.

Programs

This section presents research on programs for individuals with IDD that aim to facilitate success in competitive employment, including the ability to obtain a job, keep a job, and experience career development and advancement. The section begins with an overview of very specific instructional strategies, including specific app-based technology, and how these instructional strategies are utilized in employment programs. Following the review of specific instructional strategies, the section continues with general considerations related to program implementation. These considerations include program context and the extent to which instruction should be delivered in a classroom or in the workplace. Attention is also paid to the emergence of opportunities for postsecondary education for adults with IDD and postsecondary education as a new context for employment programs. Other considerations include how programs can address holistic circumstances that are unique to each program participant (i.e., the availability of social support).

Instructional Strategies

There is a vast amount of research on instructional techniques and approaches to teaching employment skills, including Applied Behavioral Analysis (ABA), audio-based cues, video instruction, and more (Gilson & Carter, 2016; Gilson, Carter, & Biggs, 2017; Nord et al., 2013; Park et al., 2016). ABA was developed in the 1960s for young children with autism and rests on the premise that behavior is contingent on the environment. Therefore, rewarding desired behaviors increases the likelihood that an individual will continue to display these behaviors. For several decades, ABA has also been widely utilized to enhance employment skills of adolescents and adults with IDD (Wakabayashi, 2009). One early study on this approach demonstrated the effectiveness of ABA within a job training program in improving social skills, particularly reducing noncompliance and inappropriate social interaction (Bates & Pancsofar, 1983).

Promisingly, one meta-analysis of studies examining job-related social skills training found that a wide variety of instructional approaches, including direct instruction and instruction via technology, each yielded positive effects (Park et al., 2016). This meta-analysis also found that direct instruction, where an in-person teacher delivers a curriculum, had a larger positive effect than other strategies, including video- or Internet-based approaches. An example of a direct instruction program is the WAGES program, a job-related social skill curriculum for adolescents delivered via 33 lessons with activities including resume building and mock interviews (Murray & Doren, 2013). The WAGES program has yielded improvements in adolescents' vocational expectations, social skills, and skills relevant to the workplace, such as time management and problem-solving skills (Murray & Doren, 2013).

WAGES is an example of direct instruction delivered within a classroom setting, and despite the evidence for its effectiveness, techniques other than direct instruction may be particularly useful settings outside of the classroom. For example, technology can play a critical role on the job, especially when proximity to a live coach limits an employee's interactions with co-workers, who could potentially serve as sources of natural support (Gilson & Carter, 2016). Technology such as in-ear devices can allow employees to receive cues from coaches and can also facilitate physical space from these coaches, providing opportunities for natural interaction with co-workers (Gilson & Carter, 2016). Technology can be instrumental in providing cues to help employees with IDD achieve specific job-related tasks, such as an assembly task in a factory (Mihailidis et al., 2016). It can also provide cues for more general workplace competencies, including social skills (Gilson & Carter, 2016).

Program Context

In addition to instructional strategies, the context of the program itself is an important consideration and refers to the questions of whether programs should deliver skill instruction in the workplace or in the classroom and the type of classroom setting most conducive to relevant skill instruction. Given the salience of adaptive skills for employment success, a review of the research relating to this skill category seems especially important. Hundreds of studies have examined instructional methods for teaching adaptive skills to people with IDD; the ones included in this discussion have an explicit employment focus.

For programs that are delivered in the classroom, practitioners and policy makers need to juggle teaching adaptive skills and an emphasis on inclusion (Bouck, 2010). One study utilizing data from the National Longitudinal Transition Study-2 (NLTS2), including 99,923 students with IDD, identified an overall lack of "life skills" instruction in secondary school, especially for students with mild IDD (Bouck, 2010). The author concluded that:

> given the current educational policy situation (i.e., a predisposition towards inclusive general education placements for students with disabilities and participation in the accountability system for all students), educators who believe in the value of a life skills curriculum will need to be creative in its implementation. (p. 1093)

While the importance of maximizing the inclusion of students with IDD in general education cannot be overstated, it should not be at the expense of teaching the life skills that are important for future employment. This comes with the caveat, however, that one study that analyzed data from the NLTS2 compared employment outcomes between high school students with moderate and severe IDD that received academic instruction and high school students that received instruction emphasizing functioning skills (i.e., adaptive skills; Bouck, 2010). There were no differences in employment outcomes between the two groups, possibly indicating that present methods of school-based instruction of adaptive skills including social skills are limited in its effectiveness, a conclusion that has been supported by other studies as well (Leffert & Siperstein, 2003). However, since we know that adaptive skills including social skills are perhaps the most important for employment success, these findings should not be interpreted as a call to cease instruction in these areas. Rather, the findings suggest the importance of finding new and impactful methods for school-based instruction of adaptive and social skills, particularly given the push for inclusive education as noted above.

In addition to this need to identify effective methods of school-based social skill instruction, social skill instruction should not be *restricted* to the classroom. In one longitudinal study, adults with IDD who entered employment demonstrated gains in social skills (Stephens, Collins, & Dodder, 2005). The sample for this study consisted of 2760 individuals who were receiving services from the Developmental Disabilities Division of the Oklahoma Department of Human Services, and their employers and positions varied. This suggests that the overall experience of employment, independent of explicit instruction, promotes social skills. The relationship between social skills and employment is therefore bidirectional—in addition to the agreed-upon fact that social skills facilitate employment success, time spent working provides the opportunity for social skill development.

There is a convincing body of research indicating that *early* work experience benefits job seekers with IDD, and this consistent finding is an argument for expanding instruction beyond the classroom (Benz, Lindstrom, & Yavanoff, 2000; Carter et al., 2012; Simonsen & Neubert, 2013). In one study of youth who were participating in a transition program in Oregon, those who had paid jobs while they were in the program were more likely to have competitive employment after the program had ended (Benz et al., 2000). In another study of transitioning youth, prior work experience was a significant predictor of community employment (Simonsen & Neubert, 2013). A third study demonstrates that having a paid job while in high school is strongly predictive of competitive employment 2 years after graduation (Carter et al., 2012). These findings make intuitive sense: it benefits all job seekers to have an employment history on their resume and to provide the names and contact information of former supervisors who were pleased with their performance. Also, work experience provides the opportunity to gain social skills, as described earlier. Moreover, not only is it important to have work experience, it is important to have *early* work experience. In one nationally representative study of adults with IDD, 72% of the adults who were competitively employed held their first jobs before they turned 21 years old (Siperstein et al., 2014). From this body of work, we can conclude that successful employment programs incorporate work experience. Also, this work experience should occur early—the path to employment should begin before youth transition out of the public school system.

Questions related to program context are complex, and there is not a one-size-fits-all approach. Despite opportunities to learn on the job as described in the previous paragraphs, a meta-analysis of studies that evaluated job-related social skills training programs for secondary students found that school-based programs were more effective than job-based programs in achieving social skills outcomes, specifically empathy, cooperation, and assertiveness (Park et al., 2016). It is important to note that this meta-analysis only included secondary students. In light of the documented importance of social skills, it is imperative to identify the program context that works best for each individual, based on their age, disability status, preferences, and other factors.

An additional consideration related to program context is the idea that it does not have to be a question of either/or—programs can have school-based and employment-based elements. In review of employment skill programs for secondary students with ID, Gilson et al. (2017) identified 10 studies (out of 56 included in their review) that were school-based initially but had an employment or community generalization component. Schools must have some level of involvement in explicitly preparing students with IDD for employment, since federal law mandates career development for students before they leave the public education system (Biggs & Carter, 2016). The question that remains, then, is how to use in-school programming time most effectively.

Also, the question of program context contains more nuance since the Higher Education Opportunity Act of 2008 facilitated access to postsecondary education for students with disabilities one decade ago, and as a result, as many as 40% of colleges and universities around the country are including students with IDD on campus (Grigal, Hart, & Weir, 2012). Research indicates that students with disabilities who attend postsecondary education enjoy diverse and meaningful benefits across domains, including health and social relationships (Butler, Sheppard-Jones, Whaley, Harrison, & Osness, 2016). Improving employment outcomes for people with disabilities is an explicit goal of inclusion in postsecondary education, and there is preliminary data showing that postsecondary education leads to positive employment outcomes (Butler et al., 2016; Moore & Schelling, 2015). These studies do not differentiate between students with mild and more severe IDD, and more longitudinal studies are needed in order to conclude that postsecondary education leads to long-term employment success.

Also, a focus on promising practices *within* postsecondary education programs is just beginning. Grigal et al. (2012) identified 149 institutions of higher education that serve students with IDD, and they report great variability across these programs. Programs vary in the extent to which they offer and/or emphasize a wide range of services, including career and vocational counseling, self-advocacy instruction, internships, natural supports, person-centered planning, collaboration with vocational rehabilitation agencies, and more (Petcu, Chezan, & Van Horn, 2015; Plotner & Marshall, 2016). Despite this variability, evidence suggests that the experience of postsecondary education is a valuable one for students with IDD. One study explicitly compared outcomes across an inclusive versus segregated model of postsecondary programming for students with IDD and found that students who attended both types of programs were more likely to be employed after school completion than students with IDD who never attended postsecondary education (Moore & Schelling, 2015). From some postsecondary programs, success rates are extraordinarily high; for example, 80% of graduates from one particular program achieved long-term competitive employment following graduation (Kelley & Buchanan, 2017). This program attributes its success to multiple factors, several of which are highlighted in this chapter, including the provision of work experience while participants are still in school, holistic approaches that incorporate perspectives of family members and other natural supports, and person-centered planning that takes into account individuals' preferences and skills, thus adhering to principles of self-determination (Kelley & Buchanan, 2017). It is also likely that the experience of postsecondary education results in the accumulation of social capital through relationship building and personal connections to future employers—an ingredient for employment success for people without IDD. In addition, successful postsecondary education programs should address and promote adaptive skills of program participants. According to one study, students in postsecondary education report that adaptive skills are an area of concern and difficulty, indicating that these programs should support students in these areas (Berg, Jirikowic, Haerling, & MacDonald, 2017). Emphasis on adaptive skills should not end in high school.

Finally, success of postsecondary education programs in the realm of employment comes with a caveat that is still preliminary, as research in this area is new—programs may have limited success in opening doors to diverse employment options for students with IDD. One study compared graduates of two postsecondary education programs to young adults with IDD who did not attend postsecondary education. While employment rates for the postsecondary group were higher than the comparison group, the postsecondary group and the comparison group had similar occupations (Moore & Schelling, 2015). It is possible that the graduates of secondary and postsecondary institutions chose similar occupations, but it is also possible that regardless of educational level, certain occupations remain elusive to people with IDD. Employment fields in which no graduate worked included art, design, installation/maintenance/repair, and programming/mathematician, among others. Ultimately, the field should strive for postsecondary education programs that will result in employment *and* in the ability to choose widely among potential occupations.

Holistic Considerations

In addition to program context, successful employment programs are those that are holistic in the sense that they take other environmental opportunities and constraints into account. For example, because access to transportation and related services has emerged as a significant predictor of positive employment outcomes for adults with IDD, programs can only be successful to the extent that they consider and address each individual participant's transportation opportunities and limitations (Moore et al., 2002; Noel et al., 2017). Other holistic considerations include a person's level of social support. Studies show that parental expectations predict employment such that higher expectations relate to more successful employment, presumably because parents with high expectations help their children to access the most relevant programs and also because parents' expectations might motivate children (Carter et al., 2012).

Successful programs look beyond the skill levels and characteristics of individuals served and consider the larger ecology of factors that contribute to workplace success. This is a complicated and difficult mandate, but there are programs that implicitly acknowledge the need to look beyond individual characteristics of potential employees with IDD. For example, the Bridges from School to Work Program serves youth with all disabilities (including but not limited to IDD) and is an "employer-driven program," meaning that program staff considers the needs of each individual employer, in addition to the skills and interests of the youth served by the program (Hemmeter, Donovan, Cobb, & Asbury, 2015). The skills that are targeted through intervention thus reflect the employment landscape—who is hiring in the local area, for what position, etc. More than 18,000 youth across the country have participated in the program, which provides career counseling, help with job placement, and on-the-job support (Hemmeter et al., 2015). The program has demonstrated success; at age 30, approximately 75% of the individuals who were in the Bridges Program as youth were employed (Hemmeter et al., 2015).

As we have seen throughout this section, there are a wide range of program techniques and contexts. Program techniques include ABA, the use of technology, direct instruction, or some combination of each. The program context refers to whether it is school-based or employment-based and the individual participants' ecology—factors such as availability of social support, the needs of local employers, and more.

Conclusions

The vast majority of adults with IDD have the potential to work yet lack access to competitive employment (Siperstein et al., 2013). This is despite decades of research focusing on employment for this population. From this research, we have identified the skills that are primary predictors of successful competitive employment for adults with ID, including social skills, and other skills falling under the umbrella of adaptive functioning, like transportation and self-care. There is also a solid base of research indicating which experiences lead to competitive employment; early work experience is pivotal, and it seems like research is now converging on postsecondary education as a step toward employment. We are beginning to address questions about how to deliver social skills in changing contexts, such as secondary education classrooms that are increasingly inclusive and postsecondary education programs that are welcoming more and more students with IDD. We have identified impactful instructional techniques and techniques for assessment. We also underscore the importance of assessing individual skill level, along with environmental opportunities and constraints. We have effective and reliable techniques for each type of assessment.

The current challenge, then, is to merge all that we know into a holistic approach. Research, practice, and policy must reflect the premise that there is not a one-size-fits-all approach to employment. People's unique life circumstances will dictate which employment experiences will be suitable for them and which will not. For example, individuals who receive Social Security Disability Insurance have to follow specific guidelines in order to maintain receipt of benefits (Nord et al., 2013). Also relevant are individual skills, preferences, dreams, geographic areas, and more. Skill instruction, program design, and assessment each needs to reflect the reality that a one-size-fits-all approach will not work. An approach that considers the unique circumstances of each individual participant is especially challenging in light of the "fragmented education, vocational rehabilitation, and human services systems" that serve adults with IDD, yet it is necessary nevertheless (Nord et al., 2013).

Also, as much of the research cited in this chapter has done, let's not expect getting a job to be the ultimate outcome. People with IDD, like all people, want to be at a job that they *like*. Many people with IDD want to develop and advance within their careers; they strive for higher wages and more responsibility. Research is beginning to address questions relating to job quality and has demonstrated that career development and advancement are possible for adults with IDD (Heyman, Stokes, & Siperstein, 2016). Other studies have identified links between job satisfaction and work productivity (Hall, Morgan, & Salzberg, 2014) and investigated job choice, job complexity, and salary among workers with ID (Kaye, 2009; McDaniels, 2016). Findings are bleak, as most people with disabilities who are employed are still under the federal poverty line (Nord et al., 2013). However, this body of work is a step in the right direction, as it acknowledges that getting a job is not and should not be the end of the story.

As a final consideration, a complete holistic approach will acknowledge the role of the employers as well. Employers need to recognize that hiring people with disabilities is not an act of charity but a boost to their bottom line. Inclusive hiring improves the public's perceptions of businesses, thereby creating a more robust customer base (Siperstein et al., 2006). As we merge all that we know about skills, programs, and assessment into a holistic approach to increasing the employment rate, let's bring our partners from the business sector on board as well. Through this combined effort, we will achieve the change that we have worked toward for decades.

References

Akkerman, A., Kef, S., & Meininger, H. P. (2018). Job satisfaction of people with intellectual disabilities: The role of basic psychological need fulfillment and workplace participation. *Disability and Rehabilitation, 40*(10), 1192–1199. https://doi.org/10.1080/09638288.2017.1294205.

American Association on Intellectual and Developmental Disabilities. (2018a). *Definition of intellectual disability*. Retrieved from http://aaidd.org/intellectual-disability/definition#.W0fG6tVKiCh

American Association on Intellectual and Developmental Disabilities. (2018b). *Self-determination (Joint Position Statement of AAIDD and The Arc).* Washington, DC: AAIDD.

American Psychiatric Association. (2013). *Diagnostic and statistical manual of mental disorders* (5th ed.). Washington, DC: Author.

Arias, B., Verdugo, M. A., Navas, P., & Gómez, L. E. (2013). Factor structure of the construct of adaptive behavior in children with and without intellectual disability. *International Journal of Clinical and Health Psychology, 13*, 155–166.

Bates, P., & Pancsofar, E. (1983). Project EARN: A competitive employment training program for severely disabled youth in the public schools. *British Journal of Mental Subnormality, 29*(57), 97–103. https://doi.org/10.1179/bjms.1983.015.

Bennett, G. K. (1972). Hand-tool dexterity test. In O. K. Buros (Ed.), *The seventh mental measurements yearbook* (Vol. II). Lincoln, NE: University of Nebraska Press.

Benz, M. R., Lindstrom, L., & Yavanoff, P. (2000). Improving graduation and employment outcomes of students with disabilities: Predictive factors and student perspectives. *Exceptional Children, 66*(4), 509–529.

Berg, L. A., Jirikowic, T., Haerling, K., & MacDonald, G. (2017). Navigating the hidden curriculum of higher education for postsecondary students with intellectual disabilities. *American Journal of Occupational Therapy, 71*, 1–9. https://doi.org/10.5014/ajot.2017.024703.

Biggs, E. E., & Carter, E. W. (2016). Quality of life for transition-age youth with autism or intellectual disability. *Journal of Autism and Developmental Disorders, 46*(1), 190–204. https://doi.org/10.1007/s10803-015-2563-x.

Bouck, E. C. (2010). Reports of life skills training for students with intellectual disabilities in and out of school. *Journal of Intellectual Disability Research, 54*(12), 1093–1103. https://doi.org/10.1111/j.1365-2788.2010.01339.x.

Bruininks, R. H., Woodcock, R. W., Weatherman, R. F., & Hill, B. K. (1996). *Scales of independent behavior – revised comprehensive manual*. Itasca, IL: Riverside Publishing.

Burge, P., Ouellette-Kuntz, H., & Lysaght, R. (2007). Public views on employment of people with intellectual disabilities. *Journal of Vocational Rehabilitation, 29*, 29–37.

Butler, L. N., Sheppard-Jones, K., Whaley, B., Harrison, B., & Osness, M. (2016). Does participation in higher education make a difference in life outcomes for students with intellectual disability? *Journal of Vocational Rehabilitation, 44*, 295–298. https://doi.org/10.3233/JVR-160804.

Butterworth, J., & Strauch, J. D. (1994). The relationship between social competence and success in the competitive workplace for persons with mental retardation. *Education and Training in Mental Retardation and Developmental Disabilities, 29*(2), 118–133.

Carter, E. W., Austin, D., & Trainor, A. A. (2011). Factors associated with the early work experiences of adolescents with severe disabilities. *Intellectual and Developmental Disabilities, 49*(4), 233–257. https://doi.org/10.1352/1934-9556-49.4.233.

Carter, E. W., Austin, D., & Trainor, A. A. (2012). Predictors of postschool employment outcomes for young adults with severe disabilities. *Journal of Disability Policy Studies, 23*(1), 50–63. https://doi.org/10.1177/10442073.11414680.

Cohen, J. S., & Williams, C. E. (1961). A five phase vocational training program in a residential school. *American Journal of Mental Deficiency, 66*, 230–237.

Corlett, E. N., Salvendy, G., & Seymour, W. D. (1971). Selecting operators for fine manual tasks: A study of the O'Connor finger dexterity test and the Purdue pegboard. *Occupational Psychology, 45*(1), 57–65.

Drewes, D. W. (1961). Development and validation of synthetic dexterity tests based on elemental motion analysis. *Journal of Applied Psychology, 45*(3), 179–185.

Fornes, S., Rocco, T. S., & Rosenberg, H. (2008). Improving outcomes for workers with mental retardation. *Human Resources Development Quarterly, 19*(4), 373–395. https://doi.org/10.1002/hrdq.1246.

Foss, G., & Peterson, S. L. (1981). Social-interpersonal skills relevant to job tenure for mentally retarded adults. *Mental Retardation, 19*(3), 103–106.

Gilson, C. B., & Carter, E. W. (2016). Promoting social interactions and job independence for college students with autism or intellectual disability: A pilot study. *Journal of Autism and Developmental Disorders, 46*(11), 3583–3596. https://doi.org/10.1007/s10803-016-2894-2.

Gilson, C. B., Carter, E. W., & Biggs, E. E. (2017). Systematic review of instructional methods to teach employment skills to secondary students with intellectual and developmental disabilities. *Research and Practice for Persons with Severe Disabilities, 42*(2), 89–107. https://doi.org/10.1177/1540796917698831.

Gomes-Machado, M. L., Santos, F. H., Schoen, T., & Chiari, B. (2016). Effects of vocational training on a group of people with intellectual disabilities. *Journal of Policy and Practice in Intellectual Disabilities, 13*(1), 33–40. https://doi.org/10.1111/jppi.12144.

Green, J. C., Cleary, D. S., & Cannella-Malone, H. I. (2017). A model for enhancing employment outcomes through postsecondary education. *Journal of Vocational Rehabilitation, 46*, 287–291. https://doi.org/10.3233/JVR-170863.

Gresham, F., & Elliott, S. N. (2008). *Social skills improvement system (SSIS) rating scales*. Minneapolis, MN: NCS Pearson, Inc.

Grigal, M., Hart, D., & Weir, C. (2012). A survey of postsecondary education programs for students with intellectual disabilities in the United States. *Journal of Policy and Practice in Intellectual Disabilities, 9*(4), 223–233.

Hall, J., Morgan, R. L., & Salzberg, C. L. (2014). Job-preference and job-matching assessment results and their association with job performance and satisfaction among young adults with developmental disabilities. *Education and Training in Autism and Developmental Disabilities, 49*(2), 301–312.

Harrison, P., & Oakland, T. (2015). *Adaptive behavior assessment system, third edition (ABAS-3)*. Minneapolis, MN: NCS Pearson, Inc.

Hemmeter, J., Donovan, M., Cobb, J., & Asbury, T. (2015). Long term earnings and disability program participation outcomes of the bridges transition program. *Journal of Vocational Rehabilitation, 42*(1), 1–15.

Heyman, M., Stokes, J. E., & Siperstein, G. N. (2016). Not all jobs are the same: Predictors of job quality for adults with intellectual disabilities. *Journal of Vocational Rehabilitation, 44*, 299–306. https://doi.org/10.3233/JVR-160800.

Human Resources Center. (1967). *Employment of the mentally retarded in a competitive industrial setting*. Albertson, Long Island, NY: Author.

Kaye, H. S. (2009). Stuck at the bottom rung: Occupational characteristics of workers with disabilities. *Journal of Occupational Rehabilitation, 19*(2), 115–128. https://doi.org/10.1007/s10923-009-9175-2.

Kelley, K. R., & Buchanan, S. K. (2017). College to career ready: Innovative practices that lead to integrated employment. *Journal of Vocational Rehabilitation, 46*, 327–332. https://doi.org/10.3233/JVR-170869.

Kober, R., & Eggleton, I. (2005). The effect of different types of employment on quality of life. *Journal of Intellectual Disability Research, 49*(10), 756–760. https://doi.org/10.1111/j.1365-2788.2005.00746.x.

Lambert, N., Nihira, K., & Leland, H. (1993). *Adaptive behavior scales-school: Second edition. ABS-S.2*. Austin: PRO-ED, American Association on Mental Retardation.

Leffert, J., & Siperstein, G. N. (2003). Social skills instruction for students with learning disabilities. *Current Practice Alerts, 9*, 1–4.

Lemaire, G. S., & Mallik, K. (2008). Barriers to supported employment for persons with developmental disabilities. *Archives of Psychiatric Nursing, 22*(3), 147–155. https://doi.org/10.1016/j.apnu.2007.06.014.

Luckasson, R., Borthwick-Duffy, S., Buntinx, W. H. E., Coulter, D. L., Craig, E. M., Reeve, A., et al. (2002). *Mental retardation: Definition, classification, and systems of supports* (10th ed.). Washington, DC: American Association on Mental Retardation.

Malgady, R. G., Barcher, P. R., Towner, G., & Davis, J. (1979). Language factors in vocational evaluation of mentally retarded workers. *American Journal of Mental Deficiency, 85*(5), 432–438.

Mank, D., Cioffi, A., & Yovanoff, P. (1998). Employment outcomes for people with severe disabilities: Opportunities for improvement. *Mental Retardation, 36*(3), 205–216.

Marston, D. C. (2015). Behavioral assessment and vocational training programs for individuals with intellectual disability. In *Way leads on to way: Paths to employment for people with intellectual disability* (pp. 107–122). Washington, DC: AAIDD.

McCartney, L. D. (1958). Providing occupational readiness for young mentally deficient children of the nonfamilial type. *American Journal of Mental Deficiency, 62*, 625–633.

McConaughy, E. K., Stowitschek, J. J., Salzberg, C. L., & Peatross, D. K. (1989). Work supervisors' ratings of social behaviors related to employment success. *Rehabilitation Psychology, 34*(1), 3–15.

McDaniels, B. (2016). Disproportionate opportunities: Fostering vocational choice for individuals with intellectual disabilities. *Journal of Vocational Rehabilitation, 45*, 19–25. https://doi.org/10.3233/JVR-160807.

McDermott, S., Martin, M., & Butkus, S. (1999). What individual, provider, and community characteristics predict employment of individuals with mental retardation? *American Journal on Mental Retardation, 104*(4), 346–355. https://doi.org/10.1352/0895-8017(1999)104.

Mihailidis, A., Melonis, M., Keyfitz, R., Lanning, M., Van Vuuren, S., & Bodine, C. (2016). A nonlinear contextually aware prompting system (N-CAPS) to assist workers with intellectual and developmental disabilities to perform factory assembly tasks: System overview and pilot testing. *Disability and Rehabilitation: Assistive Technology, 11*(7), 604–612.

Moore, C. L., Feist-Price, S., & Alston, R. J. (2002). Competitive employment and mental retardation: Interplay among gender, race, secondary psychiatric disability, and rehabilitation services. *Journal of Rehabilitation, 68*(1), 14–19.

Moore, E. J., & Schelling, A. (2015). Postsecondary inclusion for individuals with an intellectual disability and its effects on employment. *Journal of Intellectual Disabilities, 19*(2), 130–148. https://doi.org/10.1177/1744629514564448.

Moran, R. R., McDermott, S., & Butkus, S. (2001). Getting a job, sustaining a job, and losing ajob for individuals with mental retardation. *Journal of Vocational Rehabilitation, 16*, 234–244.

Murphy, G. (2009). Challenging behavior: A barrier to inclusion? *Journal of Policy and Practice in Intellectual Disabilities, 6*(2), 89–90.

Murray, C., & Doren, B. (2013). The effects of working at gaining employment skills on the social and vocational skills of adolescents with disabilities: A school-based intervention. *Rehabilitation Counseling Bulletin, 56*(2), 96–107.

Neeman, M. (1986). The Purdue pegboard: Its predictive validity for work potential of persons with mental retardation. *American Journal of Occupational Therapy, 40*(6), 433–434.

Noel, V. A., Oulvey, E., Drake, R. E., & Bond, G. R. (2017). Barriers to employment for transition-age youth with developmental and psychiatric disabilities. *Administration and Policy in Mental Health, 44*(3), 354–358. https://doi.org/10.1007/s10488-016-0773-y.

Nord, D., Luecking, R., Mank, D., Kiernan, W., & Wray, C. (2013). The state of the science of employment and economic self-sufficiency for people with intellectual and developmental disabilities. *Intellectual and Developmental Disabilities, 51*(5), 376–384. https://doi.org/10.1352/1934-9556-51.5.376.

O'Connor, J. (1965). O'Connor finger dexterity test. In O. K. Buros (Ed.), *The sixth mental measurements yearbook*. Lincoln, NE: University of Nebraska Press.

Obama, B. H. (2013, July). Proclamation 8999-Anniversary of the Americans with Disabilities Act, 2013. In *Compilation of presidential documents*. Washington, DC: Office of the Federal Register, National Archives and Records Administration.

Park, E., Kim, J., & Kim, S. (2016). Meta-analysis of the effect of job-related social skill training for secondary students with disabilities. *Journal of Vocational Rehabilitation, 44*, 123–133. https://doi.org/10.3233/JVR-150785.

Petcu, S., Chezan, L. C., & Van Horn, M. L. (2015). Employment support services for students with intellectual and developmental disabilities attending postsecondary education programs. *Journal of Postsecondary Education and Disability, 28*(3), 359–374.

Plotner, A. J., & Marshall, K. J. (2016). Supporting postsecondary education programs for individuals with an intellectual disability: Role of the vocational rehabilitation counselor. *Rehabilitation Counseling Bulletin, 59*(4), 195–202. https://doi.org/10.1177/0034355215583056.

Poppen, M., Lindstrom, L., Unruh, D., Khurana, A., & Bullis, M. (2017). Preparing youth with disabilities for employment: An analysis of vocational rehabilitation case services data. *Journal of Vocational Rehabilitation, 46*(2), 209–224.

Posner, B. (1966). Five days as a retarded laundry worker. *Rehabilitation Record, 7*, 1–5.

Price, R., Marsh, A. J., & Fisher, M. H. (2017). Teaching young adults with intellectual and developmental disabilities community-based navigation skills to take public transportation. *Behavior Analysis in Practice, 11*(1), 46–50. https://doi.org/10.1007/s40614-017-0202-z.

Schur, L. (2002). The difference a job makes: The effects of employment among people with disabilities. *Journal of Economic Issues, 36*(2), 339–347.

Simonsen, M. L., & Neubert, D. A. (2013). Transitioning youth with intellectual and other developmental disabilities: Predicting community employment outcomes. *Career Development and Transition for Exceptional Individuals, 36*(3), 188–198. https://doi.org/10.1177/2165143412469399.

Siperstein, G. N., Heyman, M., & Stokes, J. E. (2014). Pathways to employment: A national survey of adults with intellectual disabilities. *Journal of Vocational Rehabilitation, 41*, 165–178. https://doi.org/10.3233/JVR-140711.

Siperstein, G. N., Parker, R. C., & Drascher, M. (2013). National snapshot of adults with intellectual disabilities in the labor force. *Journal of Vocational Rehabilitation, 39*, 157–165.

Siperstein, G. N., Romano, N., Mohler, A., & Parker, R. (2006). A national study of consumer attitudes towards companies that hire people with disabilities. *Journal of Vocational Rehabilitation, 24*, 3–9.

Sparrow, S. S., Cicchetti, D., & Saulnier, C. A. (2005). *Vineland adaptive behavior scales, third edition (Vineland-3)*. Minneapolis, MN: NCS Pearson, Inc.

Spence, M. (2004). Successful vocational rehabilitation for individuals with significant mental disabilities. *Journal of Social Work in Disability & Rehabilitation, 3*(1), 37–52.

Stephens, D. L., Collins, M. D., & Dodder, R. A. (2005). A longitudinal study of employment and skill acquisition among individuals with developmental disabilities. *Research in Developmental Disabilities, 26*(5), 469–486. https://doi.org/10.1016/j.ridd/200.12.003.

Tobias, J., & Gorelick, J. (1960). The effectiveness of the Purdue pegboard in evaluating work potential of retarded adults. *Training School Bulletin, 57*, 94–104.

Tomaszewski, B., Fidler, D., Talapatra, D., & Riley, K. (2018). Adaptive behavior, executive function and employment in adults with Down syndrome. *Journal of Intellectual Disability Research, 62*(1), 41–52.

United States Department of Labor. (2014). Employment first. Retrieved from https://www.dol.gov/odep/topics/EmploymentFirst.htm

Wakabayashi, I. (2009). How has applied behavioral analysis contributed to vocational interventions for people with developmental disabilities? A review of U.S. literature. *Japanese Journal of Behavior Analysis, 23*(1), 5–32.

Wehmeyer, M. L. (1996). Student self-report measure of self-determination for students with cognitive disabilities. *Education and Training in Mental Retardation and Developmental Disabilities, 31*(4), 282–293.

White, D. A., & Dodder, R. A. (2000). The relationship of adaptive and maladaptive behavior to social outcomes for individuals with developmental disabilities. *Disability and Society, 15*(6), 897–908.

Widaman, K. F., & McGrew, K. S. (1996). The structure of adaptive behavior. In J. W. Jacobson & J. S. Mulick (Eds.), *Manual of diagnosis and professional practice in mental retardation* (pp. 97–110). Washington, DC: American Psychological Association.

Neuropsychology

26

Graham M. L. Eglit

Abnormalities in brain structure and/or function are common in developmental, psychiatric, and neurological disorders, including intellectual disabilities. These abnormalities are central to defects in cognitive processing in these conditions, which, in concert with other factors, impact an individual's academic achievement, functional independence, and vocational outcome (Yeates, Ris, Taylor, & Pennington, 2009). A central focus in clinical neuropsychology is to identify and describe the fundamental cognitive processing deficits in brain-based disorders and to explain these deficits in terms of brain structure and function (Lezak, Howieson, Bigler, & Tranel, 2012). As such, neuropsychology is essential to the understanding of intellectual disabilities, and neuropsychological assessment is a key component of patient care (Yeates et al., 2009).

This chapter will discuss defining features of neuropsychology, assumptions made by neuropsychology, essential methods in neuropsychological research, approaches to clinical neuropsychological assessment, and core domains of cognitive functioning assessed in neuropsychology. Finally, this chapter will discuss emerging research areas in neuropsychology that have great potential to advance our understanding of brain-behavior relationships in normal and clinical populations.

What Is Neuropsychology?

Neuropsychology can be defined as the study of brain-behavior relationships (Kolb & Whishaw, 2009). Behavior, in this definition, can be understood in a broad sense to include cognition, emotion, and behavioral responses; however, traditionally, cognitive processing has been the main focus of neuropsychology (Ogden, 2005). The central aim of neuropsychology is to develop a science of cognition, emotion, and behavior that explains these three phenomena in terms of the function of the human brain (Kolb & Whishaw, 2009). As the name implies, neuropsychology is an inherently trans-disciplinary field, drawing from a diverse set of research areas, including anatomy, biology, biophysics, ethology, pharmacology, physiology, physiological psychology, neurosurgery, psychometrics, cognitive psychology, and philosophy, among others (Kolb & Whishaw, 2009).

Neuropsychology itself is composed of two distinct but partially overlapping sub-disciplines. Cognitive neuropsychology attempts to understand and characterize normal cognitive function through the study of brain-damaged patients. Cognitive neuropsychology is often less inter-

---

G. M. L. Eglit (✉)
Department of Psychiatry, University of California
San Diego, La Jolla, CA, USA

Sam and Rose Stein Institute for Research on Aging,
University of California San Diego,
La Jolla, CA, USA

J. L. Matson (ed.), *Handbook of Intellectual Disabilities*, Autism and Child Psychopathology
Series, https://doi.org/10.1007/978-3-030-20843-1_26

ested in brain localization of cognitive processes and instead emphasizes the implications of brain damage-induced cognitive changes for theories of normal cognitive information processing (Ward, 2006). In contrast, clinical neuropsychology places greater emphasis on the behavioral expression of central nervous system dysfunction (Lezak et al., 2012). Accordingly, the focus in clinical neuropsychology is on brain pathology and the associated signs and symptoms of this pathology. This chapter principally focuses on clinical neuropsychology. It should be noted, however, that the research questions, methods, and findings of these two sub-disciplines overlap to a significant extent.

Although described as a clinical discipline, clinical neuropsychology has both research and applied clinical branches. Clinical neuropsychological research is aimed at characterizing cognitive phenotypes, identifying cognitive markers of disease states, describing longitudinal trajectories of cognitive deficits, and developing measures to facilitate diagnosis, cognitive profile characterization, and evaluation of response to intervention. As a clinical practice, neuropsychology attempts to infer brain functioning and characterize lesion location, clarify cognitive strengths and weaknesses, predict functional difficulties, contribute to the determination of legal competency, and inform intervention targets and measure intervention outcomes (Heaton & Marcotte, 2000).

Foundational Assumptions of Neuropsychology

An important aspect of any discipline is the assumptions made by practitioners within that discipline. The discipline of neuropsychology rests on three key assumptions, which serve as the preconditions for investigation. The first of these assumptions is the brain hypothesis. The brain hypothesis maintains that the brain is the source of behavior (Kolb & Whishaw, 2009). This assumption commits neuropsychology to materialism, which maintains that behavior can be explained by the complex interaction of physical entities and events. However, neuropsychology typically does not embrace a reductive materialism, in which behavior, such as memory, can be reduced to individual neurons. Instead, it is more commonly assumed that behavior is an emergent property of the complex interactions among numerous neurons and neuronal networks (Pennington, 2009). Moreover, the influence between the brain and behavior is typically assumed to be bidirectional. In this sense, the brain is both the source of behavior and in turn is shaped and molded by one's learning history and experience. There is now a wealth of evidence demonstrating synaptic plasticity-driven changes in neural wiring due to unique experience histories, such as those found among musicians, blind readers of Braille, and the congenitally deaf, among others (Galaburda & Pascual-Leone, 2003).

The second assumption provides more specificity to the brain hypothesis. This second assumption is the cerebral localization hypothesis (also termed domain specificity), which maintains that specific cognitive functions can be localized to regions or circuits within the brain. The cerebral localization hypothesis consists of three key claims: (1) the brain has a modular organization; (2) each module is specialized for a particular cognitive process; and (3) these modules can be reliably localized to specific brain regions (Greiffenstein, 2014). Notably, cerebral localization does not entail that these brain-behavior modules are isolated and independent of one another. Instead, efficient performance of a behavior, such as speech production, requires the orderly integration of multiple modules. In addition, many of these modules are localized to distinct regions of the brain, with information processed both simultaneously and sequentially in these modules. Taken together, a more nuanced view of cerebral localization entails not only that cognitive functions are localized but also that they are distributed in multiple regions of the brain, operate in parallel to one another, and are integrated in a hierarchical fashion, with more complex modules built on simpler ones (Kolb & Whishaw, 2009).

Finally, the third assumption of neuropsychology is the psychological or cognitive assumption. This assumption holds that the use of psychological or cognitive constructs to explain normal or abnormal behavior is more parsimonious than a wholesale inventory of correlated behavioral abnormalities or a reduction of behavior to neurophysiology (Morton & Frith, 1995; Pennington, 2009). While some may interpret this assumption as maintaining that single cognitive deficits underlie specific disorders, such as phonological processing for dyslexia or theory of mind for autism, a more nuanced and scientifically plausible understanding of this assumption is that these disorders are characterized by multiple cognitive deficits acting in combination with one another. A key task of neuropsychology is to identify these fundamental cognitive constructs and explain how their interactions explain characteristic behavioral abnormalities within specific disorders (Pennington, 2006).

Clinical Neuropsychology: The Key Role of Psychometrics

One of the defining features of clinical neuropsychology is the emphasis placed on strong psychometric theory to inform clinical assessment. In fact, this focus on psychometrics distinguishes clinical neuropsychology from the closely related discipline of behavioral neurology. While both clinical neuropsychology and behavioral neurology specialize in the behavioral expression of brain dysfunction, behavioral neurology evaluates and characterizes this behavioral expression in terms of pathological signs, which are either present or absent. In contrast, clinical neuropsychology is firmly grounded in the psychometric tradition of empirical psychology, emphasizing the measurement of continuously distributed behavioral variables with known acceptable reliability and validity (Horton & Puente, 1986; Rourke & Brown, 1986). Thus, psychometrically informed measurement of behavior is an essential feature of clinical neuropsychology.

Several core psychometric considerations in neuropsychology warrant discussion. The first of these considerations is the reliance on continuously distributed variables. Continuously distributed variables are preferred in neuropsychology because their frequency distributions often conform to a normal curve (also known as a normal distribution or bell curve). The normal curve reflects the probability of obtaining a score or range of scores when randomly sampled from the population. This probability is often described in terms of percentile ranks or their associated deviate scores. A percentile rank indicates the percentage of individuals from the population who score at or below a given test score. Deviate scores, which include z, T, scaled, and standard scores, are standardized expressions of raw scores and reflect the number of standard deviation units from which a score differs from the mean of the distribution. Deviate scores thus function to convert raw scores into percentile ranks. Conceptually, deviate scores and percentile ranks are used to characterize the degree of abnormality of an obtained score, with abnormal scores reflecting poor test performance interpreted as indicative of neuropsychological impairment (Slick, Strauss, Sherman, & Spreen, 2006). There is considerable debate over optimal cutoffs for defining abnormality/neuropsychological impairment, but most definitions range from ≥1 standard deviation below the mean (16th percentile or below) to ≥2 standard deviations below the mean (0.4th percentile or below) (Heaton & Marcotte, 2000).

The use of percentile ranks and deviate scores to detect neuropsychological impairment relies on the use of a comparison sample to derive a standard normal distribution. This process is often referred to as norming and the data resulting from this process as normative data. Conceptually, normative data should reflect the distribution of test scores in the general population. Creation of normative data for test score interpretation therefore requires a large (greater than at least 200 cases) sample in order to avoid sampling error and to increase the representativeness of the sample to the general population

(Nunnally & Bernstein, 1994). In addition, the demographic composition of the normative sample should be comparable to the general population. Recognizing that demographic factors, such as age, education, gender, and race/ethnicity, can affect neuropsychological test scores independent of brain dysfunction, demographic corrections are commonly applied to normative data in neuropsychology. In general, there are two main approaches to normative comparisons in clinical neuropsychology: (1) comparing individual test scores to normative data that approximates as closely as possible the general population and (2) comparing individual test scores to normative data that matches as closely as possible the unique subgroup to which the examinee belongs (e.g., non-Hispanic Caucasian males with 14 years of education) (Mitrushina, Boone, & D'Elia, 2005; Strauss, Sherman, & Spreen, 2006). Notably, adaption of either of these approaches may be dictated by the purposes of the evaluation. For instance, in attempting to diagnose intellectual disability or learning disability, comparison to all other persons of the same age in the general population is most appropriate. However, if the purpose of the evaluation is to characterize cognitive strengths and weaknesses to inform diagnostic considerations and/or plan classroom accommodations, comparison to the examinee's unique subgroup may be more apposite (Strauss et al., 2006). Ultimately, availability of appropriate normative data is crucial to rendering informed clinical decisions based on neuropsychological test performance (Heaton & Marcotte, 2000).

In addition to the availability of adequate normative data, the reliability of neuropsychological tests and the validity of the interpretations based on neuropsychological test scores are important psychometric considerations. Reliability reflects the degree to which a test consistently measures a given construct. There are several types of reliability, including internal consistency reliability (which indicates the extent to which items on a test measure the same construct), test-retest reliability (which reflects the temporal stability of a measure), alternate forms reliability (which denotes the similarity of scores across two measures of the same construct), and inter-rater reliability (which indicates the similarity of scores across two raters of the same test). The greater the reliability of a test, the more confidence an examiner can have in the precision of a score based on performance on that test. In contrast, validity refers to the extent to which a test actually measures what it intends to measure. Similar to reliability, there are multiple types of validity evidence that include, at a minimum, content-related validity (i.e., the degree to which the content of the test is relevant, representative, and of high technical quality), construct-related validity (i.e., the extent to which a test measures what it intends or purports to measure), criterion-related validity (i.e., the extent to which a test is related to an external outcome), and response process validity (i.e., the degree to which the response processes implemented by examinees are consistent with measurement on the intended construct) (although for further types of validity information, see American Educational Research Association, 1999; Messick, 1995). Adequate validity is essential to the interpretation of a test score as indicative of a specific construct. A considerable amount of research is dedicated to evaluating the reliability and validity of neuropsychological measures and deriving appropriate normative data in order to facilitate test selection, administration, and interpretation.

Essential Research Methods in Neuropsychology

There are several essential methods used in neuropsychology to identify brain-behavior relationships. Historically, the lesion study has been the most important method to neuropsychology. Lesion studies evaluate the impact of brain damage on patient behavior. These types of studies were central to the development of the cerebral localization hypothesis and remain key in the determination of brain-behavior relationships. The central premise of the lesion study is that the function of a given brain region can be inferred on the basis of impaired and spared abilities resulting from lesions to that area. Lesion studies can be described in terms of single dissociation studies or double dissociation studies. In single dissociation studies, a patient with brain damage exhibits impairment on one task (task A), but a relatively normal performance on a separate task (task B). This type of finding suggests that task A and task B utilize different cognitive processes that rely on separate neural regions. However, a key shortcoming of single dissociation studies is the possibility that tasks A and B require the same cognitive processes/neural regions but that task B requires more of this cognitive process than task A. Conclusions about brain-behavior relationships can be strengthened through demonstration of a double dissociation. In a double dissociation, one patient with brain damage exhibits impairment on task A, but relatively normal performance on task B, while another patient with a lesion in a different brain region is impaired on task B, but normal on task A. Double dissociations provide strong evidence for the distinctness of closely related cognitive processes and their neural correlates (Ward, 2006).

Neuroimaging represents another essential research method in neuropsychology. Neuroimaging can be classified as structural or functional neuroimaging. Structural neuroimaging refers to brain imaging methods that produce static images of the spatial configuration of different types of brain tissue. Obtaining static images of the brain is based on the fact that different types of brain tissue (e.g., the skull, gray matter, white matter, cerebrospinal fluid, etc.) have different physical properties. Correlation of a structural abnormality with specific impairment on neuropsychological testing yields valuable information on the potential causal role of that region to that neuropsychological ability. Structural imaging modalities include computerized tomography and magnetic resonance imaging, which differ in their method of measuring physical properties of different tissue types (Ward, 2006).

In contrast to structural imaging, functional imaging creates dynamic images of changes in physiological characteristics of the brain that may be correlated with task-related cognitive processes. Most functional imaging modalities are based on the fact that additional regional blood flow is needed to supply oxygen and glucose to meet the metabolic demands of carrying out specific cognitive tasks. Comparison of this task-related physiological response to a baseline level of physiological activity during a control condition can thus shed light on brain region recruitment for that cognitive task, which suggests specialization of that region for the specific cognitive function under investigation. Selection of a baseline condition is a complex process, however, and requires isolating the specific cognitive process under investigation from other cognitive processes that may be involved in the task. There are several experimental designs that have been used for this comparison, which include cognitive subtraction, factorial designs, and parametric designs (see Friston, 1997, for a review of these designs and their relative advantages and disadvantages and Kosslyn, 1999, for a discussion of the difficulties of functional neuroimaging interpretation). Positron emission tomography and functional magnetic resonance imaging are the two most common functional imaging modalities (Huettel, Song, & McCarthy, 2014).

Additional methods of neuropsychological investigation include electroencephalography (EEG), single cell recordings, transcranial magnetic stimulation, computational modeling, and animal models; however, discussion of these other methods is beyond the scope of this chapter.

Approaches to Neuropsychological Assessment

A typical assessment in neuropsychology involves the administration of a multitude of cognitive and behavioral tasks. Through integration of the qualitative and quantitative data obtained by these measures, a neuropsychologist can clarify key deficits in cognitive processing, identify syndrome-level patterns of these deficits, and surmise most likely etiologies for these syndromes (Heaton & Marcotte, 2000). Historically, several distinct assessment approaches have been followed to achieve these ends. This section provides a brief description of four of the most common assessment approaches.

Qualitative Approach (Luria)

The qualitative approach to neuropsychological assessment is most often associated with the Russian neuropsychologist A.R. Luria (Glozman, 2007). Luria assumed that mental and behavioral functions were complex constellations of more fundamental component parts and that it was the neuropsychologist's role to ascertain whether and which of these fundamental components was impaired in a patient (Lezak et al., 2012). To write, for instance, involves, at a minimum, skilled movement, spatial organization, word selection, and grapheme sequencing. Impairment in any one of these fundamental cognitive processes can result in a disturbance in writing ability. Moreover, impairment in one of these fundamental abilities is likely to impact performance on other tasks. For instance, impaired word selection may also emerge as word finding problems in spontaneous speech and difficulty identifying the names of pictures and objects. Detection and characterization of neuropsychological impairment for Luria and his followers in the qualitative approach primarily involved assessing qualitative aspects of patient behavior, placing particular emphasis on the nature of deviations or errors, while the use of psychometric instruments was considerably less common. Assessment for Luria was highly individualized, following a hypothesis-driven approach in which the particular behaviors assessed were selected by the neuropsychologist based upon the neuropsychologist's working hypothesis regarding the underlying fundamental deficits from which the patient suffered (Ardila, 1992).

The qualitative approach is not without its critics. For one, several authors have questioned whether this method can be properly classified as clinical neuropsychology, preferring instead to place Luria in the tradition of behavioral neurology. As discussed above, in contrast to clinical neuropsychology, which relies on the measurement of continuously distributed variables within a psychometric tradition, behavioral neurology emphasizes dichotomous classification based on pathological signs (Horton & Puente, 1986; Rourke & Brown, 1986). For another, given the non-standardized nature of the assessment and the lack of emphasis on quantitative measurement, the qualitative approach of Luria has been criticized for being subjective, occasionally obscure and resulting in conclusions of questionable validity (Reitan, 1976). Nonetheless, Luria's emphasis on breaking down complex mental and behavioral functions into component parts and his attention to the nature of errors were highly influential to future neuropsychological developments (Ardila, 1992).

The Fixed Battery Approach (Halstead-Reitan)

In contrast to the qualitative approach, the fixed battery approach involves administration of the same comprehensive set of tests to all examinees, regardless of presenting complaints, purposes of the evaluation, or diagnostic considerations. This approach is most commonly associated with Ward Halstead and Ralph Reitan and their battery of tests, the Halstead-Reitan neuropsychological battery (HRNB), although a number of other fixed batteries exist. Historically, the HRNB emerged from the recognition that no single neuropsychological test could accurately detect and characterize the effects of brain damage. Instead, Halstead and subsequently Reitan noted that there was considerable heterogeneity in presentation, functional ability, and test performance among individuals with brain damage (Hom & Nici, 2015). Halstead initially developed a small battery of ten tests designed to measure the complex operations of higher-level mental functions of the central nervous system. Reitan further evaluated these ten tests, as well as dozens of others, to empirically determine their ability to reliably detect the presence of brain damage, lateralization of brain damage, recovery potential, and type of lesion or disease. Tests that contributed to the accurate detection and localization of brain lesions were retained and compiled to create the Halstead-Reitan neuropsychological battery (Goldstein, 2015; Hom & Nici, 2015; Reitan & Wolfson, 2009).

The fixed battery approach has both advantages and disadvantages. In terms of advantages, fixed batteries tend to yield more information on the performance of specific tests across a wide range of clinical populations. As a result, there is often a greater wealth of empirical research to ground interpretation of test scores using fixed batteries, including better norms, greater knowledge of the psychometric properties of tests, and greater knowledge of how to integrate test data across the battery (Heaton, Grant, Anthony, & Lehman, 1981; Reitan, 1964). On the other hand, however, fixed batteries can be unnecessarily lengthy and expensive and cause examinee stress and/or exhaustion due to over testing. In addition, fixed batteries are more likely to become outdated over time as the field develops and may inadequately assess certain cognitive domains, thus requiring administration of supplemental tests (Heaton & Marcotte, 2000). Nonetheless, the fixed battery approach is common in research contexts where a premium is placed on the use of a uniform set of tests and procedures so as to facilitate comparison across study participants.

The Flexible Battery Approach (Iowa-Benton)

The flexible battery approach involves the administration of a tailor-made set of tests to assess patients in a highly individualized manner, with test selection guided by the neuropsychologist's working hypothesis about the condition(s) the examinee is known or suspected to have. This approach is most typically associated with the work of Arthur Benton at the University of Iowa. The development of this approach was based on observation of clinical examinations conducted by psychiatrists and neurologists, which were noted to be highly flexible and efficient. These examinations began with a broad mental status exam, from which the clinician pursued diagnostic possibilities through diverse questions. Exams could last from anywhere between 15 min to over an hour, depending on the degree of diagnostic uncertainty involved in the exam. Benton was impressed by the flexibility and hypothesis-driven nature of these examinations and believed that neuropsychological assessment should follow this model. In this view, neuropsychological assessment should consist of standardized objective tests, similar to Halstead-Reitan, but these tests should be implemented in a highly flexible, hypothesis-testing-driven manner, more akin to Luria (Tranel, 2009).

Benton's approach to assessment consisted of a small, core battery of between two and six standard tests administered to every patient. Then, based on the referral question and the patient's performance on the core battery, additional tests were administered to explore diagnostic possibilities. This approach might be better conceptualized as a fixed-flexible approach, given that a standard set of batteries was always administered and a large set of additional measures were implemented to pursue the remaining diagnostic hypotheses. More recently, the Iowa-Benton school has undergone subtle revision in its approach. In particular, the core battery has expanded in length, and additional information has been incorporated to guide the flexible, hypothesis-testing-driven component of the assessment, including the patient's complaints, impressions gained from an initial clinical interview, the patient's medical history, neurological findings, and neuroimaging data (Tranel, 2009).

The flexible and fixed-flexible battery approach has much to recommend it. These approaches avoid unnecessary testing and allow the neuropsychologist to probe more precisely into the nature of a patient's deficits. However, there is often less psychometric and clinical information available on how tests should be interpreted in conjunction with one another. In addition, flexible and fixed-flexible battery approaches tend to be more reliant on patient- and informant-report of cognitive symptoms to guide test selection, which may lead to imprudent test selection if patient and/or informants lack accurate knowledge of cognitive deficits (Heaton & Marcotte, 2000).

Boston Process Approach (Kaplan)

The final approach to neuropsychological assessment reviewed in this chapter is the Boston Process Approach, which is most commonly associated with the work of Edith Kaplan. At its core, the Boston Process Approach has three main aims: (1) to understand the qualitative nature of behavior assessed by clinical psychometric instruments, (2) to reconcile descriptive richness with reliable and valid assessment, and (3) to incorporate recent insights from cognitive neuroscience and experimental psychology into clinical neuropsychological assessment (Delis, Kramer, Fridland, & Kaplan, 1990). This approach stemmed from the realization that there was considerable variability in the manner in which patients exhibited the loss of one and the same specific cognitive function and that this variability often correlated with lesion size and location. For instance, performance on Wechsler Adult Intelligence Scale-IV Block Design test reliably differs among individuals with left vs. right hemisphere lesions. Patients with right hemisphere lesions are more likely to exhibit configural processing errors, in which they fail to retain the 2 × 2 or 3 × 3 configural structure of produced block constructions. In contrast, patients with left hemisphere lesions are more likely to produce internal block rotation errors while retaining the configural structure of the design. This pattern of performance is consistent with accounts of hemispheric specialization, in which the right hemisphere is dominant for global processing and the left hemisphere for local processing (Delis, Kiefner, & Fridlund, 1988; Delis, Robertson, & Efron, 1986; Robertson & Delis, 1986). Thus, in the Boston Process Approach, the emphasis is placed on the manner in which a patient solves, or fails to solve, items on a neuropsychological test, rather than just their final quantitative score on that test. Over the past two decades, there have been a growing number of tests and methods developed to provide standardized ways of scoring these process outcomes, such as those implemented in the California Verbal Learning Test-II (Delis, Kramer, Kaplan, & Ober, 2000). The Boston Process Approach is most typically associated with a flexible assessment approach, although it can be used with a fixed battery as well.

Core Domains of Neurocognition in Neuropsychology

Neuropsychological assessment involves the administration of a battery of cognitive tasks across several domains of cognition. Assessment with numerous tasks across domains is necessary because neuropsychological tasks are multi-determined, such that poor performance on any given task can be the result of several cognitive and/or non-cognitive factors. To tease apart these potential contributing factors and identify underlying spared and impaired abilities and processes requires the evaluation of the pattern of performance across tasks. In addition to the evaluation of cognition, neuropsychological assessment should include, at a minimum, a global measure of intellectual functioning; an estimate of adaptive, social, behavioral, and emotional functioning; and evaluation of academic achievement in reading and math. As other chapters in this handbook are more specifically devoted to assessment of intelligence (Chaps. 2 and 22), adaptive behavior (Chap. 23), and emotional functioning (Chaps. 8, 24, 47, 48, and 49), this section will cover issues specific to the assessment of core cognitive domains in neuropsychology. Core cognitive domains include attention, memory, executive functions, language, and visuospatial ability.

Attention

Attention refers to a set of processes that enable efficient allocation of cognitive resources to the task at hand (Cohen, Malloy, Jenkins, & Paul, 2014). Unlike other cognitive domains, attention is not a fundamental substrate of cognition, but instead governs the flow and processing of information within other cognitive domains, such as memory, language, and visuospatial abilities (Cohen et al., 2014). This ability is achieved through at least four distinct attentional processes

(Cohen et al., 2014; Mirsky, Anthony, Duncan, Ahearn, & Kellam, 1991).

The first of these processes is sensory selective attention. As the name implies, sensory selective attention involves the selection of sensory input for additional cognitive processing. This selection process occurs outside of conscious awareness and at a very early stage of information processing. In order to successfully selectively attend to sensory stimuli, three subprocesses must be integrated: the ability to filter out irrelevant information, the ability to enhance the processing of relevant/filtered information, and the ability to remain engaged on a stimulus or to disengage from a stimulus and selectively orient/attend to a new stimulus.

The second attentional process is referred to as intention. Intention is important for the selection and control of behavioral responses. Unlike sensory selective attention, intention is a controlled, effortful process that requires conscious awareness. Given its controlled, effortful nature, intention partially overlaps with aspects of executive functioning (see the executive function section below for further detail).

Third, attention is constrained by an individual's attentional capacity. Attentional capacity refers to the amount of information to which an individual can allocate additional cognitive resources at any one time. Attentional capacity is impacted by state factors, such as fatigue, and trait factors, including neural transmission and processing speed, working memory capacity, and spatial and temporal processing constraints.

The fourth core attentional process is sustained attention. Sustained attention refers to the ability to maintain levels of attention across time. Individuals with sustained attention difficulties will often exhibit variability in performance over extended periods of time, often due to loss of interest and/or susceptibility to distraction.

The neuroanatomy of attention is complex and cannot be localized to a single brain system. Instead, multiple brain systems interact as a coherent network to control attention. These regions include the inferior parietal cortex, frontal cortex, limbic system, thalamus, basal ganglia, and the midbrain (Cohen et al., 2014).

Neuropsychological assessment of attention is challenging for a number of reasons. For one, as mentioned above, it is not a fundamental cognitive substrate, but rather functions to enhance other cognitive processes. As such, pure measures of attention do not exist. For another, attention by definition varies over time and may be situation specific. Thus, attention is usually assessed on tasks that load on more than one cognitive domain and may be best evaluated by examining performance across tasks that differ with respect to attentional parameters (e.g., attentional capacity load, time on task, etc.). For sensory selective attention, key tests include letter and symbol cancellation tasks, which involve searching an array of stimuli to identify targets from distractors (Lezak et al., 2012). Intention, on the other hand, is often assessed qualitatively through tasks such as double alternating movements, alternating graphic sequences (e.g., Rampart figures), and motor impersistence (Royall, Mahurin, & Gray, 1992). Attentional capacity is most often measured with the digits forward subtest of, for instance, the Wechsler Adult Intelligence Scale-IV (Wechsler, 2008). More complex tasks of attentional capacity that also require focus typically involve varying both the amount of information attended to and the complexity of effortful processing necessary to perform some operation on that information. A popular measure of attentional capacity and focus is the paced auditory serial addition test (PASAT; Gronwall, 1977). Finally, tests of sustained attention evaluate the temporal characteristics of performance on tasks that involve detecting targets among distractors. The most common of these measures are continuous performance tests (CPT), such as the Connors CPT-II (Connors, 2000).

Memory

Memory is a remarkably complex cognitive domain composed of several distinct memory systems. These memory systems differ in both their time course and the type of information for which they are specialized. Regarding time course, there are three distinct stages of memory.

The first of these stages is the sensory storage (or sensory registration). This stage refers to the point in time in which sensory information (e.g., visual, auditory, gustatory, etc.) is initially registered. This stage is very short in duration, lasting milliseconds to seconds. If no additional processing is conducted, this sensory information quickly dissipates. The second stage of memory processing is referred to as short-term or working memory. Short-term memory has a duration of up to approximately 30 s. In addition, short-term memory has a limited storage capacity in which individuals can typically only maintain 7 ± 2 distinct items (e.g., to-be-remembered words). Unless further processing is applied to items in short-term memory, such as rehearsal or organization, these items will be forgotten. Information that has been sufficiently processed in short-term memory will be transferred into the third and final stage of memory, long-term memory. Long-term memory is relatively permanent and with unlimited capacity (Scott & Schoenberg, 2011d).

There are several different types of long-term memory, which differ in the nature of the information for which they are specialized. Squire and Zola (1996) proposed the most influential taxonomy of long-term memory. In their model, memory can be divided into declarative and non-declarative types. Declarative (explicit) memory refers to long-term memories that can be retrieved and reflected on consciously, including memory for names, people, places, events, and facts. Within declarative memory, there are two further subtypes. The first subtype of declarative memory is semantic memory. Semantic memory is memory for facts and general world knowledge, such as the name of 17th President of the United States and the day of the year on which Christmas falls. This is in contrast to episodic memory, which refers to memory for personally experienced and remembered events, such as what you ate for breakfast today. As opposed to semantic memory, episodic memory requires the active recall of the learning event. Episodic memory is the more important type of memory in neuropsychological assessment (Scott & Schoenberg, 2011d). Episodic memory can be further characterized by whether it involves verbal or visual material, whether it is learned intentionally or incidentally, and whether the episodic memory is for a recent or remote event. In addition, episodic memory can be subdivided into three subprocesses, encoding, consolidation, and retrieval. Encoding refers to the process of learning material, consolidation refers to the process of transferring new material from short- to long-term memory, and retrieval is the process of activating information from long-term memory back to short-term memory for conscious recollection (Scott & Schoenberg, 2011d).

In contrast, non-declarative (implicit) memory refers to information that can influence thought and behavior without conscious awareness. Non-declarative memory includes memory for skills, priming, classical conditioning, and nonassociative learning. Non-declarative memory is seldom evaluated in neuropsychological assessment (Squire & Zola, 1996).

Given the complexity of long-term memory, it is perhaps unsurprising that the functional neuroanatomy of this cognitive domain is similarly complex. Episodic memory is subtended by two functional neurocircuits, a medial circuit (often referred to as the Papez circuit) involving the hippocampus, fornix, mammillary bodies, anterior medial nucleus of the thalamus, and the cingulate, and a lateral circuit (often referred to as the amygdaloid circuit) involving the amygdala, thalamic nuclei, orbitofrontal cortex, piriform cortex, insula, hypothalamus, limbic striatum, and nucleus basalis of Meynert (Mishkin, 1982). Amnestic syndromes are most commonly associated with damage to three distinct regions. First, damage to the medial temporal lobe (hippocampus, parahippocampal gyrus, and entorhinal cortex) often leads to an anterograde amnesia characterized by preserved insight into memory difficulties, increased rate of forgetting, limited retrograde amnesia, and lack of confabulation. Second, patients with damage to the diencephalic regions, especially the medial thalamus region and the mammillary bodies, exhibit anterograde amnesia, but with normal rates of forgetting. Due to disruption of frontal areas, patients with damage to this are may also present with limited insight and a tendency to confabulate. Third, damage to the basal forebrain region results in profound attention difficulties that adversely impacts encoding and retrieval processes, but may leave consolidation intact. Patients with damage to this area have poor insight into their memory difficulties and demonstrate a tendency to confabulate (O'Connor & Race, 2013).

Multiple aspects of episodic memory should be evaluated in order to comprehensively characterize this domain. In particular, memory testing should include measures of verbal (e.g., list learning, story passage recall) and nonverbal (e.g., figure recall) memory. In addition, memory for these stimulus types should be conducted immediately after learning trials (i.e., immediate recall), over the course of multiple repetitions of the stimuli (i.e., learning over trials), following an approximately 20–30 min delay (i.e., delayed recall), and using a yes/no recognition testing format (i.e., recognition) (Scott & Schoenberg, 2011d).

Based on the performance on these learning and memory tasks, one can characterize the episodic memory subprocesses impaired in a patient. For instance, a patient exhibiting poor immediate recall and limited improvement over multiple repetitions of to-be-learned material but who retains most information over a delay likely has a primary deficit in encoding. A patient with poor immediate recall, but some learning over multiple repetitions of information, as well as severely impaired delayed recall that only slightly improves with recognition likely has a primary deficit in consolidation. Finally, a patient with normal immediate recall and learning, impaired delayed recall, and normal recognition likely has a primary deficit in retrieval. Of course, patients can also have deficits in multiple episodic memory processes, leading to even greater specificity in memory profile characterization and neuroanatomical lesion localization (Scott & Schoenberg, 2011d).

Multiple memory tests are available for administration. Representative tests of episodic memory include the Wechsler Memory Scale-IV (Wechsler, Holdnack, & Drozdick, 2009), California Verbal Learning Test-II (Delis et al., 2000), California Verbal Learning Test-Children's Version (Delis, Kramer, Kaplan, & Ober, 1994), and Children's Memory Scale (M. Cohen, 1997).

Executive Functions

Executive functions are a heterogeneous collection of cognitive processes. Fundamentally, executive functions are involved in integrating other cognitive domains to make decisions and initiate complex actions. There are several component processes involved in executive functions. These processes include planning, problem-solving, concept formation (i.e., identify abstract concepts), set shifting, verbal fluency, inhibitory control, and working memory. Certain aspects of memory, especially encoding strategy use and memory retrieval, also involve executive operations. These processes rely principally on the integrity of the frontal lobes. Notably, executive functions undergo rapid change during development. As such, greater inter-individual variability in executive function performance is expected among children and adolescents (Floden, 2014; Scott & Schoenberg, 2011b).

One popular model of frontal lobe functioning divides the prefrontal cortex into three functional regions. The first of these regions involves the dorsolateral prefrontal cortex. Dysfunction in this region is characterized by difficulty carrying out complex goal-directed behavior due to difficulties with organization, planning, sequencing, and selecting and implementing strategies, comprising a so-called dysexecutive syndrome. Individuals with damage to the dorsolateral prefrontal cortex may also appear mentally inflexible and perseverate on incorrect strategies and exhibit poor working memory, especially with regard to mental manipulation of information. In addition, this region supports complex aspects of attention, including selective attention and attention shifting. As such, individuals with damage to the dorsolateral prefrontal cortex may appear highly distractible. The dorsolateral prefrontal cortex is also important for implementing encoding strategies (e.g., semantically categorizing to-be-remembered material) and retrieving items from episodic long-term memory (Floden, 2014; Scott & Schoenberg, 2011b). There is some suggestion of hemispheric specialization for these operations, with the left dorsolateral prefrontal cortex specialized for encoding strategy use and the right dorsolateral prefrontal cortex for retrieval (Habib, Nyberg, & Tulving, 2003).

The second important region of the prefrontal cortex is the orbital/ventromedial region. Damage to this region often results in a "disinhibition syndrome," characterized by lack of impulse control and social filter, inability to delay gratification, and emotional dysregulation. Individuals with damage to this region often exhibit poor judgment, with their behavior governed by the pursuit of immediate reinforcers. In addition, these individuals are often hyperverbal and hyperactive, have difficulties with sustained attention, and have little insight into their behavior (Floden, 2014; Scott & Schoenberg, 2011b).

The third important functional region of the prefrontal cortex is the medial frontal region. Damage to this area results in an "apathetic syndrome." Patients often exhibit lethargy, lack of spontaneous initiation of behavior, and appear disengaged from their environment. They may also appear to be emotionally indifferent, dull, and unmotivated and to lack curiosity (Floden, 2014; Scott & Schoenberg, 2011b).

A variety of measures are available for executive function assessment. Planning can be measured with Tower tests (Delis, Kaplan, & Kramer, 2001) or through more qualitative methods such as inspection of complex figure copying strategy (Meyers & Meyers, 1995). Verbal fluency is often measured with phonemic (Benton, Hamsher, & Sivan, 1983) or semantic (Lezak et al., 2012) word generation tasks. The Wisconsin Card Sorting Test is one of the most popular measures of executive functions, measuring abstract concept formation, set maintenance and set switching, and novel problem-solving (Heaton, 1981). Complex attention can be measured in a variety of ways. The Trail Making Test is a popular measure of task set switching of attention (Reitan, 1958), and the Stroop Test (Golden, 1976) is a class measure of selective attention and the ability to suppress a prepotent response. There are considerably fewer measures available for assessment of the disinhibition syndrome. The Iowa Gambling Task is a commonly administered measure of orbital and ventromedial prefrontal cortex function and involves the ability to learn and adapt behavior to response-reward contingencies (Bechara, Damasio, Damasio, & Anderson, 1994). Assessment of the apathetic syndrome is often achieved through self- or informant-report instruments, rather than performance-based measures. Finally, the Frontal Systems Behavior Inventory evaluates common symptoms resulting from frontal lobe damage and provides separate scales for dysexecutive, disinhibited, and apathetic symptoms. This measure provides both self- and informant-report forms, enabling evaluation of discrepancies across raters, which may shed additional light on patient insight into executive deficits (Grace & Malloy, 2001).

Language

Language is arguably the most distinctly human of all cognitive abilities. While other animals are able to communicate, the scope and complexity of human language is unparalleled. At its core, language refers to the use of arbitrary symbols to convey meaning and involves the ability to comprehend, formulate, and produce spoken, written, and gestural symbolic representations. These abilities are distinct from speech, which involves the ability to control the articulatory movements necessary to produce oral expression. Most language functions are mediated by the dominant (often left) hemisphere, although the ability to comprehend and produce prosody in language involves the non-dominant (often right) hemisphere. Due to the relative importance of non-prosodic aspects of language function, this section will focus only on dominant hemisphere language abilities (Sabsevitz & Hammeke, 2014; Schoenberg & Scott, 2011; Scott & Schoenberg, 2011c).

The ability to use language depends on several constituent processes, including orthographic processing (grouping individual letters into words), phonological processing (deciphering, mentally maintaining, and retrieving the articulatory movements and sound structure of speech), and semantic processing (storing and accessing word meanings). These three fundamental language processes rely on distinct neuroanatomical regions. Orthographic processing is subserved by the left inferior temporal-occipital region. Individuals with damage to this area often exhibit adequate language production, comprehension, writing, and identification of single letters, but their reading of words is slow and effortful (Sabsevitz & Hammeke, 2014). In contrast, phonological processing involves a broader network of brain regions in the dominant hemisphere, including the supramarginal gyrus, superior temporal gyrus and sulcus, and the inferior frontal gyrus. Damage to this area can lead to difficulties selecting and sequencing phonemes during speech production and/or difficulties maintaining or manipulating phonological information in working memory (Sabsevitz & Hammeke, 2014). Semantic processing is widely distributed throughout the temporal and parietal lobe, especially the lateral temporal lobe and angular gyrus. Impairments in semantic processing can result in difficulties with spoken word comprehension, semantic categorization, and object naming (Binder, Desai, Graves, & Conant, 2009).

Language functions are typically divided into two major groups, receptive and expressive language, which both rely on varying degrees of the three abovementioned language processes. Receptive language consists of the ability to comprehend language, both written and spoken, and is localized to the posterior dominant hemisphere. Language comprehension requires processing the phonological elements of speech sounds, retaining phonological information long enough to enable processing of semantic content, and deciphering the semantic content of words and sentences. The dominant hemisphere temporal-parietal area including Wernicke's area is central to comprehension of spoken and written language (Scott & Schoenberg, 2011c). This region is located adjacent to the primary auditory cortex and is key to isolating specific phonemic characteristics of sound into known phonemic systems (Kolb & Whishaw, 2009).

In contrast, expressive language deficits involve difficulties in producing speech. To produce speech, an individual must use multiple language subprocesses, including those involved in semantics (to conceptualize the intended communication), syntax (to order the intended communication), phonology (to map semantic knowledge onto speech sounds), and motor planning (to activate the appropriate motor programs for the intended communication). These processes depend on the posterior inferior frontal lobe as well as the dorsal prefrontal cortex (Sabsevitz & Hammeke, 2014).

Assessment of expressive language should involve at a minimum an analysis of spontaneous speech for output fluency, accuracy of word retrieval, integrity of grammar/syntax, length of utterances, omission of words, meaningfulness of content, and articulatory precision of word production. In addition, tests of picture naming, verbal fluency, phrase repetition, story description, and a writing sample should be administered as part of an assessment of expressive language. Measures of auditory comprehension are key to the assessment of receptive language. Measures of auditory comprehension include simple and complex command following, word-picture matching, yes/no questions, word reading, and sentence comprehension (Sabsevitz & Hammeke, 2014; Schoenberg & Scott, 2011; Scott & Schoenberg, 2011c). Several language-specific batteries have been developed to assess each of these areas of language functioning, including the Boston Diagnostic Aphasia Examination (Goodglass, Kaplan, & Barresi, 2000) and the Western Aphasia Battery Revised (Kertesz, 2006).

Visuoperceptual, Visuospatial, and Visuoconstructional Abilities

Visuoperceptual and visuospatial abilities are central to competent everyday functioning. Most of these abilities occur automatically and outside of awareness, thus obscuring their importance. However, when the brain regions that underlie these functions are damaged, a number of deficits may occur. These deficits may include an inability to process basic aspects of stimuli (e.g., color, form, and orientation), recognize familiar objects, find one's way in the environment, copy or draw designs, or attend to objects in the left hemispatial field. The nature of these deficits and their corresponding functional neuroanatomy are the topic of this section.

Visuoperceptual and visuospatial functional anatomy involves two separate streams of visual information processing that occur in parallel, despite the unitary nature of conscious experience of the visual world. Initial analysis of visual information occurs in the primary visual cortex in the occipital lobe. Primary visual cortex is responsible for extracting very basic visual information from stimuli, including orientation, spatial frequency, and direction of visual stimulus movement. This information is then sent to brain regions involved in higher levels of visual processing, which integrate these initial visual perceptual features into integrated wholes. These higher-level visual functions are predominantly processed in the right hemisphere and involve two separate pathways, a ventral stream involved in visuoperceptual analysis for object recognition and a dorsal stream involved in visuospatial analysis to guide movement in relation to visual information (Scott & Schoenberg, 2011a).

The ventral stream runs from the occipital to temporal lobe. These regions are involved in the perception of form, color, and shape. Damage to this stream can result in difficulties discriminating between colors (termed achromatopsia), defects in form discrimination, problems with matching identical stimuli, and difficulties mentally synthesizing fragmented pictures into complete wholes. Bilateral damage to the fusiform gyrus can result in a more specific inability to recognize faces (termed prosopagnosia). More extensive damage to the occipitotemporal region can result in visual agnosias, which are characterized by an inability to recognize visually presented objects. Two types of visual agnosias have been identified. Apperceptive visual agnosia involves an inability to group together visual stimulus elements into a whole object, despite accurate perception of attributes of the object. Associative visual agnosia consists of accurate perception of a whole, integrated object, but a failure to link semantic knowledge to that perception (Bauer, 2014; Scott & Schoenberg, 2011a).

The dorsal stream runs from the occipital cortex through the superior temporal gyrus and into the parietal lobe. This region is involved in visuospatial processing and visuomotor interaction. Damage to the dorsal, visuospatial stream also results in a range of deficits, including difficulties attending to the left hemispatial field (termed attentional neglect), an inability to localize objects in space, difficulties finding one's way in the environment with respect to the self (termed egocentric disorientation), and problems with judging the relative orientation of lines. Deficits in visuoconstructional abilities, such as putting blocks together to construct designs or drawing complex figures, can result from damage to either ventral or dorsal streams, although the nature of deficits may differ depending on the region of damage (Bauer, 2014; Scott & Schoenberg, 2011a).

A variety of measures are available to assess visuoperceptual and visuospatial abilities. As a first step, sensory functioning, including visual acuity, oculomotor ability, and visual field deficits, should be evaluated (Scott & Schoenberg, 2011a). Commonly administered measures of visuoconstruction include the WAIS-IV Block Design task (Wechsler, 2008) and the Rey-Osterrieth Complex Figure Test (Meyers & Meyers, 1995). Visuospatial ability can be measured with the Judgment of Line Orientation test (Benton, Varney, & Hamsher, 1978) and hemispatial neglect by line bisection and cancellation tasks (Lezak et al., 2012). Finally, measures of visuoperceptual ability include the Benton Visual Form Discrimination test (Benton, Sivan, & Hamsher, 1983) and the Hooper Visual Organization Test (Hooper, 1983).

Future Directions in Neuropsychology

Clinical neuropsychology is a rapidly advancing field. Technological developments as well as increasing recognition of the limitations of neuropsychological assessment to nonmainstream cultural groups have led to some of the most exciting and important new research areas in neuropsychology. This section discusses three of these emerging research areas: cultural neuropsychology, connectomics, and the integration of genomics with neuropsychology.

Cultural Neuropsychology

The United States is currently undergoing a dramatic demographic shift that is changing the composition of the country (Bureau, 2011). This, among other factors, has led to growing interest on the impact of culture on neuropsychological test performance. Traditionally, neuropsychology has struggled to account for cultural contributions to test performance, leading to reduced diagnostic specificity with examinees from nonmainstream cultural groups (Cagigas & Manly, 2014). This has largely been due to a mismatch between normative standardization samples and culturally diverse examinees, with most standardization samples consisting of well-educated, English-speaking, US-born, culturally mainstream Whites. Cultural neuropsychology is an attempt to improve the discipline of neuropsychology by exploring the effects of and pathways by which cultural experiences impact brain-behavior relationships. In this way, cultural neuropsychology may be defined as the study of brain-behavior relationships that emerge out of the manner in which human being engage in culture-specific practices that shape the organization, development, and revision of their cognition and behavior (Cagigas & Manly, 2014).

There are several important strains of cultural neuropsychological research. First, a considerable amount of research has explored the causes of test sore differences across cultural groups. In the aggregate, individuals from nonmainstream cultural backgrounds typically score lower on neuropsychological tests than their culturally mainstream counterparts. This may be due to one or more of the following factors: (1) improper translation of tests, including culturally inappropriate wordings and use of inappropriate norms (van der Vijver & Hambleton, 1996); (2) discrepancies in the quality of education across cultural groups, despite similar number of years of education (Manly & Jacobs, 2002); (3) low levels of acculturation to the culture in which the tests were developed and normed (Artiola, Fortuny, Heaton, & Hermosillo, 1998); and (4) reduced comfort and confidence during the test session due to racial socialization (Cagigas & Manly, 2014). In addition, bilingualism has also been shown to affect neuropsychological test performance, leading to poorer scores on measures of vocabulary (Bialystok, Luk, Peets, & Yang, 2009), picture naming (Gollan, Fennema-Notestine, Montoya, & Jernigan, 2007), and semantic fluency (Gollan & Ferreira, 2009; Gollan, Montoya, & Werner, 2002). However, some research suggests that bilingualism confers advantages on executive functioning tasks, presumably due to greater familiarity with managing and switching between two competing representations of language among bilinguals (Bialystok, Craik, & Luk, 2008).

Second, cultural neuropsychological research has explored approaches to developing more culturally sensitive approaches to neuropsychological assessment. Chief among these efforts has been the development of separate norms for racial/ethnic or language groups. This approach has been shown to improve diagnostic accuracy (Miller, Heaton, Kirson, & Grant, 1997). However, some have noted that there is still considerable variability within racial/ethnic and language groups that remains unaccounted for. Ultimately, it has been argued that race and ethnicity may more accurately be considered proxies for, and thus confounded by, other more meaningful variables, such as language proficiency, educational quality, or socioeconomic status (Cagigas & Manly, 2014). In addition, in developing culturally sensitive neuropsychological measures, it is important not to simply assume that the validity of the measure demonstrated in the culturally mainstream group generalizes to a new cultural group. Thus, a growing body of research has sought to evaluate the construct validity of neuropsychological measures across cultural groups (Cagigas & Manly, 2014).

Third, some research has demonstrated differences in cognitive processes across individuals from different cultures. Studies have found that culture can impact a number of cognitive processes, ranging from facial (Goh et al., 2010) and visual object processing (Gutchess, Welsh, Boduroglu, & Park, 2006) to semantic categorization use to facilitate free recall (Gutchess et al., 2006). In fact, culturally specific cognitive information processing has long been recognized in neuropsychology. Luria studied remote rural communities in Uzbekistan and noted that individuals from these communities did not categorize objects based on abstract rules, like their educated adult counterparts, but rather did so based on the relationship of the stimulus objects to their everyday lives (Nell, 1999, 2000). Nonetheless, evaluation of the culture-specific nature of cognition has largely been overlooked in subsequent years. An exciting new area of research has begun to explore the manner in which cultural practices shape cognition, emotion, and behavior as well as brain structure and function (Cagigas & Manly, 2014)

Connectomics

Historically, most neuroimaging research in clinical populations has focused on regional abnormalities in brain structure and/or functioning. However, recent advances in neuroimaging and network modeling have led to growing interest in the dynamics of large-scale brain networks within normal populations and abnormalities within and between these networks in clinical populations (Menon, 2011). A central goal of this research is to provide a comprehensive characterization of the structural pathways connecting remote brain regions and the functional interactions between these remote regions. The study of these large-scale networks is referred to as connectomics and the descriptions derived therefrom as the human connectome (Sporns, 2013). Emerging findings from connectomics suggest that the human brain consists of several distinct functional networks and that these networks are organized in a modular fashion, with networks interlinked through sub-networks or core hub regions, which enable global information flow and integration (Sporns, 2013).

Adopting a network-level analysis of brain-behavior relationships has led to novel findings among several clinical populations. For instance, traditional neuroimaging studies evaluating regional abnormalities in brain structure and function among individuals with autism have often reported abnormalities in the frontal lobes, amygdala, and cerebellum (Amaral, Schumann, & Nordahl, 2008). However, many of these findings have been inconsistent across studies. Moreover, descriptions of regional abnormalities may be insufficient to characterize the distributed nature of a disorder like autism, especially given the heterogeneity of the cognitive and behavioral phenotype of this disorder. As such, evaluation of the structure, integrity, and integration of distributed functional networks may be more revealing of brain-behavior relationships in autism.

Recent research on network functional connectivity in autism has revealed important findings. For one, autism is most consistently characterized by underconnectivity between the prefrontal cortex and posterior brain regions and regional overconnectivity within the extrastriate cortex, frontal lobe, temporal lobe, amygdala, parahippocampal gyrus, and temporo-thalamic regions (Maximo, Cadena, & Kana, 2014; Murphy, Foss-Feig, Kenworthy, Gaillard, & Vaidya, 2012; Noonan, Haist, & Muller, 2009; Shih et al., 2010; Shih et al., 2011; Uddin et al., 2013; Welchew et al., 2005). These findings have been taken to suggest inefficient brain network integration, with overabundant connectivity occurring in nonessential regions, leading to poor distributed network coordination in the midst of high levels of regional noise. It remains to be determined whether regional overconnectivity in autism is a cause or an effect of observed underconnectivity across distributed brain regions (Maximo et al., 2014). For another, emerging research has shown that irregularities in specific networks are associated with distinct behavioral abnormalities. For instance, overconnectivity within specific nodes in the default mode network has been associated with lower verbal and nonverbal communication ability, more severe repetitive behaviors, and restricted, repetitive behaviors (Agam, Joseph, Barton, & Manoach, 2010; Monk et al., 2009; Weng et al., 2010). In this sense, brain network abnormalities may serve as promising intermediate phenotypes linking genes to behavior (Sporns, 2013).

Genetics and Neuropsychology

Another promising avenue is the integration of genomics with neuropsychology. Genomic research is focused on identifying genetic contributions to normal and pathological phenotypes and has been made possible by rapid advances in human genome sequencing and techniques for identifying single nucleotide polymorphisms and quantifying gene expression. Cognition has emerged as an important phenotype in these investigations, due to the contribution of cognition to functional outcomes (e.g. vocation, academic achievement) and the presence of cognitive impairment in pathological conditions as diverse as autism, schizophrenia, Parkinson's disease, and Alzheimer's disease. Neuropsychology specializes in teasing apart the multiple cognitive processes contributing to performance on cognitive tasks and linking these specific processes to brain structure and function. As such, neuropsychology is well positioned to assist in genomic research on cognitive phenotypes (Kremen, Panizzon, & Cannon, 2016).

For instance, Panizzon et al. (2011) explored performance on the California Verbal Learning Test-II (Delis et al., 2000), a word list learning episodic memory task, among twin pairs. These authors found that while the same set of genes contributed to short- and long-delay free recall, a different set of genes was associated with performance during learning trials. Short- and long-delay free recall as well as learning trials involve free recall; however, only learning trials additionally involve acquisition of to-be-remembered content. As such, these authors reasoned that there are separate genetic influences on recall and acquisition processes. This implies that episodic memory tests are not interchangeable at the genetic level. Studies that fail to take into consideration this specificity of the multiple cognitive processes contributing to a particular cognitive domain may thus obscure the genetic contribution to these processes.

Exploration of gene-environment interactions is an especially exciting area of genetic and neuropsychology research. Gene-environment interactions deal with the manner in which the response to an environmental factor varies as a function of one's genotype (Kremen et al., 2016). These interactions are important insofar as they indicate the mutability of one's genetic endowment and indicate the potential for psychosocial or behavioral intervention. For instance, Ferencz et al. (2014) created a genetic risk score based on several genes previously associated with episodic memory among individuals differing in levels of physical activity. These authors reported that the effect of the risk score on episodic memory differed across individuals with high vs. low physical activity. In particular, the genetic risk score was associated with episodic memory impairment only among individuals in the low physical activity group. In other words, physical activity was protective against genetic effects on episodic memory. Incorporation of these and other gene-environment interactions has the potential to improve interventions for cognition and can assist in the development of individualized treatment recommendations, a main goal of the emerging field of precision medicine (Collins & Varmus, 2015).

Conclusion

Neuropsychology is an expansive and rapidly developing field. Neuropsychological investigations have deepened our understanding of brain-behavior relationships in the normal brain and the manner in which these relationships are disrupted in a variety of developmental, psychiatric, and neurological disorders. Technological developments in brain imaging and genetics and the expansion of neuropsychology to previously understudied populations promise to yield further transformative insights into brain structure and function.

References

Agam, Y., Joseph, R. M., Barton, J. J., & Manoach, D. S. (2010). Reduced cognitive control of response inhibition by the anterior cingulate in autism spectrum disorders. *NeuroImage, 52*, 336–347.

Amaral, D. G., Schumann, C. M., & Nordahl, C. W. (2008). Neuroanatomy of autism. *Trends in Neurosciences, 31*(3), 137–145.

American Educational Research Association, A. P. A., & National Council on Measurement in Education. (1999). *Standards for educational and psychological testing*. Washington, DC: American Psychological Association.

Ardila, A. (1992). Luria's approach to neuropsychological assessment. *International Journal of Neuroscience, 66*, 35–43.

Artiola, I., Fortuny, L., Heaton, R. K., & Hermosillo, D. (1998). Neuropsychological comparisons of Spanish-speaking participants from teh U.S.-Mexico border region versus Spain. *Journal of the International Neuropsychological Society, 4*, 363–379.

Bauer, R. L. (2014). Visuospatial, visuoperceptual, and visuoconstructional disorders. In M. W. Parsons & T. A. Hammeke (Eds.), *Clinical neuropsychology: A pocket handbook for assessment*. Washington, DC: American Psychological Association.

Bechara, A., Damasio, A. R., Damasio, H., & Anderson, S. W. (1994). Insensitivity to future consequences following damage to human prefrontal cortex. *Cognition, 50*, 7–15.

Benton, A. L., Hamsher, K., & Sivan, A. B. (1983). *Multilingual aphasia examination* (3rd ed.). Iowa City, IA: AJA Associates.

Benton, A. L., Sivan, A. B., & Hamsher, K. (1983). *Contributions to neuropsychological assessment: A clinical manual*. New York: Oxford University Press.

Benton, A. L., Varney, N. R., & Hamsher, K. S. (1978). Visuospatial judgment: A clinical test. *Archives of Neurology, 35*, 364–367.

Bialystok, E., Craik, F. I. M., & Luk, G. (2008). Lexical access in bilinguals: Effects of vocabulary size and executive control. *Journal of Neurolinguistics, 21*, 522–538.

Bialystok, E., Luk, G., Peets, K. F., & Yang, S. (2009). Receptive vocabulary differences in monolingual and bilingual children. *Bilingualism: Language and Cognition, 13*, 525–531.

Binder, J. R., Desai, R. H., Graves, W. W., & Conant, L. L. (2009). Where is the semantic system? A critical review and meta-analysis of 120 functional neuroimaging studies. *Cerebral Cortex, 19*, 2767–2796.

Bureau, U. S. C. (2011). Population estimates. National characteristics: Vintage 2011. Retrieved from http://www.cnesus.gov/popest/data/national/asrh/2011/index.html

Cagigas, X. E., & Manly, J. J. (2014). Cultural neuropsychology: The new norm. In T. A. Hammeke & M. W. Parsons (Eds.), *Clinical neuropsychology: A pocket handbook for assessment*. Washington, DC: American Psychological Association.

Cohen, M. (1997). *Children's memory scale*. San Antonio, TX: NCS Pearson, Inc..

Cohen, R. A., Malloy, P., Jenkins, M., & Paul, R. (2014). Disorders of attention. In M. W. Parsons & T. A. Hammeke (Eds.), *Clinical neuropsychology: A pocket handbook for assessment*. Washington, DC: American Psychological Association.

Collins, F. S., & Varmus, H. (2015). A new initiative on precision medicine. *New England Journal of Medicine, 372*, 793–795.

Connors, C. K. (2000). *Conners' continuous performance test (CPT-2) computer program for windows, technical guide, and software manual*. Toronto, ON: Multi Health Systems Inc.

Delis, D. C., Kaplan, E., & Kramer, J. H. (2001). *Delis-Kaplan executive function system (D-KEFS)*. San Antonio, TX: Psychological Corporation.

Delis, D. C., Kiefner, M., & Fridlund, A. J. (1988). Visuospatial dysfunction following unilateral brain damage: Dissociation in hierarchical and hemispatial analysis. *Journal of Clinical and Experimental Neuropsychology, 10*, 421–431.

Delis, D. C., Kramer, J. H., Fridland, A. J., & Kaplan, E. (1990). A cognitive science approach to neuropsychological assessment. In P. McReynolds, J. C. Rosen, & G. Chelune (Eds.), *Advances in psychological assessment* (Vol. 7, pp. 101–132). New York: Plenum Press.

Delis, D. C., Kramer, J. H., Kaplan, E., & Ober, B. A. (1994). *California verbal learning test – children's version (CVLT-C)*. San Antonio, TX: NCS Pearson, Inc.

Delis, D. C., Kramer, J. H., Kaplan, E., & Ober, B. A. (2000). *California verbal learning test (CVLT-2)*. San Antonio, TX: Psychological Corporation.

Delis, D. C., Robertson, L. C., & Efron, R. (1986). Hemispheric specialization of memory for visual hierarchical stimuli. *Neuropsychologia, 24*, 205–214.

Ferencz, B., Laukka, E. J., Welmer, A. K., Kalpouzos, G., Angleman, S., Keller, L., & Backman, L. (2014). The benefits of staying active in old age: Physical activity counteracts the negative influence of PICALM, BIN1, and CLU risk alleles on episodic memory functioning. *Psychology and Aging, 29*(2), 440–449.

Floden, D. (2014). Frontal lobe function. In M. W. Parsons & T. A. Hammeke (Eds.), *Clinical neuropsychology: A pocket handbook for assessment*. Washington, DC: American Psychological Association.

Friston, K. J. (1997). Imaging cognitive anatomy. *Trends in Cognitive Sciences, 1*(1), 21–27.

Galaburda, A., & Pascual-Leone, A. (2003). Mechanisms of plasticity and behavior. In T. E. Feinberg & M. J. Farah (Eds.), *Behavioral neurology and neuropsychology*. New York: McGraw Hill.

Glozman, J. M. (2007). A.R. Luria and the history of Russian neuropsychology. *Journal of the History of the Neurosciences, 16*, 168–180.

Goh, J. O. S., Leshikar, E. D., Sutton, B. P., Tan, J. C., Sim, S. K. Y., Hebrank, A. C., & Park, D. C. (2010). Culture differences in neural processing of faces and houses in the ventral visual cortex. *Social Cognitive and Affective Neuroscience, 5*(2–3), 227–235.

Golden, C. J. (1976). Identification of brain disorders by the Stroop color and word test. *Journal of Clinical Psychology, 32*(3), 654–658.

Goldstein, G. (2015). Ralph Reitan and biological intelligence. *Archives of Clinical Neuropsychology, 30*, 733–739.

Gollan, T. H., Fennema-Notestine, C., Montoya, R. I., & Jernigan, T. L. (2007). The bilingual effect on Boston naming test performance. *Journal of the International Neuropsychological Society, 13*(2), 197–208.

Gollan, T. H., & Ferreira, V. S. (2009). Should I stay or should I switch? A cost-benefit analysis of voluntary language switching in young and aging bilinguals. *Journal of Experimental Psychology: Learning, Memory, and Cognition, 35*, 640–665.

Gollan, T. H., Montoya, R. I., & Werner, G. A. (2002). Semantic and letter fluency in Spanish-English bilinguals. *Neuropsychology, 16*, 562–576.

Goodglass, H., Kaplan, E., & Barresi, B. (2000). *Boston diagnostic aphasia examination - third edition (BDAE-3)*. San Antonio, TX: NCS Pearson Inc.

Grace, J., & Malloy, P. F. (2001). *Frontal systems behavior scale (FrSBe): Professional manual*. Odessa, FL: Psychological Assessment Resources.

Greiffenstein, M. (2014). Important theories in neuropsychology: A historical perspective. In K. J. Stucky, M. W. Kirkwood, & J. Donders (Eds.), *Neuropsychology study guide & board review*. New York, NY: Oxford University Press.

Gronwall, D. M. A. (1977). Paced auditory serial-addition task: A measure of recovery from concussion. *Perceptual and Motor Skills, 44*, 367–373.

Gutchess, A. H., Welsh, R. C., Boduroglu, A., & Park, D. C. (2006). Cultural differences in neural function associated with object processing. *Cogntitive, Affective, and Behavioral Neuroscience, 6*, 102–109.

Gutchess, A. H., Yoon, C., Luo, T., Feinberg, F., Hedden, T., Jing, Q., et al. (2006). Categorical organization in free recall across culture and age. *Gerontology, 52*, 314–323.

Habib, R., Nyberg, L., & Tulving, E. (2003). Hemispheric asymmetries of memory: The HERA model revisited. *Trends in Cognitive Sciences, 7*, 241–245.

Heaton, R. K. (1981). *Wisconsin card sorting test manual*. Odessa, FL: Psychological Assessment Resources, Inc.

Heaton, R. K., Grant, I., Anthony, W. Z., & Lehman, R. A. (1981). A comparison of clinical and automated interpretation of the Halstead-Reitan battery. *Journal of Clinical and Experimental Neuropsychology, 3*, 121–141.

Heaton, R. K., & Marcotte, T. D. (2000). Clinical neuropsychological tests and assessment techniques. In F. Boller, J. Grafman, & G. Rizzolatti (Eds.), *Handbook of neuropsychology*. Amsterdam, Netherlands: Elsevier Science Publishers B.V..

Hom, J., & Nici, J. (2015). Ralph M. Reitan: The pioneer of clinical neuropsychology. *Archives of Clinical Neuropsychology, 30*, 724–732.

Hooper, H. E. (1983). *Hooper visual organization test manual*. Los Angeles: Western Psychological Services.

Horton, A. M., & Puente, A. E. (1986). Human neuropsychology: An overview. In D. Wedding, A. M. Horton, & J. Webster (Eds.), *The neuropsychology handbook: Behavioral and clinical perspectives*. New York: Springer.

Huettel, S. A., Song, A. W., & McCarthy, G. (2014). *Functional magnetic resonance imaging*. Sunderland, MA: Sinauer Associates, Inc..

Kertesz, A. (2006). *Western aphasia battery, revised*. San Antonio, TX: NCS Pearson Inc.

Kolb, B., & Whishaw, I. Q. (2009). *Fundamental of human neuropsychology* (6th ed.). New York, NY: Worth Publishers.

Kosslyn, S. M. (1999). If neuroimaging is the answer, what is the question? *Philosophical Transactions of the Royal Society of London B, 354*, 1283–1294.

Kremen, W. S., Panizzon, M. S., & Cannon, T. D. (2016). Genetics and neuropsychology: A merger whose time has come. *Neuropsychology, 30*(1), 1–5.

Lezak, M. D., Howieson, D. B., Bigler, E. D., & Tranel, D. (2012). *Neuropsychological assessment* (5th ed.). New York: Oxford University Press.

Manly, J. J., & Jacobs, D. M. (2002). Future directions in neuropsychological assessment with African Americans. In F. R. Ferraro (Ed.), *Minority and cross-cultural aspects of neuropsychological assessment*. Lisse, Netherlands: Swetz and Zeitlinger.

Maximo, J. O., Cadena, E. J., & Kana, R. K. (2014). The implications of brain connectivity in the neuropsychology of autism. *Neuropsychology Review, 24*, 16–31.

Menon, V. (2011). Large-scale brain networks and psychopathology: A unifying triple network model. *Trends in Cognitive Sciences, 15*(10), 483–506.

Messick, S. (1995). Validity of psychological assessment: Validation of inferences from persons' responses and performance as scientific inquiry into scoring meaning. *American Psychologist, 50*(9), 741–749.

Meyers, J. E., & Meyers, K. R. (1995). *Rey complex figure test and recognition trial*. Odessa, FL: Psychological Assessment Resources.

Miller, S. W., Heaton, R. K., Kirson, D., & Grant, I. (1997). Neuropsychological (NP) assessment of African Americans. *Journal of the International Neuropsychological Society, 3*, 49.

Mirsky, A. F., Anthony, B. J., Duncan, C. C., Ahearn, M. B., & Kellam, S. G. (1991). Analysis of the elements of attention: A neuropsychological approach. *Neuropsychology Review, 2*, 109–145.

Mishkin, M. (1982). A memory system in the monkey. *Philosophical Transactions of the Royal Society of London B, 298*, 85–95.

Mitrushina, M. N., Boone, K. B., & D'Elia, L. F. (2005). *Handbook of normative data for neuropsychological assessment*. New York, NY: Oxford University Press.

Monk, C. S., Peltier, S. J., Wiggins, J. L., Weng, S. J., Carrasco, M., Risi, S., & Lord, C. (2009). *Abnormalities of intrinsic functional connectivity in autism spectrum disorders* (Vol. 47, p. 764).

Morton, J., & Frith, U. (1995). Causal modeling: A structural approach to developmental psychopathology. In D. Cicchetti & D. J. Cohen (Eds.), *Developmental psychopathology*. New York: John Wiley & Sons.

Murphy, E. R., Foss-Feig, J., Kenworthy, L., Gaillard, W. D., & Vaidya, C. J. (2012). Atypical functional connectivity of the amygdala in childhood autism spectrum disorders during spontaneous attention to eye-gaze. *Autism Research and Treatment, 2012*, 652408.

Nell, V. (1999). Luria in Uzbekistan: The vicissitudes of cross-cultural neuropsychology. *Neuropsychology Review, 9*(1), 45–52.

Nell, V. (2000). *Cross-cultural neuropsychological assessment: Theory and practice*. Mahwah, NJ: Erlbaum.

Noonan, S. K., Haist, F., & Muller, R. A. (2009). Aberrant functional connectivity in autism: Evidence from low-frequency BOLD signal fluctuations. *Brain Research, 1262*, 48–63.

Nunnally, J. C., & Bernstein, I. H. (1994). *Psychometric theory*. New York: McGraw HIll, Inc.

O'Connor, M. G., & Race, E. (2013). Amnestic syndromes. In M. W. Parsons & T. A. Hammeke (Eds.), *Clinical neuropsychology: A pocket handbook for assessment*. Washington, DC: American Psychological Association.

Ogden, J. A. (2005). *Fractured minds: A case-study approach to clinical neuropsychology*. New York: Oxford University Press.

Panizzon, M. S., Lyons, M. J., Jacobson, K. C., Franz, C. E., Grant, M. D., Eisen, S. A., & Kremen, W. S. (2011). Genetic architecture of learning and delayed recall: A twin study of episodic memory. *Neuropsychology, 25*, 488–498.

Pennington, B. F. (2006). From single to multiple deficit models of developmental disorders. *Cognition, 101*, 385–413.

Pennington, B. F. (2009). How neuropsychology informs our understanding of developmental disorders. *Journal of Child Psychology and Psychiatry, 50*(1–2), 72–78.

Reitan, R. M. (1958). Validity of the trail making test as an indicator of organic brain damage. *Perceptual and Motor Skills, 8*, 271–276.

Reitan, R. M. (1964). Psychological deficits resulting from cerebral lesions in man. In J. M. Warren & K. A. Akert (Eds.), *The frontal granular cortex and behavior* (pp. 295–312). New York: McGraw-Hill.

Reitan, R. M. (1976). Neuropsychology: The vulgarization that Luria always wanted. *Contemporary Psychology, 21*, 737–738.

Reitan, R. M., & Wolfson, D. (2009). The Halstead-Reitan neuropsychological tests battery for adults - theoretical, methodological and validational bases. In I. Grant & K. M. Adams (Eds.), *Neuropsychological assessment of neuropsychiatric and neuromedical disorders* (3rd ed.). New York: Oxford University Press.

Robertson, L., & Delis, D. C. (1986). "Part-whole" processing in unilateral damaged patients: Dysfunction of hierarchical organization. *Neuropsychologia, 24*, 363–370.

Rourke, B. P., & Brown, G. G. (1986). Clinical neuropsychology and behavioral neurology: Similarities and differences. In S. B. Filskov & T. J. Boll (Eds.), *Handbook of clinical neuropsychology* (Vol. 2). New York: Wiley.

Royall, D. R., Mahurin, R. K., & Gray, K. F. (1992). Bedside assessment of executive cognitive impairment: The executive interview. *Journal of the American Geriatrics Society, 40*, 1221–1226.

Sabsevitz, D. S., & Hammeke, T. A. (2014). The aphasias. In M. W. Parsons & T. A. Hammeke (Eds.), *Clinical neuropsychology: A pocket handbook for assessment*. Washington, DC: American Psychological Association.

Schoenberg, M. R., & Scott, J. G. (2011). Aphasia syndromes. In M. R. Schoenberg & J. G. Scott (Eds.), *The little black book of neuropsychology*. New York, NY: Springer.

Scott, J. G., & Schoenberg, M. R. (2011a). Deficits in visuospatial/visuoconstructional skills and motor praxis. In M. R. Schoenberg & J. G. Scott (Eds.), *The little black book of neuropsychology*. New York, NY: Springer.

Scott, J. G., & Schoenberg, M. R. (2011b). Frontal lobe/executive functioning. In M. R. Schoenberg & J. G. Scott (Eds.), *The little black book of neuropsychology*. New York, NY: Springer.

Scott, J. G., & Schoenberg, M. R. (2011c). Language problems and assessment: The aphasic patient. In M. R. Schoenberg & J. G. Scott (Eds.), *The little black book of neuropsychology*. New York, NY: Springer.

Scott, J. G., & Schoenberg, M. R. (2011d). Memory and learning: The forgetful patient. In M. R. Schoenberg & J. G. Scott (Eds.), *The little black book of neuropsychology*. New York, NY: Springer.

Shih, P., Keehn, B., Oram, J. K., Leyden, K. M., Keown, C. L., & Muller, R. A. (2011). Functional differentiation of posterior superior temporal sulcus in autism: A functional connectivity magnetic resonance imaging study. *Biological Psychiatry, 70*, 270–277.

Shih, P., Shen, M., Ottl, G., Keehn, B., Gaffrey, M. S., & Muller, R. A. (2010). Atypical network connectivity for imitation in autism spectrum disorder. *Neuropsychologia, 48*, 1780–1791.

Slick, D. J., Strauss, E., Sherman, E. M. S., & Spreen, O. (2006). Psychometrics in neuropsychological assessment. In E. Strauss, E. M. S. Sherman, & O. Spreen (Eds.), *A compendium of neuropsychological tests: Administration, and commentary*. New York, NY: Oxford University Press.

Sporns, O. (2013). Structure and function of complex brain networks. *Dialogues in Clinical Neuroscience, 15*(3), 247–262.

Squire, L. R., & Zola, M. (1996). Structure and function of declarative and nondeclarative memory. *Proceedings of the National Academy of Sciences of the United States of America, 93*, 13515–13522.

Strauss, E., Sherman, E. M. S., & Spreen, O. (2006). Norms selection in neuropsychological assessment. In E. Strauss, E. M. S. Sherman, & O. Spreen (Eds.), *A compendium of neuropsychological tests: Administration, norms, and commentary*. New York, NY: Oxford University Press.

Tranel, D. (2009). The Iowa-Benton school of neuropsychological assessment. In I. Grant & K. M. Adams (Eds.), *Neuropsychological assessment of neuropsychiatric and neuromedical disorders* (3rd ed.). New York: Oxford University Press.

Uddin, L. Q., Supekar, K., Lynch, C. J., Khouzam, A., Phillips, J., Feinstein, C., et al. (2013). Salience network-based classification and prediction of symptom severity in children with autism. *JAMA Psychiatry, 70*, 869–879.

van der Vijver, F., & Hambleton, R. K. (1996). Translating tests: Some practical guidelines. *European Psychologist, 1*, 89–99.

Ward, J. (2006). *The student's guide to cognitive neuroscience*. New York, NY: Psychology Press.

Wechsler, D. (2008). *Wechsler adult intelligence scale - Fourth edition (WAIS-IV)*. San Antonio, TX: NCS Pearson Inc.

Wechsler, D., Holdnack, J. A., & Drozdick, L. W. (2009). *Wechsler memory scale, fourth edition*. San Antonio, TX: NCS Pearson, Inc..

Welchew, D. E., Ashwin, C., Berkouk, K., Salvador, R., Suckling, J., Baron-Cohen, S., & Bullmore, E. (2005). Functional disconnectivity of the medial temporal lobe in Asperger's syndrome. *Biological Psychiatry, 57*, 991–998.

Weng, S. J., Wiggins, J. L., Peltier, S. J., Carrasco, M., Risi, S., Lord, C., & Monk, C. S. (2010). Alterations of resting state functional connectivity in the default network in adolescents with autism spectrum disorders. *Brain Research, 1313*, 202–214.

Yeates, K. O., Ris, D. M., Taylor, H. G., & Pennington, B. F. (2009). *Pediatric neuropsychology: Research, theory, and practice*. New York: The Guilford Press.

Part VII

General Issues in Treatment

Evidence-Based Treatment

27

Nicole M. DeRosa, William E. Sullivan,
and Henry S. Roane

Introduction

Individuals with intellectual disabilities have not always had access to appropriate care. In fact, throughout history there is evidence of how individuals with disabilities were shunned and considered a detriment to society. Institutions, which segregated individuals with disabilities from the mainstream public, were commonplace in the late 1800s and early 1900s. The "care" individuals received in these institutions was often inhumane, lacked a focus on the needs of each individual, and was geared toward lessening the burden of individuals with disabilities on the general public. The advent of the civil rights movement sparked a shift in the acceptance and care of individuals with disabilities. The mistreatment of individuals within institutions was revealed and led to improved conditions, as well as eventually the deinstitutionalization of people with disabilities. Movement toward incorporating individuals with disabilities into society and addressing individual needs to improve their quality of life emerged.

An integral part of meeting the needs of individuals with intellectual disabilities involves identifying and implementing effective treatments. Selecting the most appropriate treatment for individuals with intellectual disabilities can be challenging, in part due to the heterogeneity of the population. That is, the unique needs of individuals diagnosed with intellectual disability do not necessarily adhere to a "one-size fits all" approach. Thus, caregivers, practitioners, physicians, and educators are tasked with finding efficient and effective treatments to meet the specific needs of individuals with intellectual disability.

Caregivers (e.g., parents, educators) must be aware of the most effective treatment options in order to provide appropriate care for individuals with intellectual disability. Historically, limited information or guidance has been available regarding the best treatments and how to effectively implement these treatments. However, the evidence-based practice (EBP) movement brought to light the importance of applying research findings into everyday clinical practice. Thus, the feasibility of non-researchers to access the relevant research in order to identify and implement the most appropriate treatments for individuals with intellectual disabilities has not only been made possible but is also now commonplace. As a result of the EBP movement, information regarding evidence-based practices and treatments (more commonly referred to as empirically supported treatments) for a variety of populations, diagnoses, and symptoms is readily disseminated through registries, guides, and journals. The accessibility of this information

---

N. M. DeRosa (✉) · W. E. Sullivan · H. S. Roane
Department of Pediatrics, SUNY Upstate Medical
University, Syracuse, NY, USA
e-mail: derosan@upstate.edu

J. L. Matson (ed.), *Handbook of Intellectual Disabilities*, Autism and Child Psychopathology
Series, https://doi.org/10.1007/978-3-030-20843-1_27

allows for individuals with intellectual disabilities to obtain the care they need and deserve.

This chapter will first review the history or evolution of the EBP movement to guide the reader in understanding EBP as it is known today. Next, the definition of EBP will be presented along with a breakdown of the three main components: (1) research evidence, (2) clinical expertise, and (3) patient characteristics, preferences, and values. A brief discussion regarding the differences between EBP and empirically supported treatments (ESTs) is then provided, followed by a detailed section on the evaluation of ESTs for individuals with intellectual disabilities. Finally, the chapter concludes with consideration of barriers to identifying and implementing ESTs, as well as discussion of the future directions of EBP.

Evolution of Evidence-Based Practice

Evidence-based practice (EBP) is a decision-making model rooted within the field of medicine (Sackett, Rosenberg, Muir Gray, Haynes, & Richardson, 1996; Thoma & Eaves, 2015). Although the philosophical foundation of evidence-based medicine can be traced back to Europe in the eighteenth and nineteenth centuries, a more widespread movement toward evidence-based medicine across Europe and North America did not occur until the mid-twentieth century. It wasn't until the early 1990s that the more modern concept of EBP emerged (APA, 2006; Sackett et al., 1996; Zimerman, 2013). The philosophical underpinnings of EBP are closely linked to epidemiology, which focuses on the needs of populations. In the late 1930s, John R. Paul founded clinical epidemiology, which stemmed from the desire to address not only the needs of entire populations but also of individual patients (Zimerman, 2013). That is, the practices of employing measurable theories and approaches to study diseases and other health-related incidents in populations were incorporated into the everyday decision-making model of clinical practice to positively affect individual patient outcomes. The use of scientific models to inform clinical practice is the impetus behind the EBP movement.

The focus on specific patient needs within clinical practice, as well as refinement of the medical decision-making process, proliferated during the 1960s, as described in a review of EBP conducted by Sur and Dahm (2011). Alvan Feinstein, a follower of Paul's who studied mathematics prior to becoming a physician, played an integral role in incorporating basic science into the practice of medicine (Zimerman, 2013). During his involvement in a rheumatic fever epidemiological study, Feinstein identified an overreliance on clinical expertise alone when determining the presence of relevant symptoms of the disease. This led Feinstein to develop scientific-based criteria for evaluating the symptoms of study participants, which resulted in a more accurate classification of the symptoms of rheumatic fever. In turn, a dramatic decrease in the prevalence of the disease was observed following improved patient outcomes. Furthermore, given Feinstein's criticism of the ability of public health studies to appropriately train physicians in clinical care, he paved the way for improving the value of medical research by linking science and clinical experience. That is, the subjective nature of public health research limited the ability to apply the research findings to improve clinical care. Thus, Feinstein began to modify traditional medical teaching by merging statistical methods, inherent to epidemiology, with clinical reasoning to positively affect patient outcomes.

Around the same time that Feinstein's work was unfolding, events that influenced the movement toward EBP were also occurring in Canada, namely, a shift toward universal healthcare (Zimerman, 2013). One outcome of the change in the Canadian healthcare system during the mid-1960s was the emergence of new medical schools aimed at incorporating basic science into the medical curriculum. Of particular importance was the founding of McMaster University, where the first department of clinical epidemiology and biostatistics emerged, a program heavily influenced by the work of Feinstein who served as a visiting professor during the program's initial years. A key feature of the McMaster program was the development of a "problem-based learning" (Zimerman, 2013; p. 72) technique in which clinical scenarios were presented in a group format in order to incorporate both the basic sciences and clinical medicine into the teaching experience. Although Feinstein's involvement with the newly developed department was crucial in fostering the novel "problem-based learning" teaching method, much of the development of the curriculum and training techniques that influenced the future of EBP is credited to the department head at the time, David Sackett (Sur & Dahm, 2011).

Trained in Public Health, Sackett saw a need for clinical practice to be driven by science in order to better affect the health outcomes of individuals; however, he recognized that most epidemiological training programs addressed public health issues in a manner that was not readily accessible to physicians (Thoma & Eaves, 2015). At the time, clinical practice predominantly involved patient observation and physician opinion. That is, medical programs were not training physicians to be consumers of epidemiological research such that they could apply those findings to their patients. Thus, during his time as head of the Department of Clinical Epidemiology and Biostatistics, Sackett focused on structuring the curriculum such that issues related to public health and practices rooted in science (e.g., biostatistics) were readily incorporated into physician training.

In addition to a novel epidemiological curriculum that encompassed the problem-based learning approach, Sackett and colleagues published a series of articles to assist physicians with effectively accessing information from the literature that could then be applied at the bedside (Thoma & Eaves, 2015). These articles were published in the *Canadian Medical Association Journal (CMAJ)* in the early 1980s and eventually titled "Readers' Guides" (Zimerman, 2013). Following the initial publication of the Readers' Guides, Sackett and colleagues continued to disseminate information on the problem-based learning technique through workshops and additional publications. This laid the groundwork for the future of EBP (i.e., linking science and clinical medicine).

Although Sackett was pivotal in the movement toward EBP, it was Gordon Guyatt, a young faculty member at McMaster, who is officially credited with coining the term "evidence-based medicine" (Thoma & Eaves, 2015; Zimerman, 2013). Furthermore, Guyatt was instrumental in continuing the work of Sackett throughout the 1990s and further disseminating the methods of the newly termed concept of evidence-based medicine. During his time at McMaster, Guyatt further cultivated evidence-based teaching methods, as well as developed a medical residency program based on these methods (Zimerman, 2013). Additionally, Guyatt and colleagues partnered with US academic institutions in the early 1990s to form a workgroup aimed at assessing the state of evidence-based medicine at that time. Conclusions of the workgroup suggested that although Sackett's Readers' Guides affected the bedside practice of medical professionals, limitations of the work existed. More specifically, the workgroup noted that the Readers' Guides included a heavy focus on the quality of evidence but lacked focus on the application of evidence to individual patient needs. That is, guidance was needed on how to appropriately apply the evidence to specific clinical cases, particularly when the relevant evidence may be limited (Sur & Dahm, 2011).

The decision to republish Sackett's Readers' Guides into the *Journal of the American Medical Association* (*JAMA*) in the early 1990s allowed for the opportunity to address the limitations noted by the workgroup. An updated, two-part series of the Guides was developed that included a section on evaluation of clinical measurements, edited by Sackett, and a new section, "Users' Guides to the Medical Literature," headed by Guyatt. This update on the Readers' Guides more heavily focused on the application of evidence into the daily practice of clinical medicine (Zimerman, 2013). The first publication of the new series appeared in the pages of *JAMA* in 1992 and publication continued for 8 years, concluding with a total of 32 articles. During the initial years following the first publication, the evidence-based medicine literature was predominantly maintained by McMaster researchers. However, beginning in 1995 the number of outside researchers who published using the phrase "evidence-based medicine" surpassed that of McMaster researchers, indicating that these methods had successfully infiltrated the field of medicine (Zimerman, 2013). Furthermore, a PsychInfo search conducted by DiGennaro-Reed and Reed (2008) displayed a notable increase in publications using the term "evidence-based practice" beginning in the early 2000s demonstrating the breadth of the evidence-based movement beyond the field of medicine.

Although, the evidence-based movement is largely credited to the field of medicine, education and psychology are also known to have strong roots in an evidence-based approach to assessment and treatment. Of particular significance is Lightner Witmer's establishment of the first psychological clinic at the University of Pennsylvania in 1896, which is known as one of the first institutions to conduct experimentation on the implementation of various teaching strategies (APA, 2006; DiGennaro-Reed & Reed, 2008; Fagan & Wise, 2000). Additionally, in 1908 Witmer founded the journal *The Psychological Clinic*, which focused on clinical services and disabled children (Fagan & Wise, 2000). Likewise, in the late nineteenth and early twentieth century, schools began to adopt an evidence-based approach to improving students' success in the classroom through the work of G. Stanley Hall, the founder of the American Psychological Association (APA). Hall's work influenced the development of the first clinic facility to function in a public school, the Department of Scientific Pedagogy and Child Study (Fagan & Wise, 2000). Furthermore, in the 1960s, during the time in which the evidence-based medicine movement was taking off, it became increasingly common for researchers in psychology and education to conduct treatment comparisons to identify the most effective strategies to improve client and student outcomes (Biglan & Ogden, 2008). Thereafter, research in education and psychology proliferated on effective treatments for a variety of problems (e.g., mental health issues, classroom behavior, drug use) across populations (e.g., children, adolescents). Thus, history suggests that the fields of education and psychology were frontrunners in the EBP movement (APA, 2006).

Although education has a long-standing history in the EBP movement, recent developments have been significantly impactful in improving EBP within the field of education, notably the Council for Exceptional Children (CEC) *Standards for Evidence-Based Practices in Special Education* (2014). The CEC standards provide quality indicators and criteria to assist educators with evaluating the quality of research studies to aid in developing and refining teaching practices. Additionally, the What Works Clearinghouse (WWC), funded by the Institute for Educational Sciences within the US Department of Education, is a research database that systematically evaluates school-based academic and behavioral interventions in order to guide the practice of educators. The availability of resources offered by the CEC and WWC is critical, particularly given federal mandates regarding instructional strategies. That is, the *Individuals with Disabilities Education Improvement Act* (IDEA, 2004) and the *No Child Left Behind Act* (2001) both specify that instructional strategies must be selected and designed using "scientifically based research." Therefore, with the help of initiatives like the CEC and WWC, educators can be equipped with treatments that have been scientifically shown to produce favorable outcomes within the school setting.

One significant contribution that the field of psychology had on the EBP movement was APA's development of best practice guidelines. In 1992, APA created a joint task force among the Board of Scientific Affairs, the Board of Professional Affairs, and the Committee for the Advancement of Professional Practice, which resulted in the publication of the *Template for Developing Guidelines*: *Treatments for Mental Disorders and Psychosocial Aspects of Physical Disorders* (APA, 1995, 2006). The goal of the "Template" was to have structured criteria for establishing best practice guidelines that included reference to both relevant research and clinical expertise. The "Template" was later replaced with the *Criteria for Evaluating Treatment Guidelines* in 2002 (APA, 2006). The Task Force on Promotion and

Dissemination of Psychological Procedures emerged from Division 12 (Clinical Psychology) of APA in 1993, which developed criteria for identifying empirically supported treatments. This came in response to many psychological treatments being viewed as inferior to medical treatments (e.g., pharmacology), despite proven effectiveness of certain therapies (i.e., psychotherapy) dating back to the 1970s.

Although the development of treatment criteria brought about increased awareness regarding the effectiveness of psychological treatments for specific disorders, many found the criteria to be too restrictive given an emphasis on randomized controlled trials (RCTs) and treatment manuals (APA, 2006). Thus, psychologists within specific disciplines (e.g., counseling) began to develop additional guidelines that also accounted for patient variables to better inform treatment development and application within clinical practice. Additionally, the 2005 APA Presidential Task Force on Evidence-Based Practice was established in an effort to identify global practice standards to benefit all disciplines of psychology, as well as to help guide policymakers who had a newfound interest in EBP.

In the early 2000s, healthcare systems and healthcare policy began to readily adopt the notion of EBP. To date, various federal, state, and local organizations and agencies incorporate guidelines and/or mandates that specify the need for EBP. For example, several states have enacted an autism insurance mandate that provides coverage for several medical and behavioral treatments (e.g., applied behavior analysis), such that individuals with autism are able to access appropriate, EBP-derived care. Additionally, APA Task Forces continue to work toward the development and refinement of criteria for EBP across psychology disciplines (APA, 2006). Similar developments are observed across several other fields, including behavioral medicine, education, and behavior analysis (Council for Exceptional Children, 2006; Davidson, Trudeau, Ockene, Orleans, & Kaplan, 2003; DiGennaro-Reed & Reed, 2008). A major impetus for incorporating EBP across disciplines, organizations, and agencies is to maximize consumer (e.g., patients, students) outcomes, as well as to ensure physicians, psychologists, teachers, and the like are able to not only access the relevant research but also assess and evaluate it and most importantly put it into practice.

Definition of Evidence-Based Practice

The main purpose of EBP is to narrow the research-to-practice gap to ensure that treatment is not only effective for a given individual but also cost-efficient (Spencer, Detrich, & Slocum, 2012). However, variations in wording and perhaps meaning of what EBP actually entails can be found across disciplines, the impact of which will be discussed later in this chapter. Despite this, there are three general variables that encompass EBP: (1) research evidence, (2) clinical expertise, and (3) patient characteristics. For example, the Institute of Medicine (2001) defines EBP as “the integration of best research evidence with clinical expertise and patient values” (p. 147; adapted from Sackett, Strauss, Richardson, Rosenberg, & Haynes, 2000). Similarly, the 2005 APA Presidential Task Force defines EBP as the “integration of the best available research with clinical expertise in the context of patient characteristics, culture, and preferences” (p. 273). Of significance is the emphasis on research evidence rather than expert opinion alone. However, it is also important to note that EBP does not emphasize research evidence over expert opinion but instead stresses a collaboration between the two. That is, EBP encompasses professional opinion and clinical practice that is guided by research to best meet the needs of individual consumers (Cook, Tankersley, & Harjusola-Webb, 2008). Below we discuss each of the three areas of EBP.

Research Evidence. In order to consider a treatment or other clinical practice to be effective, it must be backed by a strong research base. The APA Presidential Task Force on Evidence-Based Practice indicates that “a sizeable body of evidence drawn from a variety of research designs and methodologies attests to the effectiveness of psychological practices" and furthermore states that "evidence derived from clinically relevant research on psychological practices should be based on systematic reviews, reasonable effect sizes, statistical and clinical significance, and a body of supporting evidence" (APA, 2006, p. 284). However, determining the strength of the relevant research may not always be straightforward.

Most researchers and practitioners would agree that a large amount of scientific evidence is necessary in order to be confident that a particular practice is effective and appropriate for a given individual, but the specific amount or type of evidence needed is not always agreed upon. For example, RCTs are commonly referred to as the "gold standard"; however, this type of research design may not always be the most appropriate given a specific research question or analysis. Accordingly, a number of differing criteria for determining the level of evidence available for a given treatment or practice have been developed and will be discussed in detail later in this chapter. Nonetheless, what is vital to EBP is a critical appraisal of the research.

Clinical Expertise. Clinical expertise encompasses a practitioner's theoretical understanding of the relevant topic, clinical training, knowledge and understanding of the relevant research, self-evaluation, and ongoing education and training (APA, 2006). Sackett et al. (1996) described clinical expertise as the "proficiency and judgment" practitioners obtain through their education and training (p. 71). Through clinical expertise, the relevant research can be effectively applied to individual patients given an understanding of the clinical needs of that patient. Although clinical practice should be guided by the relevant research, at times a particular evidence-based treatment targeting a specific concern may not match the client's unique needs or be feasible to implement in the relevant treatment setting. For example, for a student with an intellectual disability that has verbal outbursts in the classroom maintained by teacher attention, evidence would suggest that withholding attention when the student shouts out and providing attention contingent on an appropriate alternative behavior (e.g., the child raises his/her hand and waits to be called on) can reduce the student's vocal outbursts. However, in a classroom setting, it may be very difficult, if not impossible, to ignore the student's vocal outbursts given the impact this may have on the learning of peers. Therefore, clinical expertise may come into play such that appropriate modifications are made to the treatment to best meet the needs of the student while accounting for variables related to the treatment setting (e.g., classroom).

Furthermore, at times there may simply be an insufficient research base related to the specific needs of a given patient or consumer, and thus practitioners must rely on expert opinion and practice principles guided by science. For instance, suppose there are two separate treatments that have been shown to produce favorable outcomes for a particular concern across multiple empirical studies. However, within the literature there are *no* studies that have directly compared these treatments. In a case such as this, the practitioner must rely on their own clinical expertise to select the most appropriate treatment for their client.

Given clinical expertise encompasses not only the relevant research but also practices based on opinions, experiences, and an understanding of the patient or consumer, it is important that practitioners are aware of the limits of their knowledge, as well as their own biases when making clinical decisions (APA, 2006). One manner in which clinical expertise can be evaluated is through examining the efficacy and efficiency of clinical decision-making (e.g., patient outcomes; Sackett et al., 1996). Although relying solely on clinical expertise can produce outdated practices that negatively impact the consumer, clinical practice in the absence of clinical expertise (i.e., relying only research evidence) may result in the unique needs of individual consumers not being reliably met (Sackett et al., 1996).

Patient Characteristics. Consideration of patient characteristics entails more than the target symptoms or problem at hand. Evidence-based practice also considers patient or consumer preferences, values, sociocultural context, and strengths, as these variables are likely to affect treatment outcomes (APA, 2006). Thus, by taking patient characteristics into account, the practitioner is best equipped to apply the relevant research, in conjunction with expert opinion, to best meet the needs of the patient rather than the disorder, symptom, or concern. For example, suppose a contingency has been arranged for a patient with an intellectual disability to increase their physical activity such that for every mile the patient walks, they can earn a specific reinforcer. The effectiveness of this contingency ultimately relies on how motivated the patient is to obtain the programmed reinforcer. One way to increase the chances that the contingency will result in the desired behavior change (i.e., increase the patient's physical activity) is to assess the patient's preference for items they could earn as a reinforcer (e.g., Fisher et al., 1992). By doing so, the patient's most preferred item can be programmed into treatment. Accordingly, contingent access to a highly preferred reinforcer will likely increase the patient's physical activity to a greater extent than earning access to a low-preferred reinforcer.

Along these lines, Swift, Callahan, and Vollmer (2011) conducted a meta-analysis (described later in this chapter) that compiled data from 35 studies comparing outcomes of patients in which preference was incorporated into treatment or not. Findings suggested that for those patients in which preferences were accounted for, significantly better outcomes ($d = 0.31$) were obtained. This study highlights the benefits of incorporating patient preference into treatment and aligns with EBP. For further discussion of incorporating patient characteristics as a component of EBP, please refer to APA Presidential Task Force on Evidence-Based Practice (2006).

Evidence-Based Practice and Empirically Supported Treatments

Evidence-based practice is a comprehensive, global approach to patient or consumer care. In other words, it is a decision-making model that seeks to answer a specific question as it relates to an individual (APA, 2006; Schlosser & Sigafoos, 2008). Thus, it is important to point out that EBP differs from empirically supported treatments (EST), which are treatments with known effectiveness for a given population and/or problem. Schlosser and Sigafoos (2008) define ESTs as treatments that have “obtained a certain threshold of research evidence” (p. 61). Although significant to EBP, ESTs are one component of the decision-making process. That is, when employing EBP, practitioners will select treatments that (a) have a substantial research base (i.e., ESTs), (b) best meet the individual's needs based on the practitioner's clinical expertise, and (c) account for the individual's characteristics and preferences.

Evaluating Empirically Supported Treatments

As previously noted, EST are but one component of the EBP decision-making process. In order for a treatment to be empirically supported, it is imperative that studies investigating the efficacy of the particular treatment are carefully evaluated. Stated differently, the efficacy of any treatment must be documented in carefully controlled empirical research that demonstrates the treatment in question is responsible for the beneficial effects observed, rather than other confounding variables or chance (see Campbell & Stanley, 1963; Kazdin, 1992). Within the literature, a number of criteria have been put forth to classify treatments as evidence-based (Chambless & Hollon, 1998; Kratochwill & Stoiber, 2002; Odom et al., 2005; WWC, 2017). Common to these criteria is assessing the quality of experimental studies employing randomized controlled trials (RCTs), single-case experimental designs (SCEDs), and meta-analyses. Any particular treatment may have an accumulation of evidence supporting its efficacy; however, if that evidence is derived from low-quality sources, the confidence a clinician has in recommending that treatment might be truncated. In the following section, we introduce the aforementioned experimental methodologies (i.e., RCTs, SCEDs, and meta-analyses), the various quality assessments linked to each design, and general criteria outlining the amount of research required for a treatment to be considered an EST.

Randomized Controlled Trials

Randomized controlled trials (RCTs) are experimental group designs that are largely regarded as the "gold standard" of clinical research (Schulz, Altman, & Moher, 2010a, 2010b; Sturmey, 2014). In RCTs, participants are randomly assigned to treatment and control groups. This random assignment is done to ensure that any confounding variables that may influence the outcome of the study are equally dispersed across groups. In the treatment group, participants receive the treatment under investigation (i.e., independent variable), whereas in the control group, participants receive no treatment, an alternative treatment, or a placebo. Then, at specified times during the study, quantifiable data are collected on the participant variable (i.e., dependent variables) and compared across groups. In general, RCTs are highly regarded in clinical research because (1) they carefully control for confounding variables that may threaten internal validity and (2) large groups of participants undergo random assignment, which enhances external validity. In other words, treatment effects derived from a well-designed RCT can be attributed to the treatment under investigation and extended to the larger population. A detailed analysis of RCTs and other group designs go beyond the purpose of this chapter but are available from other sources (Machin & Fayers, 2010; Torgerson & Torgerson, 2008).

Although RCTs have a sound methodological foundation and can produce empirical evidence regarding the effectiveness of a particular treatment, not all RCTs are conducted with the same level of experimental rigor (Grossman & Mackenzie, 2005). Therefore, there are a number of methodological considerations that need to be accounted for when conducting and reporting a RCT, some of which may be difficult to achieve. For instance, Nichol, Bailey, and Cooper (2010) outlined a number of challenges with employing RCTs: (1) unclear hypotheses and multiple objectives, (2) poor selection of endpoints, (3) inappropriate subject selection criteria, (4) non-clinically relevant or feasible treatments, (5) inadequate randomization, stratification, and blinding, (6) insufficient sample size, (7) failure to use intention-to-treat analysis, and (8) failure to anticipate common practical constraints. Although we will not detail each of these potential issues, it is important to acknowledge that even though RCTs are highly regarded in terms of experimental design, the quality of an RCT can vary widely and dramatically impact the level of evidence provided.

Given the number of potential issues with conducting RCTs, it is crucial that studies be adequately described to allow for objective quality assessment. That is, there must be clear descriptions of the methodologies employed and findings obtained in order to assess the quality of the study and determine the level of evidence it provides for a particular treatment. Without such technical information, many flaws in a study may be masked or overlooked by reviewers. To ensure that RCTs receive adequate and objective quality assessment, it was necessary to standardize the manner in which RCTs are reported.

Accordingly, the Consolidated Standards of Reporting Trials (CONSORT standards) were developed and described by Schulz, Altman, Moher, and the CONSORT group in 2010. The primary aim of the CONSORT standards is to ensure that adequate information is reported to allow for a quality evaluation. A total of 25 standards are included in the CONSORT criteria. As an example, Standard 5 states that treatments for each group are described with sufficient detail to allow for replication, including how and when the treatments were administered. We will not be discussing each standard here but want to highlight the importance of these standards as they provide guidelines for how to report RCTs, allowing for quality assessment. Ultimately, this provides an objective basis to determine whether or not a treatment should be considered "evidence based."

Within the educational literature, and as an example of RCT quality assessment, WWC

(2017) developed criteria for evaluating individual studies. First, studies are screened to determine if the study is relevant to the literature base on the treatment in question. Second, the measurement system employed in the study is examined to ensure the researchers selected and used an appropriate (i.e., reliable and valid) measurement system. Third, the design of the study (e.g., RCT) and sample (e.g., participants; sample size) used in the study are assessed. Fourth, baseline data (if obtained) are examined looking for equivalence across treatment and control group for the dependent variables. Then finally, the analyses performed (e.g., statistical analyses) are assessed for appropriateness. Please see *WWC Study Review Guide, Group Design Studies* (2018) for a more detailed description of the evaluation process.

Single-Case Experimental Designs

Single-case experimental designs (SCEDs) have a long history of scientific merit and provide empirical evidence at the level of the individual. Although a thorough discussion of SCEDs is beyond the scope of this chapter, a number of detailed discussions are available elsewhere (DeRosa, Sullivan, Roane, & Kadey, in press; Kratochwill & Levin, 2014; Perone & Hursh, 2013). Like RCTs, SCEDs are experimental in nature. The goal of any SCED is to demonstrate a causal relationship between the independent variable (e.g., treatment) and dependent variable (e.g., behavior targeted for change). This causal, or functional, relation relies on the demonstration of experimental control. When the introduction of the independent variable occasions a change in the dependent variable, and the effect is reliably replicated within and/or across participants, experimental control is demonstrated. By making both within- and between-subject comparisons, threats to internal validity are controlled for, while systematic replication of the effects bolsters external validity (Martella, Nelson, & Marchand-Martella, 1999).

Like all experimental designs, the quality of a SCED also varies and requires quality assessment to determine the level of evidence provided. To assess the level of evidence presented in SCEDs, Horner et al. (2005) established specific criteria. That is, for a treatment to be considered evidence-based derived from SCEDs, five standards need to be met: (1) the treatment is operationally defined, (2) the setting in which the treatment is to be used is defined, (3) the treatment is implemented with fidelity, (4) demonstration of a functional relation between the treatment and a change in behavior, and (5) the experimental effects are replicated across a sufficient number of studies, researchers, and participants. In general, Horner and colleagues suggested that in order for a treatment to be evidence based, a series of experimental studies need to be conducted that demonstrate an effect by the treatment on behavior under specified conditions (i.e., setting and procedures). For a more detailed discussion of these standards please see Horner et al. (2005) or WWC (2017) for SCED standards used in the educational literature.

Meta-analyses

Meta-analyses are an integral part of EBP as they offer an approach to aggregate the results of multiple studies and synthesize their collective findings. Meta-analyses are based on a family of statistical procedures that combine data from multiple studies and calculate average effect sizes. An effect size is a statistical metric that estimates the magnitude of effect the independent variable has on the dependent variable. Stated differently, an effect size estimates how much of an effect the treatment had, rather than simply did it have an effect greater than what would be expected by chance (i.e., p-value).

Meta-analytic strategies have been developed for both RCTs and SCEDs. In the case of RCTs, the most commonly used effect size is Cohen's d, which standardizes differences between group means (i.e., treatment group vs. control group) expressed as a z-score. However, for SCED, Cohen's d is inappropriate as it relies on group differences, which SCEDs inherently do not provide and because SCED data do not meet many of the basic assumptions needed to conduct parametric analyses (e.g., homoscedasticity; Burns, 2012). For these reasons researchers have developed a family of nonparametric effect sizes for use with single-case data, for example, percentage of nonoverlapping data (Scruggs, Mastropieri, & Castro, 1987), percentage of all nonoverlapping data (Parker, Hagan-Burke, & Vannest, 2007), and nonoverlap of all pairs (Parker & Vannest, 2009), to name a few. Similar to RCTs, meta-analytic researchers utilize these nonparametric effect sizes to synthesize the results of numerous studies simultaneously. Thus, meta-analyses are well-suited for questions regarding a particular treatment's evidence base as they can simultaneously synthesize and analyze results from numerous studies employing various types of experimental designs. For a comprehensive review of meta-analytic strategies, please see Faith, Allison, and Gorman (Faith, Allison, & Gorman, 1996) and Schmidt and Hunter (Schmidt & Hunter, 2014).

One method for assessing the quality of individual studies, which may be included in a meta-analysis, is the *Scientific Merit Rating Scale* (SMRS), developed by the National Center on Autism's *National Standards Project* (National Autism Center, 2009). The SMRS checklist uses a 5-point rating scale to evaluate study quality across five domains. First, the research design is considered in terms of the degree to which experimental control is demonstrated and design employed. Second, a rating is given regarding the extent to which reliable and valid measurement of the dependent variable took place. Third, procedural integrity is reviewed to determine if the independent variable was implemented as intended. Fourth, participant characteristics are reviewed to determine if the treatment was evaluated within the appropriate population. And fifth, a rating is given on generalization or the extent to which the study attempted to objectively demonstrate that treatment effects occur across time, setting, stimuli, and responses.

Another method to assess the quality of studies included in meta-analyses was developed by Guyatt et al. (2008) termed the "grading of recommendation assessment, development, and evaluation" (GRADE) system. The GRADE system first assesses the quality of study design, with RCTs starting as "high evidence" and observational studies or SCEDs as "low evidence." From there, five factors may result in a study's quality of evidence being downgraded: (1) study limitations, (2) inconsistency of results, (3) indirectness of evidence, (4) imprecision, and (5) publication bias. In opposition, there are three factors that may improve the quality of evidence provided by a particular study: (1) large magnitude of effect, (2) dose response, and (3) confounding variables likely minimize the effect.

After individual studies have been assessed, the GRADE system applies a broad classification system to the entire body of work on a particular treatment across four levels: "(1) High quality—further research is very unlikely to change confidence in the estimate of effect, (2) Moderate quality—Further research is likely to have an important impact on our confidence in the estimate of effect and may change the estimate, (3) Low quality—Further research is very likely to have an important impact on our confidence in the estimate of effect and is likely to change the estimate, and (4) Very low quality—Any estimate of effect is uncertain" (Guyatt et al., 2008, p. 926). This leads to an overall rating of the quality of evidence provided across studies related to a particular treatment. The GRADE system then outlines how to decide whether the treatment in question should be recommended based on (a) quality of evidence, (b) cost/benefit analyses, and (c) patient or client values and preferences. For a more complete discussion, please refer to Guyatt et al.

Amount of Evidence Required for EBP

Although high-quality empirical research is needed in order for a treatment to be considered evidence based, one perfectly conducted study does not suffice. For a treatment to be considered evidence based, there not only has to be one quality empirical investigation demonstrating its effectiveness, there must be multiple experimental demonstrations confirming the treatments efficacy.

Horner et al. (2005) proposed that five well-designed single-case studies, conducted by at least three different research groups, across three different geographical locations, with a total of at least 20 participants, are needed to establish a treatment as evidence based. The WWC (2017) also set forth criteria to categorize treatments based on the amount of evidence in the literature. More specifically, the "extent of evidence" for a particular treatment is based on the number and sizes of the studies. A "small" amount of evidence is defined as either one study, one school, or less than 350 participants. Whereas a "medium to large" amount of evidence consists of more than one study, more than one school, and at least 350 participants. The purpose of criteria like these is used to give an indication to the practitioner of how generalizable the research findings are for a given treatment.

Similarly, the National Autism Center (2009) developed a classification system used to describe the strength of evidence available for a particular treatment. This classification system divided the level of evidence provided for a particular treatment into four categories: established, emerging, unestablished, and ineffective/harmful. An established treatment, based on the National Autism Center's criteria, has been documented in either two group experiments with at least 12 participants that display no conflicting results or three group experiments or six, single-case experiments with at least 18 participants with no more than one study that found conflicting results. Additionally, all studies need to have received high ratings (i.e., 3 or greater) on the SMRS to be considered established treatment. Table 27.1 provides an outline of the National Autism Center's criteria, and Table 27.2 provides an example of how these criteria have been applied to behavioral treatments for individuals with autism.

Barriers to Evidence-Based Practice

Despite the breadth of EBP, selecting an appropriate treatment can be met with challenges for several reasons. Differences in terminology used across disciplines, agencies, and organizations are one barrier to an evidence-based approach. Terms that are used interchangeably include

Table 27.1 A summary of the four levels of evidence from the National Autism Center's (2009) *National Standards Project*

Level of evidence	Definition
Established	1. Two group experiments or four single-case experiments with at least 12 participants without conflicting results; three group experiments or six single-case experiments with a minimum of 18 participants with no more than one study presenting conflicting results 2. Had SMRS scores of 3 or greater 3. Reported beneficial treatment effects for specific targets 4. These results may be supplemented by other lower-quality studies
Emerging	1. One group experiment or two single-case experiments with a minimum of six participants without conflicting results 2. Has SMRS scores of 2 3. Reported beneficial treatment effects on one dependent variable for a specific target 4. These studies may be supplemented by those with higher or lower SMRS scores
Unestablished	1. May or may not be based on research 2. Had beneficial effects reported on poorly controlled studies with SMRS scores of 0 or 1 3. Have claims based on testimonials, opinions, or speculation 4. Were ineffective, unknown, or adverse treatment effects based on poorly controlled studies
Ineffective/harmful	1. Had two group experiments or four single-case experiments with at least 12 participants without conflicting results; three group experiments or six single-case experiments with a minimum of 18 participants with no more than one study presenting conflicting results 2. Had SMRS scores of at least 3 3. No beneficial treatment effects for one dependent variable for a specific target or had adverse treatment effects on dependent variable

Adapted from the National Autism Center (2009).

Table 27.2 A summary of treatments for children with ASD under the age of 21 that meet three levels of evidence (Established, Emerging, and Unestablished) from the National Autism Center's (2015) *National Standards Project*

Level of evidence	Treatments
Established	1. Behavioral treatments 2. Cognitive behavior treatment package 3. Comprehensive behavioral treatment for young children 4. Language training (production) 5. Modeling 6. Natural teaching strategies 7. Parent training 8. Peer training package 9. Pivotal response training 10. Schedules 11. Scripting 12. Self-management 13. Social skills package 14. Story-based treatment
Emerging	1. Augmentative and alternative communication devices 2. Developmental relationship-based treatment 3. Exercise 4. Functional communication training 5. Imitation-based treatment 6. Initiation training 7. Language training (production and understanding) 8. Massage therapy 9. Multicomponent package 10. Music therapy 11. Picture exchange communication system 12. Reductive package 13. Sign instruction 14. Social communication intervention 15. Structured teaching 16. Technology-based treatment 17. Theory of mind training
Unestablished	1. Animal-assisted therapy 2. Auditory integration training 3. Concept mapping 4. DIR/floor time 5. Facilitated communication 6. Gluten-free/casein-free diet 7. Movement-based treatment 8. SENSE Theatre treatment 9. Sensory treatment package 10. Shock therapy 11. Social behavioral learning strategy 12. Social cognition treatment 13. Social thinking treatment

Adapted from the National Autism Center (2015).

evidence-based practice, best practice, research-based practice, empirically supported treatments, empirically validated treatment, and scientifically supported treatments (DiGennaro-Reed & Reed, 2008). More specifically, many disciplines (e.g., psychology, medicine, public health) embrace EBP as a decision-making model that incorporates several components to providing appropriate care to consumers, including the identification and selection of ESTs. However, in other disciplines (e.g., education), EBP typically refers to specific interventions or instructional

Table 27.3 A summary of criteria for empirically supported treatments (Chambless & Hollon, 1998)

Level of evidence	Definition
Efficacious	1. Two between-group design experiments (RCTs), showing treatment is superior (based on statistical significance) to another treatment or placebo 2. Three single-case design experiments ($n > 9$), demonstrating experimental control, and compared to another treatment 3. Experimental procedures detailed 4. Participant characteristics specified 5. Effects demonstrated by at least two different research groups
Possibly efficacious	1. Two experiments showing treatment is superior (based on statistical significance) to a wait-list control 2. All experiments conducted by one research group 3. Single-case design experiment(s) ($n < 3$)

Adapted from Tolin, McKay, Forman, Klonsky, & Thombs (Tolin, McKay, Forman, Klonsky, & Thombs, 2015)

Table 27.4 A summary of treatments and level of evidence for the treatment of aggression for individuals with intellectual disabilities

Level of evidence	Treatments
Evidence-based treatment	**Behavioral treatments** • Functional communication training • Differential reinforcement procedures • Function-based reinforcement; mixed-treatment packages
Lacking sufficient evidence	**Behavioral treatments** • Antecedent manipulations and changes in context • Noncontingent reinforcement alone • Antecedent exercise • Response blocking • Contingent positive punishment • Self-monitoring **Other treatments** • Mindfulness • Teaching family model • Vibroacoustic music • Aromatherapy
Inconclusive evidence	Cognitive-behavioral therapy

The level of evidence is based on Chambless and Hollon's (1998) criteria

Adapted from Healy, O., Lydon, S. and Murray, C. (Healy, Lydon, & Murray, 2014). Aggressive Behavior. In Evidence-Based Practice and Intellectual Disabilities (eds P. Sturmey and R. Didden). doi:https://doi.org/10.1002/9781118326077.ch5

strategies that have a strong empirical base. Thus, the differences in terminology regarding EBP can pose challenges to obtaining appropriate care given a multidisciplinary team approach.

Variation in criteria, regarding what constitutes a treatment as empirically supported, is another challenge that interferes with accessing appropriate treatment for individuals with intellectual disabilities. That is, while the research base for a given treatment may be classified as efficacious or established given one set of criteria, evaluation given a different set of criteria may suggest that the treatment is not empirically supported. The National Autism Center's criteria (Table 27.1) and examples of application of the criteria (Table 27.2) were discussed earlier in this chapter. Tables 27.3 and 27.4 are provided for comparative purposes where Table 27.3 outlines Chambless and Hollon's (1998) criteria for categorizing "efficacious" and "possibly efficacious treatments" and Table 27.4 provides an example of the application of these criteria. Additionally, databases outlining ESTs may not be readily available to practitioners and educators, and resources, particularly funding, may be limited, thus restricting the ability to incorporate ESTs into every day practice (DiGennaro-Reed & Reed, 2008).

Finally, the lack of evidence on the implementation of EBP poses a significant barrier to accessing appropriate care. That is, evaluation of treatment integrity and progress monitoring are considered a part of EBP; however, there is limited guidance regarding how often and in what manner these measures should be taken to ensure the decision-making model is effective (Detrich, 2008; DiGennaro-Reed & Reed, 2008). Thus, the future direction of EBP for individuals with intellectual disabilities is evaluating the model itself to understand if efficient and effective outcomes are in fact being achieved.

Summary

The history of EBP has been quite impactful in improving outcomes for patients and students with intellectual disabilities. The EBP movement was integral in shifting the general perception and understanding of individuals with intellectual disabilities, such that these individuals can now more readily access the treatments needed to enhance their quality of life. Furthermore, rigorous treatment evaluation criteria support the relevance of obtaining and implementing research-based findings in the daily care of individuals with intellectual disabilities.

Despite the long-standing history of EBP and improved care for individuals across disciplines, limitations exist in the ability to consistently and effectively access needed treatments within and across populations. Thus, continued work is needed to ensure the effective implementation of EBP, as well as to refine the current model to best meet the needs of individuals with various presenting symptomology. Although the push toward EBP became prevalent in the 1960s, with the ideas being more readily disseminated into literature and practice in the 1990s, EBP has only become *essential* within the field of medicine, education, psychology, and other health-related disciplines (e.g., speech, public health) within the past two decades. Thus, although the work of Sackett and other pioneers of the EBP movement cannot be discounted, it is now integral to move beyond an understanding and basic application and begin to focus on the implementation and effectiveness of EBP for individuals with intellectual disabilities.

References

American Psychological Association. (1995). *Template for developing guidelines: Interventions for mental disorders and psychosocial aspects of physical disorders*. Washington, DC: Author.

American Psychological Association. (2006). Presidential task force on evidence-based practice: Evidence-based practice in psychology. *American Psychologist, 61*, 271–285.

Biglan, A., & Ogden, T. (2008). The evolution of evidence-based practice. *European Journal of Behavior Analysis., 9*, 81–95.

Burns, M. K. (2012). Meta-analysis of single-case design research: Introduction to the special issue. *Journal of Behavioral Education, 21*, 175–184.

Campbell, D. T., & Stanley, J. C. (1963). Experimental and quasi-experimental designs for research. In *Handbook of research on teaching*. Chicago, IL: Rand McNally.

Chambless, D. L., & Hollon, S. D. (1998). Defining empirically supported therapies. *Journal of Consulting and Clinical Psychology, 66*(1), 7–18.

Cook, B. G., Tankersley, M., & Harjusola-Webb, S. (2008). Evidence-based special education and professional wisdom: Putting it all together. *Intervention in School and Clinic, 44*, 105–111. https://doi.org/10.1177/1053451208321566

Council for Exceptional Children. (2006). *Evidence-based professional practices proposal*. Arlington, VA: Author.

Council for Exceptional Children. (2014). *Standards for evidence-based practices in special education*. Arlington, VA: Author.

Davidson, K. W., Trudeau, K. J., Ockene, J. K., Orleans, J. K., & Kaplan, R. M. (2003). A primer on evidence-based review systems and their implications for behavioral medicine. *Annals of Behavioral Medicine, 28*, 226–238.

DeRosa, N. M., Sullivan, W. E., Roane, H. S., & Kadey, H. J. (in press). Single-case experimental designs. In W. W. Fisher, C. C. Piazza, & H. S. Roane (Eds.), *Handbook of applied behavior analysis* (2nd ed.). New York, NY: Guilford.

Detrich, R. (2008). Evidence-based, empirically supported, or best practice? A guide for the scientist-practitioner. In J. K. Luiselli, D. C. Russo, W. P. Christian, & S. Wilczynski (Eds.), *Effective practices for children with autism: Educational and behavior support interventions that work*. New York: Oxford Press.

DiGennaro-Reed, F. D., & Reed, D. D. (2008). Towards an understanding of evidence-based practice. *Journal of Early and Intensive Behavior Treatment., 5*, 20–29.

Fagan, T. K., & Wise, P. S. (2000). *School psychology: Past, present, and future* (2nd ed.). Bethesda, MD: National Association of School Psychologists.

Faith, M. S., Allison, D. B., & Gorman, B. S. (1996). Meta-analysis of single-case research. In R. D. Franklin, D. B. Allison, & B. S. Groman (Eds.), Design and analysis of single-case research (pp. 267–300). New York, NY: Psychology Press.

Fisher, W., Piazza, C. C., Bowman, L. G., Hagopian, L. P., Owens, J. C., & Slevin, I. (1992). A comparison of two approaches for identifying reinforcers for persons with severe and profound disabilities. *Journal of Applied Behavior Analysis, 25*(2), 491–498.

Grossman, J., & Mackenzie, F. J. (2005). The randomized controlled trial: Gold standard, or merely standard? *Perspectives in Biology and Medicine, 48*(4), 516–534. Johns Hopkins University Press. Retrieved July 18, 2018, from Project MUSE database.

Guyatt, G. H., Oxman, A. D., Vist, G. E., Kunz, R., Falck-Ytter, Y., Alonso-Coello, P., & Schünemann, H. J. (2008). GRADE: An emerging consensus on rating quality of evidence and strength of recommendations. *British Medical Journal (Clinical Research Ed.), 336*(7650), 924–926.

Healy, O., Lydon, S., & Murray, C. (2014). Aggressive behavior. In P. Sturmey & R. Didden (Eds.), *Evidence-based practice and intellectual disabilities*. New York: John Wiley & Sons. https://doi.org/10.1002/9781118326077.ch5

Horner, R. H., Carr, E. G., Halle, J., McGee, G., Odom, S., & Wolery, M. (2005). The use of single-subject research to identify evidence-based practice in special education. *Exceptional Children, 71*(2), 165–179.

Individuals with Disabilities Education Improvement Act of 2004, H.R. 1350, 108th Congress.

Institute of Medicine. (2001). *Crossing the quality chasm: A new health system for the 21st century*. Washington, DC: National Academies Press.

Kazdin, A. E. (1992). *Methodological issues & strategies in clinical research* (1st ed.). Washington, DC: American Psychological Association.

Kratochwill, T. R., & Levin, J. R. (2014). In T. R. Kratochwill, J. R. Levin, T. R. Kratochwill, & J. R. Levin (Eds.),. Introduction: An overview of single-case treatment research *Single-case treatment research: Methodological and statistical advances*. Washington, DC: American Psychological Association. https://doi.org/10.1037/14376-001

Kratochwill, T. R., & Stoiber, K. C. (2002). Evidence-based treatments in school psychology: Conceptual foundations of the procedural and coding manual of division 16 and the society for the study of school psychology task force. *School Psychology Quarterly, 17*(4), 341–389. https://doi.org/10.1521/scpq.17.4.341.20872

Machin, D., & Fayers, P. M. (2010). *Randomized clinical trials: Design, practice and reporting*. New York: John Wiley & Sons.

Martella, R. C., Nelson, J. R., & Marchand-Martella, N. E. (1999). *Research methods: Learning to become a critical research consumer*. Madison, WI: Allyn & Bacon.

National Autism Center. (2009). *National standards project: Phase 1*. Randolph, MA: Author.

National Autism Center. (2015). *National standards project: Phase 2*. Randolph, MA: Author.

Nichol, A. D., Bailey, M., & Cooper, D. J. (2010). Challenging issues in randomised controlled trials. *Injury, 41*, S20–S23.

No Child Left Behind Act of 2001, P.L. 107-110, 20 U.S.C. § 6319 (2002).

Odom, S. L., Brantlinger, E., Gersten, R., Horner, R. H., Thompson, B., & Harris, K. R. (2005). Research in special education: Scientific methods and evidence-based practices. *Exceptional Children, 71*(2), 137–148. https://doi.org/10.1177/001440290507100201

Parker, R. I., Hagan-Burke, S., & Vannest, K. (2007). Percentage of all non-overlapping data (PAND): An alternative to PND. *The Journal of Special Education, 40*, 194–204.

Parker, R. I., & Vannest, K. (2009). An improved effect size for single-case research: Nonoverlap of all pairs. *Behavior Therapy, 40*, 357–367.

Perone, M., & Hursh, D. E. (2013). Single-case experimental designs. In G. J. Madden, W. V. Dube, T. D. Hackenberg, G. P. Hanley, K. A. Lattal, G. J. Madden, et al. (Eds.), *APA handbook of behavior analysis* (Methods and principles) (Vol. 1, pp. 107–126). Washington, DC: American Psychological Association. https://doi.org/10.1037/13937-005

Sackett, D. L., Rosenberg, W. M. C., Muir Gray, J. A., Haynes, R. B., & Richardson, W. S. (1996). Evidence-based medicine: What it is and what it isn't. *BMJ, 312*, 71–72. https://doi.org/10.1136/bmj.312.7023.71

Sackett, D., Strauss, S., Richardson, W., Rosenberg, W., & Haynes, R. (2000). *Evidence-based medicine: How to practice and teach EBM* (2nd ed.). Edinburgh: Churchill Livingstone.

Schmidt, F. L., & Hunter, J. E. (2014). *Methods of meta-analysis: Correcting error and bias in research findings*. Thousand Oaks, CA: Sage Publications.

Schulz, K. F., Altman, D. G., & Moher, D. (2010a). CONSORT 2010 statement: Updated guidelines for reporting parallel group randomised trials. *BMC Medicine, 8*(1), 18. https://doi.org/10.1186/1741-7015-8-18

Scruggs, T. E., Mastropieri, M. A., & Castro, G. (1987). The quantitative synthesis of single-subject research: Methodology and validation. *Remediate and Special Education, 8*, 24–33.

Schlosser, R. W., & Sigafoos, J. (2008). Identifying "evidence-based practice" versus "empirically supported treatment". *Evidence-Based Communication Assessment and Intervention, 2*, 61–62.

Schulz, K. F., Altman, D. G., & Moher, D. (2010b). CONSORT 2010 statement: Updated guidelines for reporting parallel group randomized trials. *BMJ, 23*(340), c332.

Spencer, T. D., Detrich, R., & Slocum, T. A. (2012). Evidence-based practice: A framework for making effective decisions. *Education and Treatment of Children, 35*, 127–151.

Sturmey, P. (2014). Evidence-based practice an introduction. In P. Sturmey & R. Didden (Eds.), *Evidence-based practice and intellectual disabilities* (pp. 3–28). New York: John Wiley & Sons.

Sur, R. L., & Dahm, P. (2011). History of evidence-based medicine. *Indian Journal of Urology., 27*, 487–489. https://doi.org/10.4103/0970-1591.91438

Swift, J. K., Callahan, J. L., & Vollmer, B. M. (2011). Preferences. In J. Norcross (Ed.), *Psychotherapy relationships that work: Evidence based responsiveness* (2nd ed., pp. 301–315). New York: Oxford University Press.

Thoma, A., & Eaves, F. F. (2015). A brief history of evidence-based medicine (EBM) and the contributions of Dr. David Sackett. *Aesthetic Surgery Journal., 35*, 261–263. https://doi.org/10.1093/asj/sjv130

Tolin, D. F., McKay, D., Forman, E. M., Klonsky, E. D., & Thombs, B. D. (2015). Empirically supported treatment: Recommendations for a new model. *Clinical Psychology: Science and Practice, 22*(4), 317–338. https://doi.org/10.1111/cpsp.12122

Torgerson, D. J., & Torgerson, C. J. (2008). *Designing randomised trials in health, education and the social sciences: An introduction*. New York: Springer.

What Works Clearinghouse. (2017). *Standards handbook, version 4.0*. Washington, DC: US Department of Education.

What Works Clearinghouse. (2018). Study review guide. In *Group design studies*. Washington, DC: US Department of Education.

Zimerman, A. L. (2013). Evidence-based medicine: A short history of a modern medical movement. *American Medical Association Journal of Ethics., 15*, 71–76.

28

Pseudoscientific Therapies for Autism Spectrum Disorder

Bruce A. Thyer

What Is Autism Spectrum Disorder?

The standard approach to diagnosing mental disorders in the United States, and widely used throughout the world, is an authoritative book called the *Diagnostic and Statistical Manual of Mental Disorders* (DSM, American Psychiatric Association 2013). Now in its fifth edition, the DSM has been plagued by controversy from its inception. Some issues deal with the minutia of the various signs and symptoms and duration presented by a given person needed to arrive at a particular diagnosis. Others are much more fundamental, such as serious conceptual problems with the very definition of what constitutes a mental disorder and of the validity of the various categories described. Some diagnoses come and go. The DSM-2 did not include homosexuality and/or autism. Later editions did. Now the DSM-5 does not include them but does claim that obstructive sleep apnea disorder (most commonly caused by being overweight) *is* a mental disorder, as is smoking, called tobacco use disorder. Persons with cognitive impairment due to traumatic brain injury are similarly asserted to suffer from a mental disorder. Some authorities assert that any condition with a clearly evident biological cause (e.g., genetic disease, brain trauma or infection, endocrine abnormality, etc.) should not be construed as a mental disorder at all and that this latter term should be reserved for conditions which have their etiology in the person's mind, not in demonstrable physical pathology of the body.

The phenomena called autism have similarly suffered at the hands of psychiatrists making up their "minds." The DSM-5 uses the term autism spectrum disorder, while previous editions used autistic disorder. The DSM-2 did not contain any diagnosis dealing with autism at all (APA, 1968). Asperger's disorder was found in earlier editions of the DSM, but it has been eliminated by the DSM-5. Now of course the actual disorders in behavior which have been used to label someone variously as autistic, autistic disorder, Asperger's disorder, or autism spectrum disorder do not ebb and flow at the whim of the latest edition of the DSM. The field of human psychopathology is much like the former planet Pluto. For years it was considered a true planet. Now it is not. Pluto the object, whatever we call it, remains unchanged. Similarly the condition called autism spectrum disorder is similarly unmoved, and behavioral scientists and other specialists continue to study persons with this condition, attempt to discover its causes, and develop effective treatments. This can be done regardless of the current favored terminology. Many disciplines do not make use of the DSM-5 system of diagnosis at all. Behavior

B. A. Thyer (✉)
College of Social Work, Florida State University, Tallahassee, FL, USA
e-mail: Bthyer@fsu.edu

J. L. Matson (ed.), *Handbook of Intellectual Disabilities*, Autism and Child Psychopathology Series, https://doi.org/10.1007/978-3-030-20843-1_28

analysts, for example, eschew global diagnostic labels in favor of focusing on specific behavioral excesses or deficits, and working directly on behavior change, usually through programs of reinforcement, shaping and extinction (punishment is very rarely used). This is a very successful approach to helping persons with ASD and does not require using the DSM-5.

The DSM-5 criteria for autism spectrum disorder (ASD) cover one and half pages, so just a brief summary will be given here:

> Autism spectrum disorder is characterized by persistent deficits in social communication and social interaction across multiple contexts, including deficits in social reciprocity, nonverbal communicative behaviors used for social interaction, and skills in developing, maintaining, and understanding relationships. In addition to the social communication deficits, the diagnosis of autism spectrum disorder requires the presence of restricted, repetitive patterns of behavior, interests or activities. (APA, 2013, p. 31)

ASD must appear fairly young in the child's life and is often, but not always, accompanied by intellectual disability. In severe ASD the person may be unable to talk or otherwise readily communicate and may display self-injurious behavior or motor stereotypes. The precise causes of ASD remain elusive. The DSM-5 notes that a proportion of ASD cases are associated with a known genetic mutation, but there is currently no genetic test used in practice which is of value in making the diagnosis. At present the diagnosis is arrived at clinically, via observation of the client (preferably over multiple contexts and times, supplemented with valid rating and observational scales), a careful history taking, and interviews with the patient (where possible), parents, and other caregivers. That a diagnosis is arrived at clinically is true for almost all the DSM-5 disorders—there are no legitimate genetic, brain scan, blood, tissue, or other tests which contribute to the diagnoses (except for the IQ tests used to diagnosis intellectual disability). Parents naturally want to know why their child acts the way they do, but the only correct answer at present is "We do not know what causes ASD."

Clinicians can be more confident in asserting what does *not cause* ASD, although many candidate etiological factors have been forward. Of most of these, the evidence is either solely anecdotal, too weak to draw scientifically legitimate conclusions from, or solidly pointing that given candidate causes are *not* responsible—such as sugar, food dyes, heavy metals, gluten or casein intolerance, deficiencies in blood flow to the brain, hormone imbalances (e.g., secretin), vitamin deficiencies, and parental child-caring practices. Our present inability to determine the causes of ASD is frustrating and perhaps hinders the development of treatments more effective than existing therapies. However, the precise etiology of a good many of the conditions found in the DSM is similarly vague, yet intervention research aimed at testing therapies proceeds unabated, and this includes treatments for ASD.

Treatment Versus Cure

When discussing therapies for ASD, it is important to distinguish between cures versus treatments. Many conditions, medical and psychosocial, have cures. Some form of intervention is provided for the clients, and following a course of therapy, the condition is gone. Antibiotics for bacterial infections are one example of a medical cure. Gradual real-life exposure therapy for clinical phobias is a curative example drawn from psychosocial conditions. However, for many conditions, complete cures remain elusive. Most therapies for ASD are efforts to enhance functional behavior, improving social skills, for example, or reading and conversational abilities. Other treatments are aimed at reducing problematic features of ASD, such as self-injurious behavior, movement stereotypies, and self-stimulating activities such as excessively spinning objects. Taken in isolation, these treatments may be beneficial, but you still have a person who displays many other features of ASD. Perhaps their talking is improved, but other behaviors typical of ASD remain. There are promising treatments which when offered very early in the child's life, last hours per day, over several years, have apparently resulted in children who become indistinguishable from normal and function well in school with normal intelligence (see Lovass, 1987), but access to such early intensive behavioral interventions (EIBI) remains quite limited. The US Food and Drug Administration is very clear on this issue: “One thing that it is important to know about autism up front: There is no cure for autism. So, products or treatments claiming to ‘cure’ autism do not work as claimed” (see https://www.fda.gov/ForConsumers/ConsumerUpdates/ucm394757.htm).

What Are Research-Supported Therapies?

For a treatment for ASD to be considered scientific, it should be supported by a substantial amount of empirical research that has been published in quality peer-reviewed journals, with the large preponderance of the evidence indicating that the treatment is, first of all, safe. Next it is important to demonstrate that the treatment is better than doing nothing. The third is that the treatment yields results that are considerably better than those achieved by providing a credible placebo treatment to the client. The fourth is that the new treatment produces results at least equivalent to a therapy which is already considered to be effective. The fifth is that the positive results of the new treatment are maintained over time (ideally months or years). The sixth is that the new treatment does not produce unpleasant side effects (this is related to but not the same as safety). And the seventh is that the new treatment is broadly effective across a wide range of clients, boys and girls, people of different races and ethnicities, etc. To achieve these benchmarks for a novel therapy for persons with ASD requires a significant, long-term period of intensive investigations, ideally conducted by researchers independent of the inventor of the new therapy, and with no significant financial or other potential conflicts of interest which could bias the results of the investigators (e.g., they market a competing form of treatment). To accomplish these tasks, clinical researchers in ASD employ various safeguard in their research. For example, persons being considered for potential inclusion in the treatment study are carefully assessed by expert diagnosticians so we can be sure the clients are really genuine instances of ASD. Assessments of client functioning and behavior are conducted by persons independent of those delivering treatments and, ideally, are unaware of what treatment the client they are assessing will receive or has received. The therapy is delivered via some structured protocol or treatment manual, to ensure faithful adherence to the principles of the intervention. A test evaluating a treatment which was sloppily administered is not a fair evaluation of that therapy. Ideally clients who meet a study’s inclusionary criteria are randomly assigned to the new or experimental treatment and to various control conditions (e.g., no-treatment condition, waiting-list or delayed therapy condition, alternative accepted treatment, placebo treatment). This helps insure that the two or more groups of clients are genuinely similar prior to receiving treatment. If this is done, and the two or more groups are different after treatment, then these differences can be attributed to the various treatment conditions the clients received and not to spurious factors such as the passage of time, placebo influences, therapist biases, etc. Even then, it is desirable that a given new treatment which passes all these hurdles and is seemingly effective be tested *again*, by different researchers in other settings, at least once and preferably more than one more time.

Studies like this are called randomized experiments and, when applied to persons with various medical or psychosocial disorders, are given the further term of randomized clinical trial, or RCT. RCTs are generally considered to be a very credible form of evidence needed to draw legitimate conclusions about the effects of a new treatment. When such treatments are repeatedly tested in RCTs and published in scientific journals, a substantial body of literature can accumulate regarding a given newer treatment, and the results of many such RCTs can be combined and the overall results summarized. A statistical tool to do this is called a meta-analysis (MA), and in a proper MA, the various studies’ strengths and limitations can be taken into account when arriving at a conclusion. For example, a RCT study with a sample size of 200 clients would be afforded more weight in the MA calculations than one involving only 50 persons. By combining mathematically all the results obtained in the various studies, one can get a better estimate of the “true” effects of a given therapy for ASD.

A more elaborate method of combining the results of many studies on a given therapy is called a *systematic review* (SR). Systematic reviews are usually conducted by interdisciplinary treatment teams, and very diligent steps are taken to locate *all* available studies which have evaluated a given therapy. This may include studies that were published in international journals, and perhaps unpublished doctoral dissertations, and even unpublished reports. Studies are independently graded for their overall quality and methodological rigor, and the conclusions from the stronger studies are given more weight than those from weaker ones. Most SRs incorporate meta-analysis into their methodology. The best SRs are produced by two major groups, the Cochrane Collaboration (www.cochrane.org) and the Campbell Collaboration (www.campbellcollaboration.org), and very careful methodological checks and controls are used to reduce bias as much as possible. For example, completed draft reports of SRs are independently reviewed by two or more experts and suggestions made to improve it. One can search the web-based libraries of Cochrane and Campbell for completed SRs dealing with the topic of ASD.

The above processes reflect some of the practices of legitimate *scientific inquiry*, and this approach can generally be labeled *between-subject designs* because comparisons are made between groups of clients. There are other credible paths which can be used however to evaluate treatments. One major alternative is called within-subject designs. In this approach, instead of studying changes on average scores of functioning between groups exposed to different treatments, and analyzing the results using complex statistical analysis, the clinical researcher studies only one client and take multiple measures of their functioning over time prior to a new intervention being applied. Then the same measurement strategy is used to assess client functioning while the new treatment is being applied and perhaps after it is removed. The results are usually depicted via line graphs and interpreted visually, not using statistics. Just as one can look at a graph of how the stock market has performed during the past 2 weeks, one can look at a graph of client functioning. If the pretreatment data (the baseline) are stable and then after treatment is introduced, client functioning dramatically improves, and this is very evident by simply looking at the graph; one has tentative evidence the treatment produced a favorable effect. If the baseline was lengthy and stable, and substantial behavior changes occurred immediately after treatment began, this makes for more robust causal inference than if changes were small or appreciably delayed after treatment began. There are various approaches to strengthening the internal validity (our ability to make valid casual inferences about the effects of the treatment under investigation) of within-subject designs. One way is to deliberately remove and then restore a therapy with short-term effects. If improvement follows the introduction of therapy, and deterioration follows the withdrawal of treatment (and is followed by improvement when treatment is reinstituted), then the potential for causal inference is high (“Yes! Treatment caused those improvements!”). Another approach is to track the functioning of several clients with the same problem and introduce treatment to one but not the others, then to the others sequentially. If the first client who received treatment improved and the others did not, until they too received treatment, causal inferences can also be strongly enhanced. A further variant is called the $N = 1$ randomized trial, which may be possible to undertake when a client is receiving a short-lived treatment. The client’s functioning is baselined, using a valid and reliable measure (or several), and then one of two treatments is randomly allocated to be given that day. In the case of medications, it may be possible to use a blinded approach wherein the caregiver or clinician does not know if they are administering the active drug or a placebo pill. The treatment allocated is determined each day by a coin toss. Functioning is recorded for a number of days, and then the blinding is broken. The data from days the client received the active drug are compared with the days when he/she received placebo or a different medication. If the two treatments exerted differential effects, this will be clearly evident from the graphed data and an informed decision then made about what course of therapy to provide over a longer term. Certain psychosocial interventions can also be evaluated in this manner.

Within-subject designs have been used in many disciplines for a long time. The classic text *An Introduction to the Principles of Experimental Medicine* by Claude Bernard (1865/1927) described their use, and the Russian physiologist Ivan Pavlov used them extensively in his research and was awarded the Nobel Prize for his discoveries. Like between-subject designs, within-subject designs require the use of reliable and valid measures of the client's behavior, except instead of measuring behavior just a few times across many people, as in controlled before and after RCT studies, within-subject studies take many measures before and after treatment from one person. This permits a more fine-grained and detailed analysis of individual change and represents an investigatory methodology that does not require expensive grant funding. Indeed, this is a major strength of within-subject studies in that they can be incorporated into everyday clinical practice aimed at helping persons with ASD. This can and is being done but not widely (Nikles & Springer, 2015; Schork, 2015). Like between-subject designs, within-subject designs are also amenable to having their results aggregated using meta-analysis procedures. See Bellini and Akullian (2007) for an example of this. One of the founders of the evidence-based practice movement has stated that certain forms of within-subject designs are more useful for making decisions in clinical practice about the care of individual patients than are RCTs and SRs (Guyatt, Rennie, Meade & Cook, 2002). Thus there are two major pathways to making legitimate conclusions about the effects of various treatments applied to persons with ASD, between-subject designs and within-subject studies. Each has their merits and limitations.

Legitimate scientific research emphasizes the publication of results of therapeutic trials in professional journals that make use of blind peer review and accords higher weight to such studies than information disseminated via other outlets such as press releases, television talk shows, the Internet, news briefings, reports available on websites or internally produced by a treatment's developers, testimonials, and so forth. Usually an article submitted for possible publication in a professional is sent to two or more reviewers, persons with expertise in the subject matter of the paper. The reviewers do not know the authors' names, gender, or affiliations. They provide a detailed critical appraisal of the research and a recommendation to the journal's editor to accept the work, to reject it, or to suggest that it be revised by the author and resubmitted. This process of blind peer review helps ensure that high-quality work is accepted and low-quality work is not published, at least within that particular journal. This screening is why journal publications are accorded greater credibility than other means of disseminating information.

Another feature of legitimate science is its openness to being disappointed. As Thomas Huxley said "The great tragedy of science - The slaying of a beautiful hypothesis by an ugly fact." Good research on a treatment for ASD does *not* set out to prove that it works. It adopts a more neutral attitude and tries to see what the effects of the treatment *are* and is equally open to learning that the treatment was effective or that it was not. In fact good science bends over backward to prove that a treatment *does not work*! For example, if a group of kids with ASD had their functioning rated by their parents, and the kids then received a treatment which was followed by apparent improvements, rather than taking this for strong proof that the treatment worked, the researchers would design a stronger study, better capable showing that it did *not* really work. This next study might use a no-treatment control group—some kids got the treatment and others did not. If the treated group got better, their results would be compared with those of the group that did not get treatment. If the treated group was no better off than the untreated group, what would that tell the researchers about the value of their therapy? It did not work! This would perhaps be personally disappointing, but it would be better to conduct such a study, and screen out an ineffective therapy, before releasing it to the public for use with kids with ASD. Or another study might use independent observers, not the parents, to rate the kids' functioning, so as to reduce potential biases introduced by parental expectancies. Hence all the successive levels of controls introduced in good-quality RCTs help better demonstrate that something *does not work*. Through progressively more stringent investigations, as a new therapy passes each "test," we can become a little more confident that it really may work, better than the passage of time, better than placebo, etc. Then we hold our breath, so to speak, to see if others can successfully replicate our positive results. If this happens once, then again, and more times, by independent researchers around the world, we grow progressively more and more optimistic that the new treatment is genuinely helpful.

What Are Pseudoscientific Therapies?

Pseudoscience has been defined as "a body of beliefs and practices whose practitioners wish, naively or maliciously, pass for science although it is alien to the approach, the techniques and the fund of knowledge of science" (Bunge, 1998, p. 41). For a variety of reasons, the field of ASD lends itself to being vulnerable to a variety of pseudoscientific claims pertaining to the presumed etiology of the condition, methods of assessing persons with ASD, and most dangerously, methods of treating the condition. Here is how one set of writers explained this vulnerability to pseudoscience:

> "Parents are typically highly motivated to attempt any promising treating, rendering them vulnerable to promising 'cures'. The unremarkable physical appearance of autistic children may contribute to the proliferation of pseudoscientific treatments and theories of etiology. Autistic children typically appear entirely normal; in fact many of these children are strikingly attractive…The normal appearance of autistic children may lead parents, caretakers and teachers to become convinced that there must be a completely 'normal' or 'intact' child lurking inside the normal exterior…the course can vary considerably among individuals…there is a great deal of variability in response to treatments…persons with autism sometime show apparently spontaneous developmental gains or symptom improvement in a particular area for unidentified reasons. If any intervention has recently been implemented, such improvement can be erroneously attributed to the treatment, even when the treatment is actually ineffective. In sum, autism's pervasive impact on development and functioning, heterogeneity with respect to course and treatment response, and current lack of curative treatments, render the disorder fertile ground for quackery." (Herbert, Sharp, & Gaudiano, 2002, p. 24)

What are some of the features of pseudoscientific therapies? Typically these therapies are claimed to be highly effective but lack the robust evidentiary support of scientifically legitimate treatments. Sometimes the level of evidence is solely that of personal anecdote or one individual's (often a parent) experience with something. The claim is made that removing something from the diet (e.g., sugar, gluten, particular food dyes, etc.) of the person with ASD was followed by remarkable improvements. Or that adding something to their diet produced equally miraculous positive changes. Many times some form of therapy (e.g., facilitated communication) was administered which seemingly resulted in amazingly enhanced functioning, and those responsible for the care of the person with ASD make it their personal mission to promote this new miracle cure. Here is an example of the latter type of claim:

"Kalel Santiago…was diagnosed with severe, non-verbal autism. At age 9, he hadn't spoken his first word—until his parents tried a controversial treatment… a hemp oil that includes a form of cannabis. Two days later, he was speaking. Kalel's parents sprayed the compound cannabidiol (CBD) in the boy's mouth twice per day, and they say the results were astounding. "He surprised us in school by saying the vowels A-E-I-O-U. It was the first time ever," dad Abiel told Yahoo Parenting. "You can't imagine the emotion we had, hearing Kalel's voice for the first time. It was amazing. The teacher recorded him and sent it to my wife and me, and we said, 'well, the only different thing we have been doing is using the CBD.'" Soon he was saying full words. "He said, 'amo mi mama,' 'I love my mom,'" Abiel says. "I don't know how to thank [the CBD oil makers]." (https://www.parenting.com/news-break/cannabis-spray-treatment-helps-9-year-old-autism-learn-to-speak)

Anecdotes like this, especially if repeated, can be the focus of popular magazine articles, which spark a flurry of interest resulting in parent purchasing the latest remedy du jour (see for example, Borchardt, 2015), aided and abetted by companies which manufacture and sell the new purported cure for a profit. A variation on the use of testimonials from parents are assertions from various well known respected figures such as celebrities or from 'Doctors'. These people look into the camera and say, in effect, "Take it from me, this treatment really works." This line of persuasion relies on 'authority', sometimes augmented by the accoutrements of the white coat and stethoscope.

Another characteristic of pseudoscientific treatments is the use of jargon, neologisms, or simply nonsensical words of phrases to explain the effects of the new therapy, or the invocation of mysterious forces or energies heretofore unknown to mainstream science. Such language affords the patina of real scientific explanations and may deceive the uninitiated, but legitimately trained researchers will recognize a bogus argument right off the bat. Here are some additional hallmarks of pseudoscience;

- "Exploited expertise—A genuine expert in one field provides testimonials in an area outside the expert's area.
- Bogus expertise: a supposed expert claims to possess scientific or practice credentials that are simply false, or originated from diploma mills or otherwise unaccredited universities (see Thyer, 2019).
- Financial conflicts of interest: The promoters of the new ASD therapy have a financial investment in the success of the treatment.
- Inflated research support: Exaggerated claims are made on the basis of poorly designed or conducted research, or research published in journals with very low scientific standards.
- Misleading research support: Findings from a well done study are misrepresented or outright erroneously reported.
- False research support: A study is published in a scientific journal but the actual study was never conducted. Sometime even highly respected journals get hoaxed by unscrupulous authors, some of whom are even doctors!
- Grandiose claims: The new ASD treatment is said to be curative, or amazingly effective for a wide array of signs and symptoms.
- Claims of a 'Quick Fix': The ASD treatment is said to produce improvements very rapidly.
- Implausible mechanism of action: The new treatment is said to work via processes which seem highly unlikely, given what is known about ASD. For example, the treatment called Facilitated Communication is based on the premise that persons with ASD are of normal intelligence, but are trapped in bodies that do not work properly or permit them to speak. There is no evidence that this is true. Hyperbaric oxygen therapy is premised on the idea that the brains of persons with ASD suffer from enduring levels of oxygen deprivation, or of reduced blood flow to the brain. There is no evidence that this is true.
- Claims that the new ASD therapy is rejected by the mainstream treatment community because it threatens vested interests, those who may not want to see the new highly

effective treatment promoted, and therefore try and suppress it.

- The claim being made is virtually impossible to test, for example, the assertion that ASD is caused by unknown allergies. When test after test fails to reveal a putative allergen, or one dietary restriction after another fails to help the person with ASD, the fault is not with the implausible theory, but is simply that the correct test or food causing ASD has not yet been identified. Parents and caregivers may spend a fortune vainly seeking the Holy Grail of allergens." (this section in quotation marks was adapted from Hupp, Mercer, Thyer, & Pignotti, 2019).

Having provided some hallmarks of potential pseudoscientific treatments for ASD, let us examine a few by name in the following section."

Selected Examples of Pseudoscientific Treatments for ASD

Camel's Milk: Yes, some researchers have suggested camel's milk as a treatment for ASD (e.g., Al-Ayadhi & Elamin, 2013). Before rushing out to the nearest camel dairy, one should read the conclusion of a systematic review on the topic. "Based on the evidence, camel milk should not replace standard therapies for any indication in humans" (Mihic, Rainkie, Wilby, & Pawluk, 2016).

Intravenous Secretin: In 1998, Horvath et al. published a study involving three children with ASD who were administered intravenous secretin, and they claimed, with no quantitative data, that the children had improved eye contact, expansion of expressive language, and heightened alertness. Since that time a number of studies have investigated the effects of secretin, and in 2012 a comprehensive systematic review sponsored by the Cochrane Collaboration was published examining the effects of secretin on ASD. The conclusion?

"There is no evidence that single or multiple dose intravenous secretin is effective and as such currently it should not be recommended or administered as a treatment for ASD" (see https://www.cochranelibrary.com/cdsr/doi/10.1002/14651858.CD003495.pub3/full?highlightAbstract=secretin)

Omega-3 Fatty Acids Supplementation: It has been suggested that children with ASD suffer from insufficient omega-3 fatty acids and that providing dietary supplements of this product could reduce the signs and symptoms of ASD. One early case report on using this treatment was published by Johnson and Hollander (2003) and was followed by a number of other published studies. These studies were collected and analyzed in a systematic review supported by the Cochrane Collaboration in 2011. The overall finding? "To date there is no high quality evidence that omega-3 fatty acids supplementation is effective for improving core and associated symptoms of ASD" (see https://www.cochranelibrary.com/cdsr/doi/10.1002/14651858.CD007992.pub2/full?highlightAbstract=autism).

Chelation: Chelation is a form of treatment which is said to remove stored or circulating stored toxic metals (e.g., lead, mercury), with these metals being the etiological agent causing ASD. With this theory in mind, some practitioners concluded that chelation medical therapies which removed these metals from the body of the child with ASD would help reduce signs and symptoms. This is done via administering a chelating substance which binds to heavy metals to the patient, intravenously, by mouth, or by injection, with the metals subsequently being excreted via urine. The procedure is not without risks, as harmful effects and deaths have been reported. Chelation therapy for ASD was the subject of a 2015 Cochrane-sponsored systematic review. The conclusion?—"The quality of the evidence is poor. Only one trial was included in this review, and we judged it to have high or uncertain risk of bias and methodological problems that limited the interpretation of outcomes presented. Given the deleterious effects of chelation, misinterpretation and misuse of the study of Adams et al. to justify the use of chelation for ASD is unethical and potentially places children unnecessarily in harm's way. Moreover, if these findings are in fact valid, they actually undermine the heavy metal toxicity theory and the rationale for chelation treatment, suggesting that it should not be used in the first place" (cited from https://www.cochranelibrary.com/cdsr/doi/10.1002/14651858.CD010766.pub2/full?highlightAbstract=autism).

If the theory is heavy metals cause autism, heavy metals are removed through chelation therapy, and the child does not improve, this tends to falsify the underlying theory of this treatment. If the theory is incorrect, continuing to deliver treatment based on a demonstrably false theory is unlikely to prove helpful, hence the suggestion by the authors of the above SR that chelation should not even be attempted.

Acupuncture: Acupuncture is based on a theory that the human body is surrounded by an invisible energy field undetectable to science (thus far). These lines of energy intersect at various places on the human body, points called meridians, and that human physical illness and emotional distress are caused by blockages in the free flow of this energy, sometimes called qi or chi. By inserting small needles under the skin at precise meridian positions, it is said that the proper flow of qi can resume and health restored. Variations of conventional acupuncture include pressing or tapping on these meridians (acupressure), passing small amounts of electric current through the needles, twirling the inserted needles, and holding smoldering bundles of herbs near the meridians (called moxibustion). An even more implausible form of acupuncture is called Tong Ren, which involves the Tong Ren practitioner holding a plastic doll with the meridian points depicted on it, and pounding these points with a small magnetic hammer, while thinking of the patient. This is said to produce healing in the distant patient, who may not even be aware that he/she is being remotely treated. Tong Ren is being advertised as a legitimate therapy for persons with ASD (see https://theory.yinyanghouse.com/treatments/tongren_for_autism).

In 2011, the Cochrane Collaboration published a systematic review of the effects of conventional acupuncture for ASD. The conclusion?—"Current evidence does not support the use of acupuncture for treatment of ASD. There is no conclusive evidence that acupuncture is effective for treatment of ASD in children and no RCTs have been carried out with adults" (cited from https://www.cochranelibrary.com/cdsr/doi/10.1002/14651858.CD007849.pub2/full?highlightAbstract=autism).

For clinical researchers, acupuncture lends itself very nicely to placebo-controlled trials, wherein some patients receive "real" acupuncture, with the needles being placed in the theoretically correct positions, and other randomly assigned to have the needles placed in meridians that are deliberately chosen because they are incorrect for the condition being treated, or by randomly placing the needles on spots *not* said to be meridians. For many conditions, when real acupuncture has been compared to sham acupuncture, fake treatment proves as useful as real treatment, suggesting the entire theory and protocols of convention acupuncture are simply an elaborate placebo. This was the conclusion of Moffet (2009), following a systematic review of these studies. If this is the case, we have no reason to anticipate that acupuncture for persons with ASD will produce more any benefits than placebo treatments, since such negative results have been found in the treatment of many other conditions.

Hyperbaric Oxygen Therapy: The rationale for hyperbaric oxygen therapy (HBOT) as a treatment for ASD is the theory that the brains of persons with ASD suffer from reduced blood flow and/or reduced levels of oxygen in the blood going to the brain. HBOT involves placing the patient with ASD, usually with a trusted caregiver, in a hyperbaric chamber, and exposing both to increased air pressure and an oxygen-enriched atmosphere, usually for several hours at a time, for treatments being daily or less frequently for long periods of time, perhaps weeks. Clinic-based treatments can be done in hard chambers, but portable chambers can be acquired for home use. HBOT is very expensive. One review published by Dunleavy and Thyer (2014) concluded:

"Studies reviewed did not offer credible evidence to suggest that HBOT is an effective treatment for autism. Conclusion: It is premature to call HBOT an effective treatment for Autism and ASD. Individuals clinically treated with HBOT outside the context of a RCT should have the effects of the therapy evaluated using rigorous single-subject designs" (p. 1). A more comprehensive systematic review sponsored by the Cochrane Collaboration in 2016 found that: "To date, there is no evidence that hyperbaric oxygen therapy improves core symptoms and associated symptoms of ASD. It is important to note that adverse effects (minor-grade ear barotrauma events) can occur. Given the absence of evidence of effectiveness and the limited biological plausibility and possible adverse effects, the need for future RCTs of hyperbaric oxygen therapy must be carefully considered" (cited from https://www.cochranelibrary.com/cdsr/doi/10.1002/14651858.CD010922.pub2/full#CD010922-abs-0007). Of note here is that the authors not only concluded that the treatment has little evidence to support its clinical use but also that the underlying biological theory is so implausible as to suggest this therapy should not even be studied any more.

Bicom Therapy: The Bicom apparatus (a picture of it can be seen here, (https://lyme-knowledge.wordpress.com/2016/06/30/rife-bio-resonance/) or simply by Googling Bicom is an impressive-looking supposedly medical device which is said to detect pathogens and stressors afflicting someone and causing ill health. It is said to do this by detecting some type of unique "vibration" of the harmful agent residing in the patient's body. Then, once detected, the machine is said to be able to feedback a counter or corrective vibration into the patient's body to eliminate the problem. This is called bioresonance therapy. The Bicom diagnoses not through any legitimate test but through the putative assessment of bodily energies unknown to science, and the therapy too involves vibrations or energies which science cannot otherwise detect. Bicom therapy is being advertised to treat persons with ASD, and caregivers are told:

"Bioresonance therapy has shown promising results when used amongst patients diagnosed with autism – both young and old. The technique utilizes the body's own electromagnetic waves in order to promote improved healing capabilities, better metabolism and improved detoxification. Even though bioresonance therapy is not able to correct the damage that autism has dealt to the patient's brain, this therapy has been shown to enhance the function of the patient's brain; thus allowing them to experience an improvement in their symptoms…."(cited from: https://bioresonance.com/bioresonance-therapy-and-autism/)

Here is another explanation of how the Bicom device works:"Bioresonance therapy is a therapy with patient's own electromagnetic frequency patterns. The patient's own electromagnetic oscillations of his body are received by electrodes working as an antenna and fed into the device. The BICOM device changes the body's own information with the help of special electronic systems into therapy signals, which are returned to the patient by the output cable. Due to this method the electromagnetic pathologic information in the body is eliminated i.e. reduced. The patient and the therapy device enter a feedback cycle." (cited from https://www.quackwatch.org/01QuackeryRelatedTopics/Cancer/bioresonance.html)

One can read online testimonials about the powerful effects of bioresonance therapy delivered via the Bicom (http://www.bioresonance-therapy.com.sg/testimonials-adhdorautism). The treatment is not approved by the Federal Food and Drug Administration for use with humans (only animals), but unscrupulous clinicians use this bogus device and a convincing line of jargon and neologisms to persuade parents and caregivers to bring their child in for diagnostic and therapy services. Some parents purchase the machine themselves and use it at home with their child with ASD. Treatments are very expensive, and another red flag is the vast number of diseases and conditions the Bicom is said to be able to cure. Bicom therapy could be held up as a perfect poster child for pseudoscientific treatments for

ASD. The mechanism of action is theoretically implausible. The level of evidence remains at the testimonial stage, despite more than two decades of use. It is expensive. It uses scientific-sounding jargon to impress. Tellingly, what does the company's own website say, in tiny print at the bottom of the page? "In conventional medicine, however, Bicom bioresonance has not been subject to scientific research and is not yet recognized" (cited from https://www.regumed.com/bioresonance-for-doctors-naturopathic-practitioners/method/costs.html).

This latter confession brings us to the ethics of offering pseudoscientific treatments.

The Ethics of Pseudoscientific Therapies for ASD

Supporters of pseudoscientific therapy often counterarguments such as those presented in this chapter with the view that there is no harm in providing treatments which provide hope and even that the delivery of placebo therapies is legitimate. In juxtaposition to this, the World Medical Association has published its *Declaration of Helsinki—Ethical Principles for Medical Research Involving Human Subjects* (https://www.wma.net/policies-post/wma-declaration-of-helsinki-ethical-principles-for-medical-research-involving-human-subjects/), saying:

"Use of Placebo

> 33. The benefits, risks, burdens and effectiveness of a new intervention must be tested against those of the best proven intervention(s), except in the following circumstances:
> Where no proven intervention exists, the use of placebo, or no intervention, is acceptable; or where for compelling and scientifically sound methodological reasons the use of any intervention less effective than the best proven one, the use of placebo, or no intervention is necessary to determine the efficacy or safety of an intervention and the patients who receive any intervention less effective than the best proven one, placebo, or no intervention will not be subject to additional risks of serious or irreversible harm as a result of not receiving the best proven intervention. Extreme care must be taken to avoid abuse of this option.

Unproven Interventions in Clinical Practice

> 37. In the treatment of an individual patient, where proven interventions do not exist or other known interventions have been ineffective, the physician, after seeking expert advice, with informed consent from the patient or a legally authorized representative, may use an unproven intervention if in the physician's judgement it offers hope of saving life, re-establishing health or alleviating suffering. This intervention should subsequently be made the object of research, designed to evaluate its safety and efficacy. In all cases, new information must be recorded and, where appropriate, made publicly available."

In general the advocates of pseudoscientific ASD therapies have no motivation to conduct properly controlled experimental evaluations of their treatments. They have nothing to gain except unwelcome news; thus few accumulate any substantial body of research evidence. The clinical use of unproved interventions is very strictly limited; thus practitioners who knowingly provide treatments that are essentially placebo-type of therapies are practicing unethically. This appears to be the case for a number of the pseudoscientific interventions described in this chapter. And interventions which simply lack sufficient evidence to draw a conclusion should be tested against best available treatments.

The American Medical Association also weighs in on this topic.

"Code of Medical Ethics Opinion 2.1.4

> A placebo is a substance provided to a patient that the physician believes has no specific pharmacological effect on the condition being treated. The use of placebo, when consistent with good medical care, is distinct from interventions that lack scientific foundation…In the clinical setting, the use of a placebo without the patient's knowledge may undermine trust, compromise the patient-physician relationship, and result in medical harm to the patient.
> Physicians may use placebos for diagnosis or treatment only if they:

(a) Enlist the patient's cooperation. The physician should explain that it can be possible to achieve a better understanding of the medical condition by evaluating

> the effects of different medications, including the placebo.
>
> (b) Obtain the patient's general consent to administer a placebo. The physician does not need to identify precisely when the placebo will be administered. In this way, the physician respects the patient autonomy and fosters a trusting relationship, while the patient may still benefit from the placebo effect.
>
> (c) Avoid giving a placebo merely to mollify a difficult patient. Giving a placebo for such reasons places the convenience of the physician above the welfare of the patient. Physicians can produce a placebo-like effect through the skillful use of reassurance and encouragement, thereby building respect and trust, promoting the patient-physician relationship, and improving health outcomes." (cited from: https://www.ama-assn.org/delivering-care/use-placebo-clinical-practice)."

> The Code of Ethics for Behavior Analysts is particularly strong in advocating for the provision of research-based therapies for clients with ASD. For example:
> "2.09 Treatment/Intervention Efficacy. (a) Clients have a right to effective treatment (i.e., based on the research literature and adapted to the individual client). Behavior analysts always have the obligation to advocate for and educate the client about scientifically supported, most-effective treatment procedures. Effective treatment procedures have been validated as having both long-term and short-term benefits to clients and society" (cited from https://www.bacb.com/wp-content/uploads/170706r_compliance_code_english.pdf).

No other profession serving persons with ASD includes such a strong ethical requirement regarding the practitioner's obligation to provide scientifically supported treatments.

There are good reasons for avoiding placebo-equivalent treatments. A thorough review of clinical trials comparing real therapies to placebo treatments found that the latter had very little meaningful impact on clinical outcomes in medicine. The authors concluded "we found little evidence that placebos in general have powerful clinical effects…The use of placebo outside the aegis of properly designed clinical trials cannot be recommended" (Hrobjartsson & Gotzsche, 2001, p. 1599). Another reason is that a profession which relies primarily on placebo influences to obtain patient-reported improvements (e.g., chiropractic, naturopathy, acupuncture) is held in low self-esteem by health-care professionals operating from more of an evidence-based practice orientation. And certainly one does not need years of graduate and post-graduate training to be a placebo therapist. The hallmark of a legitimate health-care professional is one's capacity to deliver treatments that are *more* beneficial than credible placebos.

Apart from placebo influences, there are a large number of reasons why genuinely ineffective treatment may appear to work. Lilienfeld, Ritschel, Lynn, Cautin, and Latzman (2014) provide a comprehensive discussion of 25 additional reasons. These were grouped into three categories: (1) erroneous perceptions of client change, in its absence (15 possible reasons), (2) misinterpretations of actual client change stemming from extra-therapeutic factors (eight factors), and (3) misinterpretations of actual client change stemming from nonspecific treatment factors (three factors, include placebo). Anyone evaluating treatment outcome studies for ASD should keep these alternative explanations in mind when attempting to account for apparent client improvements. They conclude their compelling article with the following observation: "Science, which is a systematic approach to reducing uncertainty in our inferences… is ultimately our best prescription against being deceived by inadequate evidence" (Lilienfeld et al., 2014, p. 378). Not all purveyors of pseudoscientific treatments for ASD are charlatans, but it matters little to the recipient of such services whether the therapist is honestly misguided or a knowing purveyor of autistic snake oil. The end result is the same. No improvements beyond placebo factors. Loss of money. False hope. Wasted time. Disillusionment. Possible injury.

A chapter-length treatment of topic cannot do justice to the field as only a few examples of pseudoscientific treatments can be briefly described. The reader is referred to several of the book-length analyses devoted to the topic, e.g., Jacobson, Foxx, and Mulick (2005), Foxx and Mulick (2015), and Offit (2010) as well as to some excellent chapters (e.g., Tizikow & Holburn, 2011). This chapter's focus on pseudoscientific therapies for ASD may leave the reader a bit frustrated in that little mention was made of well-established research-supported treatments.

Nor was there discussion of the possible role of prescription psychotropic medications. This latter omission can be partially justified on the basis that very little is known. In the United States, the Federal Food and Drug Administration has only approved two medications for use in treating ASD, risperidone and aripiprazole, and these are intended to reduce irritability, an important but very limited goal (LeClerc & Easley, 2015), although the off-label use of everything under the sun is common among prescribers—tranquilizers to subdue patients, anti-psychotic agents, antidepressants, hormones like oxytocin, stimulant medications, and so forth.

The American Psychiatric Association provides the following guidance touching on this issue:

“Clinical decision-making without established research evidence to guide practice requires informed clinical judgments drawing on the best available research, adherence to the ethical principles of beneficence and non-maleficence, and sound theoretical reasoning. When usual treatments have failed, psychiatrists may offer non-standard or novel interventions using a shared decision-making approach grounded in the patient’s informed consent and a thorough discussion of risks, benefits, and alternatives to the innovative treatment. Since innovative practice sometimes leads to important scientific advances, it should not be categorically discouraged; however, because it may prove ineffective or even harmful, psychiatrists should proceed with caution in their use of clinical innovation. When considering use of clinical innovation, psychiatrists should consider first consulting colleagues and exploring other resources to ensure that careful thought has been given to possible alternatives as well as to the safest and most effective use of innovative interventions.” (cited from file:///C:/Users/Bthyer/Downloads/APA-Commentary-on-Ethics-in-Practice.pdf)

Clinically innovative practices is a nice way of saying off-label prescribing, using drugs for purposes for which they not been approved. More honest way is to describe it as clinical experimentation on vulnerable human beings. Note the caveat that this is justifiable only when usual treatments have failed. This should be thoroughly documented, with empirical data, before any such off-label prescribing is undertaken. I suggest some more prescriptive guidelines are in order, such as:

- Medication should be tried only after less intrusive methods of treatment have been given a legitimate trial. Examples could include behavior analysis, environmental modification, parent training, etc.
- Whenever medication is intended to produce changes in behavior, affect, or cognition, a credible baseline series of measures of these variables must be taken, prior to introducing the medication. These measures should have acceptable levels of reliability and validity. When the health and safety of the patient with ASD is at risk, this requirement for a baseline may be omitted.
- When medication is provided, ongoing valid assessments of the outcome measures (behavior, affect, cognition) must be obtained on a regular basis, and any changes in the medication regimen should be based on these data. The data must be properly recorded and available in the client’s records.

These recommendations are completely consistent with the fifth step of evidence-based practice “evaluating our effectiveness and efficiency” (Straus, Glasziou, Richardson, & Haynes, 2011, p. 3) as well as in partial compliance with relevant behavior analyst ethical standards:

“(a) Behavior analysts conduct current assessments prior to making recommendations or developing behavior-change programs. The type of assessment used is determined by client’s needs and consent, environmental parameters, and other contextual variables. When behavior analysts are developing a behavior-reduction program, they must first conduct a functional assessment. (b) Behavior analysts have an obligation to collect and graphically display data, using behavior-analytic conventions, in a manner that allows for decisions and recommendations for behavior-change program development” (cited from

Section 3.01 of https://www.bacb.com/wp-content/uploads/170706r_compliance_code_english.pdf)
Other stipulations could readily be made concerning the monitoring of potential side effects, the value of periodic drug holidays, the need for appropriate medication tapers rather than abrupt discontinuance, and the required use of written informed consent from the caregivers for such treatments. See Cohen and Jacobs (1998) for one example of such a form.

Summary

Pseudoscientific therapies for persons with ASD are widely available. They are avidly sought after by despairing parents and caregivers and equally avidly promoted by their advocates. Some advocates are honestly mislead, and others are more akin to therapeutic hucksters and quacks, eager to dishonestly earn money or to acquire a reputation as a marvelous healer. There are many reasons why ineffective therapies can appear to "work," and it requires intense effort over many years to develop a sufficiently strong research base so as to be able to ascertain if a treatment is effective or not. Most interventions tested are not effective, which tends to discourage such long-term research programs. This chapter has provided some guidelines to help recognize a potentially pseudoscientific therapy and described a selected number of these. Far too many remain unrecognized as a bogus treatment. The marketing and sales of pseudoscientific treatments can be a big business, yielding sizeable returns for those who promote them.

There is a growing literature on identifying pseudoscientific treatments, and increasingly practitioners who take a more evidence-based practice approach to care are willing to confront and disclose professionals who dishonestly promote bogus ASD and other therapies under the umbrella of their credentials as a medical doctor, psychologist, social worker, or other types of practitioner. Such efforts should be encouraged, although at times it seems akin to playing "whack-a-mole," in that while temporarily suppressed (e.g., facilitated communication) the practice springs forth anew, perhaps under a different name (e.g., rapid prompting method). The president of the American Speech-Language-Hearing Association deserves credit, for example, for taking on a testimonial editorial that appeared in the *Wall Street Journal* which advocated for a pseudoscientific treatment called the rapid prompting method, which is warmed-over facilitated communication, a long discredited therapy said (erroneously) to permit inarticulate persons with ASD to type fluently (Davis-McFarland, 2018).

Psychologist Jean Mercer has established a Facebook page called *Psychology CE Watch—When Approved Courses Don't Meet Stated Standards* (see https://www.facebook.com/groups/161745967794736/), devoted to disclosing professional continuing education programs which contain pseudoscientific content and coordinating complaints to the American Psychological Association when such CE content does not adhere to the APA's (minimal) required standards of research support. All professionals are welcome to join this page and help identify such programs and to submit complaints through the proper channels. Several previously approved CE programs had had their APA approval removed through this initiative. A couple of divisions of the APA have undertaken a similar initiative, to identify and coordinate complaints about bogus CE courses. This is described on the above website.

Eventually it is hoped, more caregivers of person with ASD who receive pseudoscientific treatments will not just complain but will file lawsuits alleging malpractice at the hands of the professionals delivering the therapy. Winning such lawsuits will be an effective way to discourage providing illegitimate treatments, as will major professional association issuing more position statements condemning the practice of selected pseudoscientific treatments. Science-based treatments are slowly growing in number but are being outpaced by the proliferation of bogus therapies. Professionals in the field of developmental disabilities need to become less tolerant of these cuckoos in our nest, and push them out, before these fledglings come to dominate the marketplace, to the disadvantage of persons with ASD and their families.

References

Al-Ayadhi, L. Y., & Elamin, N. E. (2013). Camel milk as a potential therapy as an antioxidant in autism spectrum disorder. *Evidence-based Complementary and Alternative Medicine, 2013*, 602834.

American Psychiatric Association (1968). Diagnostic and statistical manual of mental disorders (2nd edition). Washington, DC: Author.

American Psychiatric Association. (2013). Diagnostic and statistical manual of mental disorders (5th edition). Washington, DC: Author.

Bellini, S., & Akullian, J. (2007). A meta-analysis of video modeling and video self-modeling interventions for children and adolescents with autism spectrum disorders. *Exceptional Children, 73*, 264–287.

Bernard, C. (1865/1927). *An introduction to the study of experimental medicine*. New York: Macmillan.

Borchardt, D. (10 June, 2015). Desperate parents of autistic children trying cannabis despite lack of studies. *Forbes*. Retrieved from https://www.forbes.com/sites/debraborchardt/2015/06/10/desperate-parents-of-autistic-children-trying-cannabis-despite-lack-of-studies/#1f29aa266616

Bunge, M. (1998). *Philosophy of science: From problem to theory, volume one (revised edition)*. New Brunswick, NJ: Transaction.

Cohen, D., & Jacobs, D. (1998). A model consent form for psychiatric drug treatment. *International Journal of Risk and Safety in Medicine, 11*, 161–164.

Davis-McFarland, E. (28 September, 2018). RPM for autism not supported by science (letter). *Wall Street Journal*. A14.

Dunleavy, D. J., & Thyer, B. A. (2014). Is hyperbaric oxygen therapy an effective treatment for autism: A review. *Journal of Adolescent and Family Health, 6*(1), Article 5. Retrieved from http://scholar.utc.edu/jafh/

Foxx, R. M., & Mulick, J. A. (Eds.). (2015). *Controversial therapies for autism and intellectual disabilities* (2nd ed.). New York: Routledge.

Guyatt, G., Rennie, D., Meade, M. O., & Cook, D. J. (2002). Introduction: The philosophy of evidence-based medicine. In G. Guyatt & D. Rennie (Eds.), *Users guide to the medical literature* (pp. 5–71). Chicago: AMA Press.

Herbert, J. D., Sharp, I. R., & Gaudiano, B. A. (2002). Separating fact from fiction in the etiology and treatment of autism. *The Scientific Review of Mental Health Practice, 1*, 23–43.

Horvath, K., Stefanos, G., Sokolski, K. N., Nabors, L., & Tildon, J. T. (1998). Improved social and language skills after secretin administration in patients with autistic spectrum disorders. *Journal of the Association for Academic Minority Physicians, 9*, 9–15.

Hrobjartsson, A., & Gotzsche, P. C. (2001). Is the placebo powerless? An analysis of clinical trials comparing placebo with no treatment. *New England Journal of Medicine, 344*, 1594–1602.

Hupp, S., Mercer, J., Thyer, B. A., & Pignotti, M. (in press). Pseudoscience in child and adolescent psychotherapy: Skeptical guide for therapists and parents. In S. Hupp (Ed.), *Pseudoscience in child and adolescent psychotherapies: Ineffective, implausible, and potentially harmful treatments*. New York: Springer.

Jacobson, J. W., Foxx, R. M., & Mulick, J. A. (Eds.). (2005). *Controversial therapies for developmental disabilities: Fad, fashion, and science in professional practice*. Mahwah, NJ: Lawrence Erlbaum.

Johnson, S. M. & Hollander, E. (2003). Evidence that eicosapentaenoic acid is effective in treating autism. The Journal of Clinical Psychiatry, 64, 848–849.

LeClerc, S., & Easley, D. (2015). Pharmacological therapies for autism spectrum disorder: A review. *Pharmacy and Therapeutics, 40*, 389–397.

Lilienfeld, S. O., Ritschel, L. A., Lynn, S. J., Cautin, R. L., & Latzman, R. D. (2014). Why ineffective psychotherapies appear to work: A taxonomy of causes of spurious therapeutic effectiveness. *Perspectives on Psychological Science, 9*, 355–387.

Lovass, I. (1987). Behavioral treatment and normal educational and intellectual functiong in young autisic children. Journal of Abnormal Child Psychology, 20, 555–566.

Mihic, T., Rainkie, D., Wilby, K. J., & Pawluk, S. A. (2016). The therapeutic effects of camel milk: A systematic review of animal and human trials. *Journal of Evidence-based Complementary and Alternative Medicine, 21*, NP110–NP126.

Moffet, H. H. (2009). Sham acupuncture may be as efficacious as true acupuncture: A systematic review of clinical trials. *Journal of Alternative and Complementary Medicine, 14*, 213–216.

Nikles, J., & Springer, G. (Eds.). (2015). *The essential guide to n – of – 1 trials in health*. New York: Springer.

Offit, P. A. (2010). *Autism's false prophets: Bad science, risky medicine, and the search for a cure*. New York: Columbia University Press.

Schork, N. J. (2015). Time for one-person trials. *Nature, 520*, 609–611.

Straus, S. E., Glasziou, P., Richardson, W. S., & Haynes, R. B. (2011). *Evidence-based medicine: How to practice and teach it* (4th ed.). New York: Churchill Livingstone.

Thyer, B. A. (2019). Predatory doctoral programs: Warnings for social workers. *Journal of Social Work Education., 55*(1), 5–10.

Tizikow, J. E., & Holburn, S. (2011). Identifying fad therapies for autism spectrum disorders and promoting effective treatment. In J. Matson & S. Peter (Eds.), *International handbook of autism and pervasive developmental disorders* (pp. 307–319). New York: Springer.

29

Caregiver Treatment Choices

Sarah Carlon, Jennifer Stephenson,
and Mark Carter

Critical Nature of Treatment and Intervention Selection

There are a number of treatment and intervention options available for people with intellectual disabilities. These range from evidence-based treatments (as described in Chap. 28), such as those derived from the principles of applied behavior analysis (ABA), to unsupported treatments (as described in this chapter), such as complementary and alternative medicine (CAM) treatments. On balance, it can be argued that interventions with empirical support represent better options for the majority of individuals with intellectual disabilities compared to interventions without such support.

Families, schools, and governments have finite financial resources, and the economic burden of intellectual disabilities can be significant. For example, the cost of supporting one individual with comorbid intellectual disability and autism spectrum disorder (ASD) over the life span has been estimated to cost US$2.4 million in the United States (US) and US$2.2 million in the United Kingdom (UK) (Buescher, Cidav, Knapp, & Mandell, 2014). In comparison, in both the US and the UK, the estimated cost of supporting an individual with ASD but without intellectual disability over the life span was US$1.4 million (Buescher et al., 2014). In Australia, the total government expenditure on services for people with intellectual disabilities has been estimated at A$3.361 billion each year (Doran et al., 2012).

In addition to financial costs, individuals with intellectual disabilities, family members of these individuals, and professionals who work with them have limited time, resources, and energy to devote to implementing treatments and interventions. Given the finite nature of these resources, it is critical that money, time, and energy are used effectively. While there is no one treatment or intervention that is guaranteed to be effective for all individuals with intellectual disabilities, there are clearly treatments and interventions that are evidence-based and others that are not supported by empirical evidence (Bowen & Snow, 2017; Courtade, Test, & Cook, 2015; West, McCollow, Kidwell, Umbarger, & Cote, 2013). An investment in the use of an evidence-based treatment or intervention is an investment in a treatment or intervention that is more likely to be effective. In contrast, if parents or professionals choose to invest in treatments or interventions that lack empirical support, not only are these treatments/interventions less likely to work, but there is also an opportunity cost. Time, energy, and resources will be devoted to the treatment or intervention that is less likely to be effective, and as a result

S. Carlon (✉) · J. Stephenson · M. Carter
Macquarie University, Sydney, NSW, Australia
e-mail: sarah.carlon@mq.edu.au; jennifer.stephenson@mq.edu.au; mark.carter@mq.edu.au

J. L. Matson (ed.), *Handbook of Intellectual Disabilities*, Autism and Child Psychopathology Series, https://doi.org/10.1007/978-3-030-20843-1_29

this may lead to the parent or professional forgoing the use of evidence-based treatments and interventions.

Parents of individuals with intellectual disabilities and/or ASD and professionals working with individuals with these diagnoses are typically using a range of treatments and interventions including those that are unsupported by research evidence and/or those that are controversial (Burns & Ysseldyke, 2009; Carter, Stephenson, & Strnadová, 2011; Goin-Kochel, Mackintosh, & Myers, 2009; Haessler et al., 2016; Matson & Neal, 2009; Matson & Williams, 2015). It is therefore important to consider the range of factors related to treatment and intervention decisions made by both parents and professionals in order to understand how these decisions are made and how better decision-making may be supported in the future.

Factors Related to Treatment and Intervention Decisions

In recent years the decision-making, particularly of parents, regarding treatment and intervention use has become a research area of interest. As a result, an increasing number of research studies have been published in this area. It is important to note that most of the published research in this area is related to decision-making regarding treatments and interventions for individuals with ASD, but comorbidity of intellectual disabilities and ASD is common (as addressed in Chap. 49). This research will be presented along with research related to decision-making for treatments and interventions for intellectual disabilities and other specific diagnoses that are characterized by intellectual disabilities, such as Fragile X and Down syndrome.

Decision-Making of Parents

Introduction

Factors that are related to the decisions of parents regarding intervention use have been explored in the research literature. These include both factors that parents have explicitly identified as influencing their decision-making, known as *declared decision-making factors*, and other underlying factors that may influence parent decision-making but that parents may not necessarily be aware of, known as *implicit decision-making factors* (Carlon, Carter, & Stephenson, 2013; Wilson, Hamilton, Whelan, & Pilkington, 2018). *Declared decision-making factors* include factors such as recommendations from others, research evidence, and pragmatic considerations, while *implicit decision-making factors* include parent and child factors such as age, gender, and time since diagnosis (Carlon et al., 2013; Wilson et al., 2018).

Declared Decision-Making Factors

In the research literature, a range of factors have been explicitly identified by parents of individuals with intellectual disabilities, ASD, or both, as influencing their treatment and intervention decision-making. It should be acknowledged that most of this literature relates to the decision-making of parents of children with ASD, which is suggestive for parents of children with intellectual disabilities (e.g., Bowker, D'Angelo, Hicks, & Wells, 2011; Dinora & Bogenschutz, 2018; Tzanakaki et al., 2012). However, where studies have addressed the decision-making of parents of children with intellectual disabilities, these have been included (e.g., Foran & Sweeney, 2010; Prussing, Sobo, Walker, & Kurtin, 2005). In addition, some studies addressed both the decision-making of parents of children with intellectual disabilities and of parents of children with ASD (e.g., Robert, Leblanc, & Boyer, 2015; Wodehouse & McGill, 2009); and some addressed the decision-making of parents of children with ASD including some children with comorbid intellectual disability diagnoses (e.g., Hanson et al., 2007; Shyu, Tsai, & Tsai, 2010). Declared decision-making factors that have been frequently examined in the literature are presented below.

Advice and Recommendations

Parents of children with intellectual disabilities and of children with ASD receive information and advice about treatments and interventions available from many sources. These may include other parents, medical doctors, the Internet, teachers or educators, friends/relatives, autism organizations/associations, popular media, books/authors on autism, therapists (including speech therapists, occupational therapists, behavior analysts, audiologists, and physical therapists), psychologists, disability support groups and newsletters, research literature, childcare providers, and diagnostic services (Carlon et al., 2013; Carlon, Stephenson, & Carter, 2015; Dinora, Bogenschutz, & Lynch, 2017; Prussing et al., 2005).

A common theme in the literature related to the type of information offered to parents by professionals, particularly around the time of their child's diagnosis, is that, while some professionals provide useful advice and guidance, this is not always the case. The information provided can be limited and professionals may not always provide advice that parents consider to be useful in their decision-making (see Carlon, Stephenson, & Carter, 2015; Dinora & Bogenschutz, 2018; Sansosti, Lavik, & Sansosti, 2012; Tzanakaki et al., 2012; Wodehouse & McGill, 2009). Parents can be expected to make complex decisions about treatment and intervention use that can leave them feeling unsupported:

> "… several families indicated that they 'were getting the runaround from professionals and schools… no recommendations… no road map.'" (Sansosti et al., 2012, p. 88)

> "i am using no treatments now, since diagnosis ive been left alone, its like they say shes this, now get on with it." (Mackintosh, Goin-Kochel, & Myers, 2012, p. 57)

> "When a mother of a 4-year-old was asked what advice she would give a pediatrician about how to work with families of children with autism, she said, "Listen. I want them to listen to me. Don't be afraid to offer advice. It is okay. I need it. Don't worry about offending me. Honestly. Be honest with me. Don't give me some run-around because I will know if you are just telling me what I want to hear. Be real."" (Levy et al., 2016, p. 575)

> "My biggest gripe about when you get a diagnosis is that you're a parent — you're not a doctor, you're not a speech therapist, you're not an occupational therapist, but you're given a pack and given the FaHCSIA money[1] and told to make the decision as to how you are going to treat your own child. It all depends on, possibly it all depends on, the first person you see and their opinion that they give you, or you just educate yourself and then try to figure out what your child needs — but it's a bit like a doctor saying, 'You've got cancer. How would you like to treat it?'" (Carlon, Stephenson, & Carter, 2015, p. 122)

Most of the research related to advice and recommendations is limited to the examination of the sources of advice reported to be used by parents, and some qualitative examination of the parent perceived usefulness of such advice. Some researchers, however, have also examined the weight of importance that parents place on recommendations from different sources when making treatment and intervention decisions. Two recent survey studies conducted in the US have provided some insight into the relative level of influence that different sources of information had on parent decision-making regarding ASD interventions and treatments. Dinora et al. (2017) reported that the parents of children with ASD who participated in their study rated developmental pediatricians, their spouse, school staff, speech language pathologists, and primary care physicians as having the most influence (moderate to high influence) on their intervention decisions. Television shows, magazines, social media, and other people were rated as having the least influence (Dinora et al., 2017). Participants in Deyro, Simon, and Guay (2016) were asked to rank six information sources (professional referral, general media, other parents with a child or children with ASD, autism organizations, books, and scientific journals) based on the level of influence on treatment and intervention deci-

---

[1] In Australia, the Federal Government Department of Families, Housing, Community Services and Indigenous Affairs (FaHCSIA) provided eligible families of children under 7 years of age with a confirmed ASD diagnosis up to A$12,000 over 2 years to fund early intervention services provided by a range of approved providers. Parents were provided with information (but not advice) about these services from *Autism Advisors*. The parents were required to choose how to spend these funds.

sions. The source ranked as most influential by the greatest number of participants was professional referrals (48.5%), followed by other parents (20%) and scientific journals (10%). A total of 34.3% of participants indicated that the general media was the least influential source, followed by scientific journals (24.4%), then books, and other parents (5% each).

In Australia, Carlon, Carter, and Stephenson (2015) asked parents of preschoolers with ASD to rate the level of importance they placed on a range of factors (including advice from different sources) when making the decision to use the intervention they nominated as the most important intervention currently being used with their child. On a scale of 1–5 (1 = very unimportant, 5 = very important), advice from therapists received a mean rating of 4.5, advice from teachers or educators 4.1, advice from medical doctors 4.0, advice from other parents 3.9, and advice from friends or relatives 3.2. Consistent with the findings of Deyro et al. (2016), this indicates that parents placed a greater weight on recommendations received from professionals than those received from other sources (Carlon, Carter, & Stephenson, 2015).

Nevertheless, research related to the weight placed by parents on different sources of advice is limited. The findings of Carlon, Carter, and Stephenson (2015), Deyro et al. (2016), and Dinora et al. (2017) indicate that the sources of information that are most frequently addressed in the research literature may not necessarily be the most influential on parent decision-making. For example, other parents are most frequently identified as sources of advice in the literature related to parent decision-making regarding treatments and interventions for ASD (Carlon et al., 2013), yet in Carlon, Carter, and Stephenson (2015), Deyro et al. (2016), and Dinora et al. (2017), parents indicated that other parents had less influence on their decision-making than other sources of advice (such as professionals).

Child Factors

Individual needs. Unsurprisingly, the individual needs or characteristics of the child are frequently reported in the literature as factors considered in decision-making for parents of children with intellectual disabilities and of children with ASD (Carlon et al., 2013; Edwards, Brebner, McCormack, & MacDougall, 2018; Finke, Drager, & Serpentine, 2015; Prussing et al., 2005; Robert et al., 2015). There is also some evidence that parents may place considerable weight on their child's individual needs in their decision-making. In the Carlon, Carter, and Stephenson (2015) survey of Australian parents of preschoolers with ASD, the participants were asked to indicate the level of importance that a range of factors had on both their decisions to use a nominated intervention and to reject a nominated intervention. All participants indicated that the child's individual needs were important in the decision to use the intervention nominated as the most important intervention currently used with their child. It was also the factor with the highest mean importance ratings in both the decisions to use and to reject the nominated interventions.

Age. The age of the child appears to be a factor that is considered by parents in some treatment and intervention decisions, but it has not been as widely examined as an explicitly stated factor in decision-making as other child factors (Carlon et al., 2013; Wilson et al., 2018). However, the implicit relationship between child age and treatment/intervention decisions has been extensively examined and is discussed in section "Declared Decision-Making Factors".

Side effects or adverse effects. Parents of children with intellectual disabilities and of children with ASD have reported side effects or adverse effects of treatments as reasons for discontinuing treatments (Bowker et al., 2011; Prussing et al., 2005). In addition, concerns about the safety and possible side effects of medications have also led to parents rejecting the use of conventional medications and choosing to use CAM treatments as alternatives (Bilgic et al., 2013; Hanson et al., 2007; Wong, 2009). However, it appears that not all parents view CAM treatments as a safe alternative. For example, Prussing et al. (2005) reported that some parents decided not to try CAM treatments with their children with Down syndrome due to concerns regarding efficacy and safety. Nonetheless, although the specific treatments considered by parents as safer or riskier options may vary from parent to parent, the evidence suggests that parents may consider the safety of treatments in terms of actual or possible side effects or adverse effects when deciding whether to commence and/or discontinue the use of these treatments.

Perceived enjoyment. Whether or not the parent believed the child would enjoy the intervention or treatment, including consideration of possible resistance to the intervention or treatment, has been reported to be considered by parents across several studies (e.g., Carlon, Carter, & Stephenson, 2015; Christon, Mackintosh, & Myers, 2010; Shyu et al., 2010). These have been limited to the ASD population, including one study with some participants with comorbid intellectual disability diagnoses.

Perceived child progress. Frequently in the literature addressing decision-making for ASD treatments and interventions, parents have indicated that their perception of their child's progress has influenced their decision to either continue using (Edwards et al., 2018) or to discontinue the use of a treatment/intervention (Bowker et al., 2011; Christon et al., 2010; Edwards et al., 2018). While some parents discontinued treatments and interventions due to a perceived lack of progress, others did so because they believed that the treatment or intervention was no longer required, either because the child's skills had progressed to a level where the treatment/intervention was no longer necessary and/or because the child had completed the treatment/intervention (Bowker et al., 2011; Christon et al., 2010; Finke et al., 2015).

Emotion-Based Factors

The influence of emotion-based factors has mostly been examined in qualitative research regarding parent decision-making for both interventions for ASD and for intellectual disabilities. A sense of hope, both for improvement (Carlon, Carter, & Stephenson, 2015; Finke et al., 2015; Tzanakaki et al., 2012) and for a cure (Carlon, Carter, & Stephenson, 2015; Finke et al., 2015), has been identified by parents as a consideration in treatment and intervention decision-making. Parents have also reported following their own intuition or "gut feelings" when selecting interventions (Carlon, Carter, & Stephenson, 2015; Prussing et al., 2005). Participants in Carlon, Carter, and Stephenson (2015) rated the importance of factors in their decision to use a nominated intervention. They ranked emotion-based factors significantly higher in importance than advice/recommendations. However, these parents placed significantly more weight on service characteristics, research evidence, and child factors than on emotion-based factors. In contrast, when asked about factors influencing decisions to reject a nominated intervention, emotion-based factors were ranked significantly higher than both pragmatic factors, research evidence, and advice/recommendations. For the decision to reject the nominated intervention, emotion-based factors received the highest mean importance ranking of all of the examined categories of factors.

Pragmatic Factors

Unsurprisingly, there are a range of pragmatic considerations that parents of children with intellectual disabilities and of children with ASD take into account when making decisions about treatment and intervention use. One of the first considerations of parents may be whether a specific treatment or intervention is available and accessible to the family. The availability and accessibility of the treatment/intervention have been reported frequently as decision-making factors in the literature (see Carlon et al., 2013; Dinora et al., 2017; Edwards et al., 2018). Some interventions can be restricted to specific diagnostic criteria and/or only offered to individuals with a specific level of functioning, which in turn can limit intervention options (Foran & Sweeney, 2010; Robert et al., 2015).

Related to availability, parents commonly report that there are waiting lists for services, particularly those that are publicly funded (Dinora et al., 2017; Foran & Sweeney, 2010; Robert et al., 2015; Sansosti et al., 2012; Valentine, 2010). In addition to access to intervention services provided as part of public health or education systems, access (or lack thereof) to government funding support and/or to insurance cover has been reported to influence parent treatment and intervention decision-making in several studies (Dinora et al., 2017; Edwards et al., 2018; Grant, Rodger, & Hoffmann, 2016; Mackintosh et al., 2012; Shepherd, Csako, Landon, Goedeke, & Ty, 2018; Valentine, Rajkovic, Dinning, & Thompson, 2010). The cost of treatments and interventions, whether related to access to external funding or not, is a logical consideration for parents. As such, it has been reported across several studies (Carlon et al., 2013; Carlon, Carter, & Stephenson, 2015; Edwards et al., 2018; Robert et al., 2015; Sansosti et al., 2012; Shepherd et al., 2018).

Other pragmatic considerations include the convenience to or impact on the family (Edwards et al., 2018; Rajkovic, Thompson, & Valentine, 2010). Some parents have chosen to discontinue interventions or not implement interventions suggested by professionals because they did not suit their family (Robert et al., 2015). This can be related to a failure to *fit the schedule* of the child and family, and/or to the treatment or intervention being perceived as inconvenient by the family (Rajkovic et al., 2010; Robert et al., 2015; Yingling, Hock, Cohen, & McCaslin, 2017). Time constraints were also reported to be related to intervention decisions across several studies (Carlon et al., 2013).

Research Evidence

Mixed results have been reported regarding the impact of research evidence on parent decision-making regarding treatment and intervention use for both intellectual disabilities and ASD. Although research has been reported to be considered by parents in a number of studies (Carlon, Carter, & Stephenson, 2015, 2017; Deyro et al., 2016; Dinora et al., 2017; Hanson et al., 2007; Prussing et al., 2005; Valentine, 2010), parents continue to use interventions that are not supported by research evidence (Goin-Kochel et al., 2009; Matson & Williams, 2015). In some studies where participants were directly asked about consideration of research evidence, this term was not defined for parents (e.g., Carlon, Carter, & Stephenson, 2015), so even though the parents indicated that research was important in their decision-making, their interpretation of research evidence may not have been the same as the researchers' interpretation of this term. This is possibly because parents may interpret *research evidence* to mean their own efforts to find information about treatments and interventions or anecdotal reports of treatment/intervention effectiveness, as opposed to the findings of empirical studies.

There is limited research addressing parents' ratings of the empirical support for treatments and interventions. Deyro et al. (2016) asked parents of children with ASD in the US to rate the effectiveness of and level of research support for 26 treatments/interventions for ASD. The authors compared these parent ratings to the ratings provided by the National Autism Centre in the National Standards Report as a result of a comprehensive review of the research support for treatments and interventions for ASD (excluding medication; Deyro et al., 2016). The ratings of parents corresponded with the ratings provided by the National Autism Centre for only 9 of the 26 treatments/interventions. In addition, for most of the treatments/interventions rated, parent ratings of research evidence did not correlate with their ratings of the effectiveness of the treatments/interventions (Deyro et al., 2016). Carlon et al. (2017) collected similar data from Australian parents in a pilot study of a parent education package providing information about evidence-based practice, guidelines for selecting interventions, and guided access to two websites that provided information about the scientific efficacy of a range of interventions for ASD. They found that even after the guided access to the websites, parents' ratings of research evidence did not necessarily correspond with the ratings provided on the websites. Ratings of the research support for interventions were positively correlated with the participants' current use of and desire to use interventions, indicating that the participants tended to rate interventions that they were currently using and those that they had a strong desire to use as having strong research support, regardless of the information about research efficacy provided on the websites (Carlon et al., 2017).

Intervention/Treatment and Service Characteristics

Characteristics of the treatment/intervention along with those of service providers delivering specific interventions or treatments have also been reported to be considered by parents of children with intellectual disabilities and with ASD in their decision-making. Intervention characteristics considered have included whether or not the intervention was a diagnosis-specific intervention or more general in nature (Carlon, Carter, & Stephenson, 2015) and whether it would be compatible with other interventions/treatments already being used with the child (Rajkovic et al., 2010; Valentine et al., 2010). In addition, there is some evidence that parents may also consider whether the claims of the proponents of the intervention/treatment are feasible and/or if they fit with the parent's beliefs about the etiology of their child's condition (Bilgic et al., 2013; Prussing et al. 2005).

The characteristics of the staff, including staff experience (Carlon, Carter, & Stephenson, 2015), whether they appeared to be professional (Carlon, Carter, & Stephenson, 2015), and the parent's impressions of their intentions and whether they *fit* with the child and family (Dinora et al., 2017), are also reported to affect parental decision-making. As with the other factors discussed above, there is limited research addressing the weight of importance that parents place on these characteristics in comparison to other factors when making treatment and intervention decisions. Dinora et al. (2017) reported that the fit of the provider with the parent and child was the strongest influence on parent decision-making about ASD treatments and interventions for their US sample. In addition, high importance was placed on staff characteristics by Australian parents of preschoolers with ASD in the survey conducted by Carlon, Carter, and Stephenson (2015).

Implicit Decision-Making Factors

The factors described in the section above were those that were explicitly identified by parents as related to their treatment and intervention decision-making. There are other factors that have been examined by researchers because they have the potential to influence treatment and intervention decisions. These implicit, underlying factors are factors that parents do not explicitly declare and of which they may not be consciously aware. By their nature, implicit factors have tended to be examined in studies using correlational approaches. These data provide an indication of the relative impact of the possible implicit factors relative to others addressed in each study.

It should also be noted that researchers have used available datasets such as health insurance claim data (e.g., Chang, Lin, Tung, Chiang, & Hsu, 2014), service providers' existing records (e.g., Olsson, Elgmark Andersson, Granlund, & Huus, 2017; Paton et al., 2011), and health record databases (e.g., Osunsanmi & Turk, 2016) to examine the relationships between treatments/interventions used by individuals with intellectual disabilities and factors such as gender, age, school class placement (specialized or mainstream), comorbid diagnoses, presence of problem behavior, and living circumstances. The person(s) responsible for deciding whether to use each of the treatments and interventions in these studies is unknown. Decisions may have been made by medical staff, educators, the individual with an intellectual disability, or family members, for example. While these studies provide some insight into the range of factors that may be associated with the use of specific treatments and interventions for intellectual disabilities and/or ASD, they cannot provide unambiguous insight into the decision-making of parents or other caregivers when the decision-maker is unknown. For this reason, the research examined in this section is limited to studies in which data were collected directly from the parent(s), including secondary analysis of such data. Similarly, other research addressing the relationships between implicit factors and healthcare service use and/or expenditure on healthcare for children with intellectual disabilities (e.g., Adams et al., 2016; Lin et al., 2013; Wilkins et al., 2010) has not been considered here because it does not provide insight into the specific treatments/interventions employed by these medical and allied health service providers and related parent decision-making.

Implicit Factors

In recent years researchers have examined relationships between a range of possible implicit decision-making factors and parent treatment/intervention decisions for intellectual disabilities (e.g., Sheehan, Kimona, Giles, Cooper, & Hassiotis, 2018; Zablotsky et al., 2015), Fragile X (e.g., Bailey Jr. et al., 2012; Laxman et al., 2018; Martin et al., 2013), and ASD (e.g., Denne, Hastings, & Hughes, 2017; Irvin, McBee, Boyd, Hume, & Odom, 2012; Mire, Hughes, Manis, & Goin-Kochel, 2018; Salomone, Charman, McConachie, & Warreyn, 2015; Zablotsky et al., 2015). The possible implicit factors examined can be broadly classified as either child-related factors, parent-related factors, or family-related factors (Wilson et al., 2018).

Although age was a factor that was explicitly stated as influencing parent decision-making in a number of studies, it has also been widely examined, using correlational approaches, in cases where the parents did not explicitly state that the child's age was a factor that they considered in their decision-making (Denne et al., 2017; Goin-Kochel, Myers, & Mackintosh, 2007; Laxman et al., 2018; Lindly, Thorburn, Heisler, Reyes, & Zuckerman, 2017; Martin et al., 2013; Miller, Schreck, Mulick, & Butter, 2012; Mire, Gealy, Kubiszyn, Burridge, & Goin-Kochel, 2017; Mire, Raff, Brewton, & Goin-Kochel, 2015; Owen-Smith et al. 2015; Salomone et al., 2015, 2016; Sheehan et al., 2018; Shepherd et al., 2018; Thomas, Ellis, McLaurin, Daniels, & Morrissey, 2007; Witwer & Lecavalier, 2005; Wong & Smith, 2006). Other child factors that have been examined include the gender of the child (Laxman et al., 2018; Lindly et al., 2017; Martin et al., 2013; Owen-Smith et al., 2015; Patten, Baranek, Watson, & Schultz, 2013; Salomone et al., 2015, 2016; Sheehan et al., 2018; Shepherd et al., 2018; Witwer & Lecavalier, 2005; Wong & Smith, 2006), the type of diagnosis and/or perceived severity of symptoms associated with diagnosis (Bailey Jr. et al., 2012; Hall & Riccio, 2012; Laxman et al., 2018; Lindly et al., 2017; Mire et al., 2017; Owen-Smith et al., 2015; Patten et al., 2013; Salomone et al., 2016; Sheehan et al., 2018; Thomas et al., 2007; Witwer & Lecavalier, 2005; Zablotsky et al., 2015), time since diagnosis (Miller et al., 2012; Salomone et al., 2016; Zuckerman, Lindly, & Chavez, 2017), comorbid conditions (Bailey Jr. et al., 2012; Laxman et al., 2018; Martin et al., 2013; Sheehan et al., 2018; Zablotsky et al., 2015), age at diagnosis (Lindly et al., 2017; Zuckerman et al., 2017), and presence of challenging behavior (Laxman et al., 2018; Sheehan et al., 2018).

Possible relationships between treatment/intervention decisions and family-related factors have also been examined in the literature. Family income or socioeconomic status has been examined across several studies, and the findings have been mixed (Bailey Jr. et al., 2012; Denne et al., 2017; Irvin et al., 2012; Laxman et al., 2018; Miller et al., 2012; Mire et al., 2017; Owen-Smith et al., 2015; Patten et al., 2013; Thomas et al., 2007); family size was examined in only a small number of studies (Lindly et al., 2017; Shepherd et al., 2018), as was health insurance coverage (Lindly et al., 2017; Thomas et al., 2007; Zablotsky et al., 2015). In comparison, the geographic location of the family was relatively more widely examined (Lindly et al., 2017; Mire et al., 2018; Salomone et al., 2015, 2016; Thomas et al., 2007). Examination of the relationship between ethnic background and treatment/intervention use has mainly occurred in the North American context, where the results have been mixed (Hall & Riccio, 2012; Irvin et al., 2012; Lindly et al., 2017; Owen-Smith et al., 2015; Patten et al., 2013; Reyes et al., 2018; Thomas et al., 2007). Other parent-specific factors examined have included the parents' age (Miller et al., 2012; Shepherd et al., 2018; Wong & Smith, 2006), parent education levels (Denne et al., 2017; Hall & Riccio, 2012; Lindly et al., 2017; Miller et al., 2012; Mire et al., 2017; Owen-Smith et al., 2015; Patten et al., 2013; Salomone et al., 2015, 2016; Shepherd et al., 2018; Thomas et al., 2007; Wong & Smith, 2006), marital status (Hall & Riccio, 2012; Owen-Smith et al., 2015), parents' beliefs or perceptions about their child's disability (including etiology; Dardennes et al., 2011; Lindly et al., 2017; Mire et al., 2017; Reyes et al., 2018), and parent/caregiver stress (Irvin et al., 2012; Thomas et al., 2007).

In general, when relationships between implicit factors and intervention/treatment use have been examined, few significant relationships have been found. Where significant relationships have been found, these findings have not necessarily been replicated in other studies. For example, some beliefs about ASD have correlated with the use of certain treatments and interventions, but the specific beliefs and treatments/interventions these are associated with have varied from study to study (see Dardennes et al., 2011; Lindly et al., 2017; Mire et al., 2017; Reyes et al., 2018). This is possibly a reflection of the idiosyncratic nature of parent decision-making.

Nevertheless, a few consistent findings have emerged. The relationship between the child's gender and treatment/intervention use was examined across a number of studies and was not found to be associated with the use of a particular treatment or intervention in the majority of studies in which it was examined (See Laxman et al., 2018; Lindly et al., 2017; Wilson et al., 2018). Another consistent finding that has emerged across the studies is that when parent education levels and CAM use have been examined, those who are more highly educated have been found to be more likely to use CAM interventions (Hall & Riccio, 2012; Owen-Smith et al., 2015; Patten et al., 2013; Salomone et al., 2015; Wong & Smith, 2006).

Summary

The majority of research related to parent decision-making regarding treatments and interventions has been conducted with parents of preschoolers and school-age children with ASD. Factors influencing parent decision-making include both those explicitly declared by parents as influencing their decisions and other implicit underlying factors that have been examined using correlational research (Carlon et al., 2013; Wilson et al., 2018). The most frequently examined declared decision-making factors include advice or recommendations from others, child factors (such as individual child characteristics), and pragmatic considerations (such as cost, funding, and accessibility). There is limited research related to the weight or level of importance that parents place on the different factors as they make decisions, but the evidence to date indicates that multiple factors are considered by parents as they make decisions and that those factors that are most frequently reported in the research are not necessarily those that have the most influence on parent decision-making.

A vast range of implicit underlying parent, child, and family factors may also be related to parent decision-making. These potential relationships have been examined using correlational research. Few significant relationships have been found in studies examining implicit factors, and where relationships have been found these have not tended to be replicated across studies. There are two consistent findings that have emerged: (1) child gender has not appeared to be related to treatment and intervention decisions; and (2) higher parent education levels have tended to be correlated with the use of CAM treatments. With these two exceptions, overall, the findings in this area have been inconsistent, and this may be reflective of the complex interplay between a range of competing declared decision-making factors and idiosyncratic implicit child, parent, and family factors.

Decision-Making of Teachers

Introduction

Students with intellectual disabilities may be educated in a range of contexts, including inclusive settings where a regular class teacher is responsible for their education through to self-contained settings with a special educator. Teacher decision-making in any setting is complex, and using theories of natural decision-making may be best seen as a process for solving problems, with the solutions constrained by a range of teacher and contextual factors, including values, attitudes, beliefs, resourcing, and system demands (Robinson, 1998; Robinson & Donald, 2015). Conventional theories of decision-making assume that once teachers know about effective interventions, they will make a rational decision to use them and that they will monitor the impact on their students and retain effective practices. The well-documented research-to-practice gap in special education indicates that this does not happen (Gersten, Vaughn, Deshler, & Schiller, 1997). A first step in understanding teacher decision-making in natural contexts is to identify factors that may influence teacher decision-making and the characteristics of teachers that determine which factors are considered. The factors influencing decisions may be conflicting, and an understanding of teacher decision-making requires an understanding of how teachers resolve those conflicts and decide on a course of action (Robinson, 1998).

Evidence-Based Practice

Although the ideal of researchers and the thrust of recent policies such as No Child Left Behind in the US would be that in the main special educators and teachers in segregated and inclusive settings would choose to use evidence-based interventions with research evidence to support them, the research-to-practice gap is widely recognized as a problem within special education (Greenway, McCollow, Hudson, Peck, & Davis, 2013; McLeskey & Billingsley, 2008). Gersten et al. (1997) suggested principles that would support uptake and use of effective practices. Interventions must be feasible and practical for classrooms (the reality principle) and only involve a reasonable amount of change to teacher practices. The changes must be linked to student outcomes. Teachers need the opportunity to practice an intervention with feedback and to link the concepts underpinning research to classroom realities. Simply knowing about an intervention is not sufficient for a teacher to decide to use it.

In the literature addressing the research-to-practice gap, researchers have suggested a range of reasons why teachers do not use evidence-based practices, but there is little investigation of teacher perspectives on the use of research. For example, Cook and Cook (2004), in writing about the education of students with learning disabilities, suggest that within natural decision-making, teachers largely ignore research findings as a factor. They suggest this because teacher preparation does not present research well, there are difficulties in the translation of research to practice, and school cultures may not value research. Similarly, Hornby, Gable, and Evans (2013) summarized the reasons generally why special education teachers are resistant to evidence-based practice as distrust of educational research and interventions described as best practice, a preference for guidance from experienced teachers, a preference for existing practices and lack of appropriate education, and professional development. McLeskey and Billingsley (2008) also noted the two factors of inadequate teacher education about research and the nature of research itself but added other factors related to teaching contexts, such as large caseloads and lack of support that may not always support best practices.

Guckert, Mastropieri, and Scruggs (2016) interviewed highly qualified special educators teaching students with learning difficulties, autism, and severe disabilities and examined their documentation to determine their perceptions and use of evidence-based practices. They found that although all teachers believed they were using evidence-based practices, there was a wide range in awareness of evidence-based practice and research. The aware teachers trusted research and could explicitly state that a practice was evidence-based, used it, discussed it with others, and relied on it. The partially aware group did not provide specific descriptions of evidence-based practices and was hesitant about sharing research evidence with colleagues, sometimes implemented and occasionally relied on evidence-based practice. The unaware group was skeptical about using evidence-based practices and reported it was not common to discuss the source of strategies chosen with colleagues and that they sometimes implemented evidence-based practices but rarely relied on them. Aware teachers reported personalizing research in explicit and effective ways, while the partially aware teachers may not have always implemented strategies effectively and described barriers such as time demands. The unaware teachers were less aware of the sources and components of strategies and were more likely to make unsuitable adaptations.

Decision-Making and Intellectual Disabilities

There appears to be little research directly and specifically examining the decision-making of teachers in regard to students with intellectual disabilities (Greenway et al., 2013), although it appears to be an area of emerging interest, particularly in relation to decision-making for students with severe intellectual disabilities. Greenway et al. (2013) offer one of the few studies specifically investigating teacher decision-making for students with intellectual and developmental disabilities. They interviewed nine teachers about their experiences and perceptions regarding evidence-based practices. Like the participants in Guckert et al. (2016), they had varied understandings of evidence-based practices, but they also discussed their access to resources, interactions with other people and organizations, and the interactions between these. Some teachers valued the autonomy they had as special educators but noted it came with less accountability, less supervision, and less collaboration. They reported that in this context, district policies about evidence-based practices were absent or not seen by others as relevant. Teachers wanted evidence-based resources to be made available, such as curricula and more access to professional learning, consultancy, and research literature, so it appears access to sources of reliable information may limit the use of some practices.

Andzik, Chung, Doneski-Nicol, and Dollarhide (2017) interviewed teachers of students with intellectual or developmental disabilities about the factors affecting decisions to use augmentative and alternative communication (AAC). Teachers described barriers to implementing AAC which were similar to those reported in earlier studies such as the lack of training in AAC, limited preparation time, and variable support from speech language pathologists and others. These practical factors were seen to limit student access to AAC, as did teacher knowledge of assistive technology.

More specifically, Ruppar, Gaffney, and Dymond (2015) investigated teacher decision-making in respect to literacy for secondary students with severe disabilities most of whom had intellectual disabilities. They identified four major factors that contributed to teacher decision-making about literacy. Teacher beliefs concerning the student, learning, and teaching determined the degree of individualization of lessons. Their expectations of the student influenced the goals they set, with those teachers who held lower expectations of their students being more likely to have set restrictive goals. Teachers with high self-efficacy were less likely to seek expert help or professional learning. In addition, Ruppar et al. (2015) identified a range of contextual factors which interacted with the teacher factors including staffing, in-class assistance and teaching materials available, level of support from colleagues, administrators and policies, philosophies about curriculum, personal and professional experiences, and professional development.

This research then, mostly recent, which describes specifically teachers of students with intellectual disabilities, and mostly those teaching students with severe disability, has identified a range of interacting teacher and contextual factors. Within teacher factors include teacher understanding of evidence-based practices, teacher knowledge, teacher attitudes and beliefs about students and teaching, and teacher self-efficacy. Contextual factors include access to resources and reliable information about evidence-based practices, access to support and collaboration with others, administrative and policy issues, and limited time.

Despite this lack of research directly relevant to students with intellectual disabilities, research on the education of students with disability more generally offers further useful information on factors influencing teacher decision-making. Surveys of special educators explore reported practices in relation to teacher characteristics. In addition, investigations addressing the acceptability of interventions to special and general educators and research, often qualitative, which explores special educator reports of their beliefs, attitudes, and practices and the reasons they give for selecting interventions may all contribute.

Practices and Teacher Characteristics

Multiple surveys of special education teachers have revealed that although they may use evidence-based practices, it is common for them to also use unproven and disproven practices. Burns and Ysseldyke (2009) surveyed both special education teachers (about a third of whom taught students with intellectual disabilities) and school psychologists in the US and found that some sound interventions, such as direct instruction, were used frequently. However, there was also high frequency of some ineffective interventions; for example, modality instruction was used as frequently as applied behavior analysis (an intervention with strong empirical support). Although they did not directly examine teacher characteristics in relation to reported use, they suggest that factors such as perceived effectiveness, resource demands, alignment with the teacher's own theories, and intrusiveness may be relevant. They also noted that respondents were members of the Council for Exceptional Children and thus may have been more aware of current research and best practices than nonmembers.

Carter et al. (2011) replicated the Burns and Ysseldyke (2009) survey with Australian special educators, and although they found high levels of use of a number of evidence-based practices, such as direct instruction and applied behavior analysis, they also reported moderate-to-high levels of use of a number of interventions with poor research support, such as modality instruction and perceptual motor programs. There were differences in use depending on the nature of students taught, with social skills training used more with students with severe and profound disabilities. There was no correlation between practices used and qualifications in special education, although the sample was drawn from members of the Australian Association of Special Education, who like the Burns and Ysseldyke (2009) sample may have been more aware of current research. A survey of special education teachers in the Czech Republic (Carter, Strnadová, & Stephenson, 2012) revealed a similar pattern, with high frequency of use reported for the evidence-based practices, applied behavior analysis, and direct instruction but also frequent use of practices without strong empirical support, such as perceptual motor training. Here teachers of students with severe and profound disability generally reported more use of both evidence-based and non-evidence-based strategies, and use of perceptual motor and psycholinguistic training was associated with higher levels of special education training.

Knight, Huber, Kuntz, Carter, and Juarez (2018) surveyed special educators teaching students with intellectual disabilities and/or autism about instructional practices. They found considerable variability in the practices teachers reported that they used but, like previous surveys, found use of both evidence-based strategies such as direct instruction and non-evidence-based interventions such as sensory integration. They also looked at factors that influenced teacher decision-making and found teachers were more likely to use interventions when they had received training or resources related to the intervention in the previous year. Other influences were setting (more use of evidence-based practices in special education classes), caseload, and age of students. The educational level and experience of teachers had little relation to the use of evidence-based practices. The teachers themselves reported that student need, the skills to be taught, and their perceptions of effectiveness of the practice influenced decisions. Decision-making was not generally influenced by research support, expert advocacy, or preservice education, but also recent training and recommendations from other teachers were influential. Similarly, Lawson and Jones (2017) who focused more on minute-to-minute decision-making than intervention selection and who both interviewed and observed teachers found that knowledge of individual students was a factor in all decision-making. Lawson and Jones (2017) also noted consultation with others, training, and mandated programs as influences.

Despite the results from Carter et al. (2011, 2012) and Knight et al. (2018) suggesting that there is little relationship between qualifications in special education and self-reported intervention choices, other research suggests that the nature of teacher qualifications does have an impact on instructional decision-making for students with disability generally. Teachers with special education qualification get better academic results than those without (Feng & Sass, 2012; Hanushek, Kain, & Rivkin, 2002) which suggests use of more effective strategies. Survey research shows special educators who had completed relevant coursework or professional learning were more likely to report the use of effective practices regarding transition (Morningstar & Benitez, 2013). In a survey of special educators working with students with emotional and behavioral difficulties, Stormont, Reinke, and Herman (2011) found special educators were better able to identify evidence-based and non-evidence-based behavioral interventions and were more confident in their intervention choice than regular educators. Bell, Cihak, and Judge (2010) surveyed student teachers in an alternative special education licensure program and found knowledge and use of assistive technology were strongly related to teacher preparation. Participants in this survey also reported perceptions of barriers to the use of assistive technology which included lack of time, knowledge, and funding.

Teacher Attitudes and Beliefs

For students with intellectual disabilities who are educated in inclusive settings, there is also little specific research. There is a view that positive attitudes to inclusion are more likely to lead to teachers making decisions to implement effective practices but that teachers are likely to hold more negative attitudes to the inclusion of students with intellectual disabilities (Avramidis & Norwich, 2002). Carlson, Hemmings, Wurf, and Reupert (2012) noted the importance of positive teacher attitudes to inclusion as an element motivating the use of effective practices. They observed and interviewed six teachers, some of whom taught students with intellectual disabilities, described by their principals as exemplary inclusive teachers. All used evidence-based strategies such as feedback, cooperative learning, and elements of direct instruction. The researchers suggested that other evidence-based strategies such as mastery learning were not used because teachers did not know about them. Use of effective practices was supported by collaboration with families and specialists, good support systems, and appropriate professional learning.

There is little direct evidence linking beliefs and attitudes to intervention decisions, beyond the negative attitudes to inclusion for students with disability. Jordan, Glenn, and McGhie-Richmond (2010) suggested perhaps a quarter of teachers in inclusive settings may believe that a disability is a fixed condition and that students may not benefit from instruction. They make minimal adaptations to instruction and prefer a pullout service model, taking less responsibility for student learning. Similarly, Ruppar, Roberts, and Olson (2018) linked deficit views of disability as fixed and located in the student to different visions of teacher expertise characterized by different teacher behaviors. They interviewed teachers of students with significant disabilities, including intellectual disabilities, their supervisors, and university personnel preparing special educators to explore their visions of expertise. They identified two contrasting visions, strength-based and deficit-based, which were associated with instructional approaches. In the strength-based view, teachers used their knowledge of the student to choose age-appropriate curriculum content and instructional designs, based on student strengths and abilities. In contrast, in a deficit-based vision, the focus was on caregiving and managing behavior rather than instruction. Roberts, Ruppar, and Olson (2018), noting the influence of educational leaders on instructional decision-making, interviewed principals and administrators and found that these leaders "were not able to articulate evidence-based instructional practices" (p. 14). They tended to emphasize caregiving and thus could be considered to hold a deficit view and low expectations of students with severe disabilities and influence teacher practices through these beliefs.

Acceptability

Research on the acceptability of interventions to teachers also provides some relevant information related to teacher decision-making, although this work has been mostly carried out with teachers of students with learning difficulties or behavioral disorders and on behavioral intervention (Boardman, Arguelles, Vaughn, Hughes, & Klingner, 2005; Gajria & Salend, 1996). In relation to these teachers, Boardman et al. (2005, p. 168) stated that "Teachers sought instructional practices that were feasible, were appropriate for their students, were accompanied by all necessary materials and professional development support, and could be individualized for multilevel classrooms." This supported the earlier findings of Gajria and Salend (1996) from a review of research on treatment acceptability for adaptations made for students with disability in mainstream classes. They noted that although choice of adaptations was not solely related to perceived effectiveness, that was a consideration, but for behavioral interventions, the nature of the intervention, the time and resources needed, its theoretical base, and any negative side effects were also considered. For instruction, teachers generally made only superficial changes to lessons rather than adopting new strategies or adjusting curriculum. They preferred interventions that did not need additional resources, did not involve other people, were not time-consuming, and fitted in with their existing practices. Whinnery, Fuchs, and Fuchs (1991) found little difference between special and regular educators of students with mild disabilities in regard to the perceived effectiveness and acceptability of instructional strategies and social behavior interventions addressed in a teacher survey. As in other studies, acceptability was related to perceptions of interventions as effective, but they also noted that teachers may not be aware of some effective interventions.

Policies

Ruppar et al. (2015) noted that policy contexts are one of the factors that might affect teacher beliefs and policies might support or limit the decisions teachers make. The teachers in Greenway et al. (2013) indicated that there were either no relevant district policies for students with intellectual and developmental disabilities or that policies were waived and so policies had little impact on their decision-making. It may be that policies designed for typically developing students are not appropriate for students with disability. Similarly, Boardman et al. (2005) noted that the teachers in their study (teachers of students with learning difficulties or behavior disorders) were often left to make their own decisions, even when districts suggested particular programs. Some studies have explored the impact of district and school policies on teacher behavior. Timberlake (2016) interviewed teachers of students with significant cognitive disabilities and found that they took responsibility for curriculum and instructional decisions, although they did recognize some policy boundaries such as graduation requirements and alternate assessments. Hudson et al. (2016) found special educators expressed varying levels of confidence in curriculum decisions made at higher levels and did not always agree that policy decisions around evidence-based practices for general education were applicable to their students. Lawson and Jones (2017) thought teachers of students with severe intellectual disabilities in Florida, USA, were more subject to policy decisions than those in the UK.

Summary

Overall then, the literature on special educators generally is consonant with research on teachers of students with intellectual disabilities and confirms there are multiple teacher and contextual factors that can interact in various ways to affect teacher decision-making. Table 29.1 presents an overall summary of the factors that have been identified.

Generally, it seems that the principles enunciated by Gersten et al. (1997) 20 years ago to support the use of evidence-based interventions by special educators still hold. Teachers are likely to use interventions that are feasible and practical within the resources available to them (time and materials). They need to be linked to student outcomes so that teachers perceive that the interventions are effective for their students. Where teachers receive education and training in the use of interventions, they are more likely to use them. Finally, teachers who know and understand research and the theories and principles underlying it are more likely to use evidence-based interventions supported by research.

Table 29.1 Factors influencing teacher decision-making

Within teacher factors
Knowledge and understanding, especially of evidence-based practices
Attitudes to the use of research
Teacher qualifications
Teacher training about specific interventions
Teacher theories and philosophies of learning and teaching
Teacher attitudes and beliefs about students with intellectual disabilities (strength-based or deficit-based)
Teacher perceptions of the effectiveness of an intervention
Teacher perceptions of student need
Teacher knowledge of the student
Teacher self-efficacy
Contextual factors
Resources available, particularly time
Feasibility of interventions
Supports available for an intervention
Side effects of an intervention
Fit with existing practices
Policies at school, district, and system level
Interaction and collaboration with others
Available staffing
Skills and knowledge to be taught
Setting
Level of student disability

Decision-Making of Other Professionals

Professionals other than teachers are also responsible for making decisions about the treatments and interventions used with individuals with intellectual disabilities. Notably, there has been much less research in this area than in that of parent or teacher decision-making. In particular, there is very little data related to treatment or intervention decision-making in group homes and vocational programs beyond examination of the treatment decisions of intellectual disability nurses (Williams, Roberts, Irvine, & Hastings, 2010) and some comparisons of medication and service use by individuals with intellectual disabilities living with their parents compared with those living in group homes (e.g., Harrington & Kang, 2016; Osunsanmi & Turk, 2016; Tsiouris, Kim, Brown, Pettinger, & Cohen, 2013). Treatments and interventions for challenging behaviors exhibited by individuals with intellectual disabilities within residential settings have received attention in recent years, with a particular focus on the use of psychotropic and antipsychotic medications (e.g., Bowring, Totsika, Hastings, Toogood, & McMahon, 2017; Doan et al., 2014; O'Dwyer et al., 2017). However, these studies have tended to focus on identifying a range of implicit factors that may be associated with the use of medication without consideration of who was making treatment decisions or the decision-making process undertaken (see Bowring et al., 2017; Doan et al., 2014; Kelly & Su, 2015; O'Dwyer et al., 2017). Thus, this literature provides little insight into how the decisions were made beyond the fact that challenging behavior exhibited by individuals with intellectual disabilities tends to be positively associated with medication use in residential settings (see Bowring et al., 2017; Doan et al., 2014). Other researchers have examined the experiences and practices of residential care staff regarding medication use, including their attitudes toward and understanding of medications (e.g., de Kuijper & van der Putten, 2017; Joos et al., 2014; Lalor & Poulson, 2013). This research has tended to focus on the outcomes of treatment decisions as opposed to how the decisions were initially made and therefore also provides little insight into treatment decision-making.

Although Williams et al. (2010) considered the process of decision-making employed by intellectual disability nurses, it should be noted that other research related to treatment/intervention decisions for adolescents and adults with intellectual disabilities has addressed the outcomes of treatment/intervention decisions as opposed to how those decisions were made (e.g., Harrington & Kang, 2016; Lalor & Poulson, 2013; O'Dwyer et al., 2017; Osunsanmi & Turk, 2016). Most of the research regarding the treatment and intervention decision-making of professionals relates to those working in early intervention, and, similar to the parent decision-making literature, much of this relates to the population of children diagnosed with ASD.

Special education and general teachers may work in early intervention services. The decision-making of teachers working in early intervention is often more collaborative than teachers working in schools. These teachers are often working in teams with other professionals, and best practice guidelines indicate that collaboration with other professionals and, in particular, the family is important (Division for Early Childhood, 2014). Therefore, the decision-making process for those teachers working in early intervention is different to those working in schools. In addition, much of the literature on decision-making of early intervention professionals does not differentiate between the decision-making of teachers and other professionals and paraprofessionals working in early intervention. Hence, it is more appropriate to include research literature related to the decision-making of teachers working in early intervention in the current section than the previous one.

A growing interest has been shown in the decision-making of early intervention professionals in recent years, particularly in relation to ASD interventions. Stahmer, Collings, and Palinkas (2005) conducted focus groups with early intervention providers in the US working with children with ASD regarding the intervention techniques and strategies used. Participants reported using a range of interventions and tended to rate the interventions that they used as being evidence-based, when only about a third of those nominated as evidence-based had research support. Paynter and colleagues have more recently conducted research in Australia examining the knowledge and use of practices by ASD early intervention service providers (Paynter et al., 2017; Paynter & Keen, 2015) and allied health professionals working with individuals with ASD (not necessarily within the early intervention age group; Paynter, Sulek, Luskin-Saxby, Trembath, & Keen, 2018) and the factors related to their decisions to employ different practices. They found that a range of evidence-based, emerging, and unsupported interventions were being used by the early intervention and allied health professionals but that evidence-based practices were being used more than emerging practices and emerging practices more than unsupported practices (Paynter et al., 2017, 2018; Paynter & Keen, 2015). The intervention practices reported to be used most frequently by the early intervention providers in Paynter et al. (2017) were evidence-based practices, and those used least frequently were unsupported interventions. There were, however, some unsupported interventions that were commonly being used more than once per week, including facilitated communication and multisensory environments. The role/profession of the early interventionist did not appear to be related to the use of these unsupported practices, as there were no significant differences between the use of these interventions by teachers, allied health professionals/social workers, and paraprofessionals (childcare workers). Teachers did report using academic interventions more frequently than allied health professionals/social workers and paraprofessionals (Paynter et al., 2017). This indicates that the professional background of the provider may influence some (but not all) of their intervention decisions.

The use by early intervention and allied health professionals of some interventions that do not have empirical support leads to the question of whether research evidence is a factor that these professionals consider in their decision-making. Stahmer et al. (2005) observed that none of the participants involved in their study reported engaging in professional reading. They concluded that:

> "It appears that program marketing, availability of training, provider preference, and external factors such as parent requests influence the use of specific practices more than whether the practice has any evidence of efficacy." (Stahmer et al., 2005, p. 74)

Similarly, Leong, Carter, and Stephenson (2013) surveyed early intervention providers in Malaysia and Singapore and found that even though half of the participants reported that research evidence on the efficacy for sensory integration was the reason that they employed this intervention, few reported using academic journals as sources of information about sensory integration. In contrast, 74.2% of the Australian early intervention professionals surveyed by Paynter et al. (2017) reported that they used research literature (e.g., academic books and academic journals) as sources of information about intervention practices for ASD, and 60.6% used treatment reviews (e.g., "good practice guidelines"). These participants rated both research literature and treatment reviews as trustworthy sources. However, more commonly used sources of information included internal professional development (100% of participants), therapists (e.g., speech therapists; 97%), workshops or other external professional development (89.4%), teachers (81.8%), general web searches (e.g., google; 78.8%), and parents of children with ASD (77.3%). The source of information most commonly used by the sample of allied health professionals working with individuals with ASD but not limited to the early intervention years was workshops or external professional development (100%; Paynter et al., 2018). Ninety-nine percent of participants in Paynter et al. (2018) reported receiving information from research literature, 77.8% from research and professional websites, and 72.7% from treatment reviews. Other frequently used sources included other therapists (99%), internal professional development or training (95.9%), autism associations or organizations and information from general web searches (both 89.9%), teachers and parents of children with ASD (both 89.8%), and professional associations (87.9%).

The allied health professionals surveyed by Paynter et al. (2018) rated research literature as the most trustworthy source of information, followed closely by workshops or other professional development, research and professional websites, treatment reviews, and internal professional development or training which also received a high mean rating of trustworthiness. The source of information with the highest mean trust rating received from early intervention professionals in the Paynter et al. (2017) study was internal professional development. Workshops or other professional development were also considered to be trustworthy, along with a range of other sources including therapists, research literature, and websites (Paynter et al., 2017). This indicates that early intervention and allied health professionals may attribute a similar weight of importance to information received from sources of research evidence to that received from other sources such as other professionals and professional development. Stahmer et al. (2005) also observed that the participants in their US sample tended to rate interventions as evidence-based if they had attended professional development workshops related to the intervention. In addition, 76.2% of the Malaysian early intervention providers surveyed by Leong et al. (2013) stated that they used sensory integration due to benefits described at a conference. Further, occupational therapists were reported to be the most common sources of both information and training regarding sensory integration in Malaysia and Singapore, and over 80% of the participants in the Leong et al. (2013) study reported that advice from occupational therapists was the reason that they used sensory integration. Findings from these studies in the US, Australia, and Asia suggest that workshops, professional development, and advice from other allied health professionals (such as speech therapists and occupational therapists) may have significant influence on early intervention and allied health professional's decision-making.

However, similar to parent decision-making, there are very limited data related to the relative importance placed by early intervention professionals on different factors in decision-making. Paynter et al. (2017) asked the intervention providers to rate the level of importance of 15 possible factors in their decisions to use intervention practices. The child's strengths and needs had the highest mean importance ratings, followed by the family's values, professional judgment, resources available to implement the intervention at their organization, and research evidence. The factors with the lowest mean importance rating were the interventionist's university or other post-school training, their own intuition or "gut feelings," whether they thought the child would enjoy it, and their hope that the intervention would work for the specific child (Paynter et al., 2017). Although 62.1% of participants indicated that research evidence was very important in their decision-making and a further 30.3% considered it somewhat important, there were 8.5% of early intervention providers in this study who considered research evidence to be unimportant in their decision-making (Paynter et al., 2017). Clearly, for at least some professionals, factors other than research evidence influence intervention decisions.

Goldbart, Chadwick, and Buell (2014) considered the rationales provided by speech therapists in the UK for the intervention approaches that they employed with children and adults with profound and multiple disabilities. Although this study cannot provide insight into the weight of importance placed by these professionals on each of the factors in decision-making, it does provide further insight into the range of factors that influence decision-making. The most frequently stated rationales for the use of intervention approaches were the empowerment, development, and behavioral needs of the individual, followed by the opportunity to develop the communication environment and family and carer skills. Unlike the Paynter et al. (2017) sample, these professionals did not frequently consider the views and wishes of the family/carers as a rationale for using any of the specific approaches and also rarely stated that they considered the views of other professionals (Goldbart et al., 2014). Published research evidence was only sometimes provided as a rationale for the use of the intervention approaches examined. The importance of clinical expertise and theories was also mentioned occasionally across the intervention approaches. Some rationales are provided by the participants that were not anticipated by Goldbart et al. (2014) related to practical and organizational issues, such as availability of resources and service structures.

Related to practical and organizational factors that have been considered in the decision-making of professionals, Cheung, Trembath, Arciuli, and Togher (2013) surveyed Australian speech therapists working with individuals with ASD about specific workplace factors that may act as either barriers or enablers to implementing evidence-based practice. The vast majority of participants (97%) indicated that they considered the application of evidence-based practice (EBP) as necessary in the practice of speech-language pathology, and 76% indicated that they had searched for research evidence on a regular basis. Interestingly, these participants viewed themselves in the minority in terms of regular practice for speech therapists, with only 23% agreeing with the statement that “Most speech-language pathologists work according to the principles of EBP when providing services to children with ASD” (Cheung et al., 2013, p. 399), and 41% agreed that there was a divide between research and practice. This is unsurprising considering the evidence that there are some early intervention and allied health professionals who report using interventions that are not evidence-based and that research evidence is not always reported to be an important consideration in professional decision-making (Goldbart et al., 2014; Leong et al., 2013; Paynter et al., 2017, 2018; Stahmer et al. 2005). Workplace culture and support were identified by Cheung et al. (2013) as important features that may either enable or act as barriers to the implementation of evidence-based practice. One participant commented

> “Management staff of my organization are not clinicians and therefore have limited understanding of the need to use EBP. Resources for accessing research are therefore not deemed to be a priority and service delivery decisions are made from an administrative point of view, rather than being evidence-based.” (Cheung et al., 2013, p. 400)

Time constraints within the workplace and the cost of resources such as subscriptions to journals and professional development courses were identified as further barriers to completing professional reading and other professional development that participants believed were necessary for the implementation of evidence-based practice. Finally, less than half of the participants believed that they had an adequate understanding of the process of evidence-based practice involving “(a) defining questions, (b) identifying sources for clinical evidence, (c) finding research evidence, (d) appraising the literature, (e) applying findings to relevant clinical questions for a specific client’s needs, and, finally, (f) evaluating the effectiveness of this process” (Cheung et al., 2013, p. 401). This indicates that even in a sample of professionals who appear motivated to employ evidence-based interventions, there may be several barriers to the implementation of these interventions in practice.

Recommendations

Recommendations for Parents

Evidence reviewed in this chapter suggests that parents tend to consider, or are influenced by, a complex range of factors in decision-making. Consideration of research evidence, however, does not typically feature prominently in decision-making. In addition, providing guidance in interpreting research evidence may not necessarily influence parents once decisions have been made (Carlon et al., 2017). This is hardly surprising given the extensive research on human cognitive biases and the well-documented tendency to accept information that is consistent with an established or initial viewpoint and dismiss disconfirming evidence (e.g., Edwards & Smith, 1996; Taber & Lodge, 2006). This has a number of potential implications for interactions with parents.

It is likely to be of importance that parents are provided with clear quality information *before* they have committed to decisions regarding intervention. This should include “red flags” that are often associated with unproven and/or pseudoscientific interventions such as claims of a cure, interventions that address a wide range of different problems, or diagnoses and exclusive reliance on testimonials (see Stephenson, 2004; Travers, 2016). Such material should also include reliable information on the current evidence base for interventions. Unfortunately, while a number of such sites exist for families of children with ASD, such as the Raising Children Network (Raising Children Network, 2006-2018) and Research Autism (The National Autistic Society, 2018) websites, there do not appear to be equivalent sites specifically for families of children with intellectual disabilities. There is also a need for professionals who provide first line of contact to be in a position to offer evidence-based recommendations. This means that such professionals must have an up-to-date knowledge of the evidence base for interventions as well as the skill to work with families to match and tailor interventions to their specific needs. On the flip side, parents should not be reserved in asking professionals to explain the basis for an intervention recommendation. In fact, as suggested previously, such information should be an integral part of the guidance offered by professionals.

Where parents do decide to invest resources into an intervention, particularly an unproven intervention, it is important to have a strategy to assist in evaluating effectiveness. Kay and Vyse (2005) offer some useful suggestions in this regard. They suggest that parents first identify the changes they expect from the selected intervention and attempt to operationally define the relevant behaviors. For example, parents might be anticipating new words to be spoken by their child or a decrease in self-injurious behavior. This approach reduces the risk that any unrelated maturation or development that is observed in the child will be incorrectly attributed to the intervention. Second, some decision rules can be developed with parents to decide when to continue or discontinue the intervention. A test can then be developed to evaluate the intervention. Simple single-case research designs can be adapted to the purpose with the most basic of these designs involving collection of data on the targeted behaviors before and during intervention. While attempting to rationalize a prior decision represents typical human behavior, particularly when individuals are invested in the decision (Kunda, 1990), the general approach suggested by Kay and Vyse (2005) provides parents with an objective approach for evaluating their decisions as well as offering a possible exit strategy.

Recommendations for Professionals

There is a clear evidence that educational professionals working with individuals with disabilities do not necessarily have an accurate perception of the research base supporting interventions (e.g., Burns & Ysseldyke, 2009; Carter et al., 2011,

2012) and that factors other than the strength of research evidence may weigh strongly on decision-making (Boardman et al., 2005; Carter, Stephenson, & Hopper, 2015). While a variety of contextual factors (e.g., resources, policies, time, student characteristics) should assist in shaping decisions, the degree of research support should undoubtedly be a key consideration. Thus, there is a strong case for improving understanding of the research base for commonly employed interventions among professionals including teachers and early interventionists. This needs to be addressed at the preservice level and updated with in-service training. Caution should be exercised, however, when accessing online sources to identify evidence-based practice. For example, Test, Kemp-Inman, Diegelmann, Hitt, and Bethune (2015) found 43% of websites identified as providing information to teachers about evidence-based practice in special education were not trustworthy.

There are implications for teacher educators as well. It is clear that teacher education programs and in-service professional learning are needed to build an understanding of research, evidence-based practices, and their applications to particular settings (Boardman et al., 2005; Chorzempa, Smith, & Sielo, 2018). Teachers need to look for training in particular evidence-based strategies, but those providing training need to be clear about the impacts on student outcomes and consider the features of interventions that make them acceptable to teachers. The work of Ruppar and colleagues (Ruppar et al., 2018) also demonstrates the need for teachers to carefully consider their attitudes to students with severe disabilities and adopt a position that recognizes the importance of education rather than care.

In addition to making decisions regarding their own practice, some professionals, particularly those in the area of early intervention, may be asked to provide advice to parents, caregivers or other professionals. For example, Leong and colleagues (Leong et al., 2013; Leong, Stephenson, & Carter, 2011) found that advice from occupational therapists was a major factor in the adoption of sensory integration therapy in early intervention centers in Malaysia and Singapore. In addition, there is evidence that parents both value (Carlon, Carter, & Stephenson, 2015; Dinora & Bogenschutz, 2018; Grant et al., 2016) and may desire more guidance from professionals (Carlon, Stephenson, & Carter, 2015; Grant et al., 2016; Levy et al., 2016). Consequently, a strong argument can be forwarded that it is critical that professionals are up to date with regard to the evidence base for interventions (Carlon, Carter, & Stephenson, 2015; Sansosti et al., 2012; Trembath, Hawtree, Arciuli, & Caithness, 2016). In addition, clearly communicating the research base for interventions to parents can be considered an important component of professional practice (Trembath et al., 2016), and this argument logically can be extended to communication with other professionals. Further, Leong et al. (2013) have argued that organized efforts to establish professional relationships between academics and service providers may assist in the dissemination of accurate information to professionals.

Recommendations for Future Research

A number of recommendations for future research arise from this chapter. We currently have substantial research on decision-making of parents of children with ASD but very limited research on other diagnostic groups. Given the distinctiveness of ASD, it is not reasonable to assume that the results of research with this diagnostic category are automatically generalizable to other groups. There is a clear need for further research on other populations. Similarly, the vast majority of the existing research relates to children who are of preschool and school age. Consequently, there is a prescient need for research with regard to decision-making processes with regard to adults who may be accessing vocational services or living in supported residential settings. In particular, there is very limited intervention research with regard to supporting parents and teachers and individuals with intellectual disabilities in more effective decision-making.

Research on teacher decision-making for students with intellectual disabilities is also limited. There has been recent interest in the decision-making of teachers of students with severe disabilities, including severe intellectual disabilities but mostly relating to teachers of segregated classes. There is much less research on teachers of students with mild intellectual disabilities, in both inclusive and segregated settings. Although there has been a lot of interest in promoting the use of evidence-based practices by teachers, there is little work that explores the reasons why teachers decide to use or not use evidence-based practices. Future directions for research would include more work on teacher decision-making, especially for students with less severe intellectual disabilities, and more work on teacher perspectives on the use of evidence-based practices.

References

Adams, D., Handley, L., Simkiss, D., Walls, E., Jones, A., Knapp, M., … Oliver, C. (2016). Service use and access in young children with an intellectual disability or global developmental delay: Associations with challenging behaviour. *Journal of Intellectual and Developmental Disability, 43*, 232–241. https://doi.org/10.3109/13668250.2016.1238448

Andzik, N. R., Chung, Y.-C., Doneski-Nicol, J., & Dollarhide, C. T. (2017). AAC services in schools: A special educator's perspective. *International Journal of Developmental Disabilities, 65*, 89. https://doi.org/10.1080/20473869.2017.1368909. Advance online publication.

Avramidis, E., & Norwich, B. (2002). Teachers' attitudes towards integration / inclusion: A review of the literature. *European Journal of Special Needs Education, 17*, 129–147. https://doi.org/10.1080/08856250210129056

Bailey, D. B., Jr., Raspa, M., Bishop, E., Olmsted, M., Mallya, U. G., & Berry-Kravis, D. (2012). Medication utilization for targeted symptoms in children and adults with fragile X syndrome: US survey. *Journal of Developmental and Behavioral Pediatrics, 33*, 62–69. https://doi.org/10.1097/DBP.0b013e318236c0e1

Bell, S. M., Cihak, C. F., & Judge, S. (2010). A preliminary study: Do alternative certification route programs develop the necessary skills and knowledge in assistive technology? *International Journal of Special Education, 25*, 110–118.

Bilgic, A., Congologlu, A., Herguner, S., Turkoglu, S., Bahali, K., Gurkan, K., … Turkbay, T. (2013). Use of complementary and alternative medicine in children with autism spectrum disorders: A multicenter study. *Archives of Neuropsychiatry, 50*, 237–243. https://doi.org/10.4274/npa.y6389

Boardman, A. G., Arguelles, M. E., Vaughn, S., Hughes, M. T., & Klingner, J. (2005). Special education teachers' views of research-based practices. *Journal of Special Education, 39*, 168–180. https://doi.org/10.1177/00224669050390030401

Bowen, C., & Snow, P. (2017). *Making sense of interventions for children with developmental disorders*. Guildford: J&R Press.

Bowker, A., D'Angelo, N. M., Hicks, R., & Wells, K. (2011). Treatments for autism: Parental choices and perceptions of change. *Journal of Autism and Developmental Disorders, 41*, 1373–1382. https://doi.org/10.1007/s10803-010-1164-y

Bowring, D. L., Totsika, V., Hastings, R. P., Toogood, S., & McMahon, M. (2017). Prevalence of psychotropic medication use and association with challenging behaviour in adults with an intellectual disability. A total population study. *Journal of Intellectual Disability Resarch, 61*, 604–617. https://doi.org/10.1111/jir.12359

Buescher, A. V., Cidav, Z., Knapp, M., & Mandell, D. S. (2014). Costs of autism spectrum disorders in the United Kingdom and the United States. *JAMA Pediatrics, 168*, 721–728. https://doi.org/10.1001/jamapediatrics.2014.210

Burns, M. K., & Ysseldyke, J. E. (2009). Reported prevalence of evidence-based instructional practices in special education. *The Journal of Special Education, 43*, 3–11. https://doi.org/10.1177/0022466908315563

Carlon, S., Carter, M., & Stephenson, J. (2013). A review of declared factors identified by parents of children with autism spectrum disorders (ASD) in making intervention decisions. *Research in Autism Spectrum Disorders, 7*, 369–381. https://doi.org/10.1016/j.rasd.2012.10.009

Carlon, S., Carter, M., & Stephenson, J. (2015). Decision-making regarding early intervention by parents of children with autism spectrum disorder. *Journal of Developmental and Physical Disabilities, 27*, 285–305. https://doi.org/10.1007/s10882-014-9415-z

Carlon, S., Carter, M., & Stephenson, J. (2017). Pilot study of a parent guided website access package for early intervention decision-making for autism spectrum disorder. *Australasian Journal of Special Education, 41*, 141–156. https://doi.org/10.1017/jse.2017.7

Carlon, S., Stephenson, J., & Carter, M. (2015). Parent perspectives on sources of information about autism spectrum disorder interventions in Australia. *Australasian Journal of Special Education, 39*, 113–127. https://doi.org/10.1017/jse.2015.9

Carlson, L., Hemmings, B., Wurf, G., & Reupert, A. (2012). The instructional strategies and attitudes of effective inclusive teachers. *Special Education Perspectives, 21*, 7–20.

Carter, M., Stephenson, J., & Hopper, T. (2015). Factors in instructional decision-making, ratings of evidence and intended instructional practices of Australian final year teacher education students. *Australian Journal of Teacher Education, 40*, 85–103. https://doi.org/10.14221/ajte.2015v40n6.5

Carter, M., Stephenson, J., & Strnadová, I. (2011). Reported prevalence by Australian special educators of evidence-based instructional practices. *Australasian Journal of Special Education, 35*, 47–60. https://doi.org/10.1375/ajse.35.1.47

Carter, M., Strnadová, I., & Stephenson, J. (2012). Reported prevalence of evidence-based instructional practices by special educators in the Czech Republic. *European Journal of Special Needs Education, 27*, 319–335. https://doi.org/10.1080/08856257.2012.691229

Chang, Y. C., Lin, J. D., Tung, H. J., Chiang, P. H., & Hsu, S. W. (2014). Outpatient physical therapy utilization for children and adolescents with intellectual disabilities in Taiwan: A population-based nationwide study. *Research in Developmental Disabilities, 35*, 498–505. https://doi.org/10.1016/j.ridd.2013.12.001

Cheung, G., Trembath, D., Arciuli, J., & Togher, L. (2013). The impact of workplace factors on evidence-based speech-language pathology practice for children with autism spectrum disorders. *International Journal of Speech-Language Pathology, 15*, 396–406. https://doi.org/10.3109/17549507.2012.714797

Chorzempa, B. F., Smith, M. D., & Sielo, J. M. (2018). Practice-based evidence: A model for helping educators make evidence-based decisions. *Teacher Education and Special Education, 42*, 82. https://doi.org/10.1177/0888406418767254. Advance online publication.

Christon, L. M., Mackintosh, V. H., & Myers, B. J. (2010). Use of complementary and alternative medicine (CAM) treatments by parents of children with autism spectrum disorders. *Research in Autism Spectrum Disorders, 4*, 249–259. https://doi.org/10.1016/j.rasd.2009.09.013

Cook, B. G., & Cook, L. (2004). Bringing science into the classroom by basing craft on research. *Journal of Learning Disabilities, 37*, 240–247. https://doi.org/10.1177/00222194040370030901

Courtade, G. R., Test, D. W., & Cook, B. G. (2015). Evidence-based practices for learners with severe intellectual disability. *Research and Practice for Persons with Severe Disabilities, 39*, 305–318. https://doi.org/10.1177/1540796914566711

Dardennes, R. M., Al Anbar, N. N., Prado-Netto, A., Kaye, K., Contejean, Y., & Al Anbar, N. N. (2011). Treating the cause of illness rather than the symptoms: Parental causal beliefs and treatment choices in autism spectrum disorder. *Research in Developmental Disabilities, 32*, 1137–1146. https://doi.org/10.1016/j.ridd.2011.01.010

de Kuijper, G., & van der Putten, A. A. J. (2017). Knowledge and expectations of direct support professionals towards effects of psychotropic drug use in people with intellectual disabilities. *Journal of Applied Research in Intellectual Disabilities, 30*(S1), 1–9. https://doi.org/10.1111/jar.12357

Denne, L. D., Hastings, R. P., & Hughes, C. J. (2017). Common approaches to intervention for the support and education of children with autism in the UK: An internet-based parent survey. *International Journal of Developmental Disabilities, 64*, 105–112. https://doi.org/10.1080/20473869.2016.1275439

Deyro, M. C., Simon, E. W., & Guay, J. (2016). Parental awareness of empirically established treatments for autism spectrum disorders. *Focus on Autism and Other Developmental Disabilities, 31*, 184–195. https://doi.org/10.1177/1088357614559210

Dinora, P., & Bogenschutz, M. (2018). Narratives on the factors that influence family decision making for young children with autism spectrum disorder. *Journal of Early Intervention, 40*, 195. https://doi.org/10.1177/1053815118760313. Advance online publication.

Dinora, P., Bogenschutz, M., & Lynch, K. (2017). Factors that may influence parent treatment decision making for young children with autism spectrum disorder. *Journal of Social Work in Disabililty and Rehabilitation, 16*, 377–395. https://doi.org/10.1080/1536710X.2017.1392395

Division for Early Childhood. (2014). DEC recommended practices in early intervention/early childhood special education. Retreived from http://www.dec-sped.org/recommendedpractices

Doan, T., Ware, R., McPherson, L., van Dooren, K., Bain, C., Carrington, S., … Lennox, N. (2014). Psychotropic medication use in adolescents with intellectual disability living in the community. *Pharmacoepidemiology and Drug Safety, 23*, 69–76. https://doi.org/10.1002/pds.3484

Doran, C. M., Einfeld, S. L., Madden, R. H., Otim, M., Horstead, S. K., Ellis, L. A., & Emerson, E. (2012). How much does intellectual disability really cost? First estimates for Australia. *Journal of Intellectual and Developmental Disability, 37*, 42–49. https://doi.org/10.3109/13668250.2011.648609

Edwards, A. G., Brebner, C. M., McCormack, P. F., & MacDougall, C. J. (2018). From 'Parent' to 'Expert': How parents of children with autism spectrum disorder make decisions about which intervention approaches to access. *Journal of Autism and Developmental Disorders, 48*, 2122. https://doi.org/10.1007/s10803-018-3473-5. Advance online publication.

Edwards, K., & Smith, E. E. (1996). A disconfirmation bias in the evaluation of arguments. *Journal of Personality and Social Psychology, 71*, 5–24.

Feng, L., & Sass, T. R. (2012). *What makes special-education teachers special? Teacher training and achievement of students with disabilities*. Atlanta, GA: Andrew Young School of Policy Studies, Georgia State University.

Finke, E. H., Drager, K. D. R., & Serpentine, E. C. (2015). "It's not humanly possible to do everything": Perspectives on intervention decision-making processes of parents of children with autism spectrum disorders. *Perspectives on Language Learning and Education, 22*, 13–21. https://doi.org/10.1044/lle22.1.13

Foran, S., & Sweeney, J. (2010). Accessing specialist early intervention services for pre-school children. *Learning Disability Practice, 13*, 30–35. https://doi.org/10.7748/ldp2010.03.13.2.30.c7606

Gajria, M., & Salend, S. J. (1996). Treatment acceptability: A critical dimension for overcoming teacher resistance to implementing adaptations for mainstreamed students. *Reading and Writing Quarterly, 12*, 91–108. https://doi.org/10.1080/1057356960120107

Gersten, R., Vaughn, S., Deshler, D., & Schiller, E. (1997). What we know about using research findings: Implications for improving special education practice. *Journal of Learning Disabilities, 30*, 466–476. https://doi.org/10.1177/002221949703000501

Goin-Kochel, R. P., Mackintosh, V. H., & Myers, B. J. (2009). Parental reports on the efficacy of treatments and therapies for their children with autism spectrum disorders. *Research in Autism Spectrum Disorders, 3*, 528–537. https://doi.org/10.1016/j.rasd.2008.11.001

Goin-Kochel, R. P., Myers, B. J., & Mackintosh, V. H. (2007). Parental reports on the use of treatments and therapies for children with autism spectrum disorders. *Research in Autism Spectrum Disorders, 1*, 195–209. https://doi.org/10.1016/j.rasd.2006.08.006

Goldbart, J., Chadwick, D., & Buell, S. (2014). Speech and language therapists' approaches to communication intervention with children and adults with profound and multiple learning disability. *International Journal of Language and Communication Disoders, 49*, 687–701. https://doi.org/10.1111/1460-6984.12098

Grant, N., Rodger, S., & Hoffmann, T. (2016). Intervention decision-making processes and information preferences of parents of children with autism spectrum disorders. *Child: Care Health and Development, 42*, 125–134. https://doi.org/10.1111/cch.12296

Greenway, R., McCollow, M., Hudson, R. F., Peck, C., & Davis, C. A. (2013). Autonomy and accountability: Teacher perspectives on evidence-based practice and decision-making for students with intellectual and developmental disabilities. *Education and Training in Autism and Developmental Disabilities, 48*, 456–468.

Guckert, M., Mastropieri, M. A., & Scruggs, T. E. (2016). Personalizing research: Special educators' awareness of evidence-based practice. *Exceptionality, 24*, 63–78. https://doi.org/10.1080/09362835.2014.986607

Haessler, F., Gaese, F., Huss, M., Kretschmar, C., Brinkman, M., Peters, H., … Pittrow, D. (2016). Characterization, treatment patterns, and patient-related outcomes of patients with fragile X syndrome in Germany: Final results of the observational EXPLAIN-FXS study. *BMC Psychiatry, 16*, 318. https://doi.org/10.1186/s12888-016-1020-5

Hall, S. E., & Riccio, C. A. (2012). Complementary and alternative treatment use for autism spectrum disorders. *Complementary Therapies in Clinical Practice, 18*, 159–163. https://doi.org/10.1016/j.ctcp.2012.03.004

Hanson, E., Kalish, L. A., Bunce, E., Curtis, C., McDaniel, S., Ware, J., & Petry, J. (2007). Use of complementary and alternative medicine among children diagnosed with autism spectrum disorder. *Journal of Autism and Developmental Disorders, 37*, 628–636. https://doi.org/10.1007/s10803-006-0192-0

Hanushek, E. A., Kain, J. F., & Rivkin, S. G. (2002). Inferring program effects for special populations: Does special education raise achievement for students with disabilities? *Review of Economics and Statistics, 84*, 584–599. https://doi.org/10.1162/003465302760556431

Harrington, C., & Kang, T. (2016). Disparities in service use and expenditures for people with intellectual and developmental disabilities in California in 2005 and 2013. *Intellectual and Developmental Disabilities, 54*, 1–18. https://doi.org/10.1352/1934-9556-54.1.1

Hornby, G., Gable, R. A., & Evans, W. (2013). Implementing evidence-based practice in education: What international literature reviews tell us and what they don't. *Preventing School Failure: Alternative Education for Children and Youth, 57*, 119–123. https://doi.org/10.1080/1045988x.2013.794326

Hudson, R. F., Davis, C. A., Blum, G., Greenway, R., Hackett, J., Kidwell, J., … Peck, C. A. (2016). A socio-cultural analysis of practitioner perspectives on implementation of evidence-based practice in special education. *The Journal of Special Education, 50*, 27–36. https://doi.org/10.1177/0022466915613592

Irvin, D. W., McBee, M., Boyd, B. A., Hume, K., & Odom, S. L. (2012). Child and family factors associated with the use of services for preschoolers with autism spectrum disorder. *Research in Autism Spectrum Disorders, 6*, 565–572. https://doi.org/10.1016/j.rasd.2011.07.018

Joos, E., Mehuys, E., Van Bocxlaer, J., Remon, J. P., Van Winckel, M., & Boussery, K. (2014). Medication management in Belgian residential care facilities for individuals with intellectual disability: An observational study. *Journal of Policy and Practice in Intellectual Disabilities, 11*, 27–33. https://doi.org/10.1111/jppi.12064

Jordan, A., Glenn, C., & McGhie-Richmond, D. (2010). The supporting effective teaching (SET) project: The relationship of inclusive teaching practices to teachers' beliefs about disability and ability and about their roles as teachers. *Teaching and Teacher Education, 26*, 259–266. https://doi.org/10.1016/j.tate.2009.03.005

Kay, S., & Vyse, S. (2005). Helping parents separate the wheat from the chaff: Putting autism treatments to the test. In J. W. Jacobson, R. M. Foxx, & J. A. Mulick (Eds.), *Controversial therapies for developmental disabilities: Fad, fashion, and science in professional practice* (pp. 265–277). Mahwah, NJ: Lawrence Erlbaum.

Kelly, S., & Su, Y. (2015). Psychotropic and anticonvulsant medication: Individuals with intellectual and developmental disabilities who transitioned to the community from an institution. *Intellectual and Developmental Disabilities, 53*, 289–300. https://doi.org/10.1352/1934-9556-53.4.289

Knight, V. F., Huber, H. B., Kuntz, E. M., Carter, E. W., & Juarez, A. P. (2018). Instructional practices, priorities and preparedness for education students with autism and intellectual disability. *Focus on Autism and Other Developmental Disabilities, 34*, 3. https://doi.org/10.1177/1088357618755694. Advance online publication.

Kunda, Z. (1990). The case for motivated reasoning. *Psychological Bulletin, 108*, 480–498.

Lalor, J., & Poulson, L. (2013). Psychotropic medications and adults with intellectual disabilities: Care staff perspectives. *Advances in Mental Health and Intellectual Disabilities, 7*, 333–345. https://doi.org/10.1108/AMHID-03-2013-0017

Lawson, H., & Jones, P. (2017). Teachers' pedagogical decision-making and influences on this when teaching students with severe intellectual disabilities. *Journal of Research in Special Educational Needs, 18*, 196. https://doi.org/10.1111/1471-3802.12405. Advance online publication.

Laxman, D. J., Greenberg, J. S., DaWalt, L. S., Hong, J., Aman, M. G., & Mailick, M. (2018). Medication use by adolescents and adults with fragile X syndrome. *Journal of Intellectual Disability Research, 62*, 94–105. https://doi.org/10.1111/jir.12433

Leong, H. M., Carter, M., & Stephenson, J. (2013). Sensory integration therapy in Malaysia and Singapore: Sources of information and reasons for use in early intervention. *Education and Training in Autism and Developmental Disabilities, 48*, 421–435.

Leong, H. M., Stephenson, J., & Carter, M. (2011). The use of sensory integration therapy by intervention service providers in Malaysia. *International Journal of Disability, Development and Education, 58*, 341–358. https://doi.org/10.1080/1034912X.2011.626608

Levy, S. E., Frasso, R., Colantonio, S., Reed, H., Stein, G., Barg, F. K., … Fiks, A. G. (2016). Shared decision making and treatment decisions for young children with autism spectrum disorder. *Academic Pediatrics, 16*, 571–578. https://doi.org/10.1016/j.acap.2016.04.007

Lin, L. P., Kuan, C. Y., Hsu, S. W., Lee, T. N., Lai, C. I., Wu, J. L., & Din, J.-L. (2013). Outpatient visits and expenditures for children and adolescents diagnosed with autism spectrum disorders and co-occurring intellectual disability: An analysis of the national health insurance claims data. *Research in Autism Spectrum Disorders, 7*, 1625–1630. https://doi.org/10.1016/j.rasd.2013.09.003

Lindly, O. J., Thorburn, S., Heisler, K., Reyes, N. M., & Zuckerman, K. E. (2017). Parents' use of complementary health approaches for young children with autism spectrum disorder. *Journal of Autism and Developmental Disorders, 48*, 1803. https://doi.org/10.1007/s10803-017-3432-6

Mackintosh, V. H., Goin-Kochel, R. P., & Myers, B. J. (2012). What do you like/dislike about the treatments you're currently using? *Focus on Autism and Other Developmental Disabilities, 27*, 51–60. https://doi.org/10.1177/1088357611423542

Martin, G. E., Ausderau, K. K., Raspa, M., Bishop, E., Mallya, U., & Bailey, D. B., Jr. (2013). Therapy service use among individuals with fragile X syndrome: Findings from a US parent survey. *Journal of Intellectual Disabililty Research, 57*, 837–849. https://doi.org/10.1111/j.1365-2788.2012.01608.x

Matson, J. L., & Neal, D. (2009). Psychotropic medication use for challenging behaviors in persons with intellectual disabilities: An overview. *Research in Developmental Disabilities, 30*, 572–586. https://doi.org/10.1016/j.ridd.2008.08.007

Matson, J. L., & Williams, L. W. (2015). The curious selection process of treatments for autism spectrum disorders. *Research in Autism Spectrum Disorders, 9*, 21–25. https://doi.org/10.1016/j.rasd.2014.10.004

McLeskey, J., & Billingsley, B. S. (2008). How does the quality and stability of the teaching force influence the research-to-practice gap? A perspective on the teacher shortage in special education. *Remedial and Special Education, 29*, 293–305. https://doi.org/10.1177/0741932507312010

Miller, V. A., Schreck, K. A., Mulick, J. A., & Butter, E. (2012). Factors related to parents' choices of treatments for their children with autism spectrum disorders. *Research in Autism Spectrum Disorders, 6*, 87–95. https://doi.org/10.1016/j.rasd.2011.03.008

Mire, S. S., Gealy, W., Kubiszyn, T., Burridge, A. B., & Goin-Kochel, R. P. (2017). Parent perceptions about autism spectrum disorder influence treatment choices. *Focus on Autism and Other Developmental Disabilities, 32*, 305–318. https://doi.org/10.1177/1088357615610547

Mire, S. S., Hughes, K. R., Manis, J. K., & Goin-Kochel, R. P. (2018). Autism treatment: Families' use varies across U.S. regions. *Journal of Disability Policy Studies, 29*, 97. https://doi.org/10.1177/1044207318766597. Advance online publication.

Mire, S. S., Raff, N. S., Brewton, C. M., & Goin-Kochel, R. P. (2015). Age-related trends in treatment use for children with autism spectrum disorder. *Research in Autism Spectrum Disorders, 15-16*, 29–41. https://doi.org/10.1016/j.rasd.2015.03.001

Morningstar, M. E., & Benitez, D. T. (2013). Teacher training matters: The results of a multistate survey if secondary special educators regarding transition from school to adulthood. *Teacher Education and Special Education, 36*, 51–64. https://doi.org/10.1177/0888406412474022

O'Dwyer, M., Peklar, J., Mulryan, N., McCallion, P., McCarron, M., & Henman, M. C. (2017). Prevalence, patterns and factors associated with psychotropic use in older adults with intellectual disabilities in Ireland. *Journal Intellectual Disability Research, 61*, 969–983. https://doi.org/10.1111/jir.12391

Olsson, L. M., Elgmark Andersson, E., Granlund, M., & Huus, K. (2017). Habilitation service utilization patterns among children with mild intellectual disability. *Journal of Policy and Practice in Intellectual Disabilities, 14*, 233–239. https://doi.org/10.1111/jppi.12174

Osunsanmi, S., & Turk, J. (2016). Influence of age, gender, and living circumstances on patterns of attention-deficit/hyperactivity disorder medication use in children and adolescents with or without intellectual disabilities. *Journal of Child and Adolescent Psychopharmacoly, 26*, 828–834. https://doi.org/10.1089/cap.2014.0139

Owen-Smith, A. A., Bent, S., Lynch, F. L., Coleman, K. J., Yau, V. M., Pearson, K. A., … Croen, L. A. (2015). Prevalence and predictors of complementary and alternative medicine use in a large insured sample of children with autism spectrum disorders. *Research in Autism Spectrum Disorders, 17*, 40–51. https://doi.org/10.1016/j.rasd.2015.05.002

Paton, C., Flynn, A., Shingleton-Smith, A., McIntyre, S., Bhaumik, S., Rasmussen, J., … Barnes, T. (2011). Nature and quality of antipsychotic prescribing practice in UK psychiatry of intellectual disability services. *Journal of Intellectual Disability Research, 55*, 665–674. https://doi.org/10.1111/j.1365-2788.2011.01421.x

Patten, E., Baranek, G. T., Watson, L. R., & Schultz, B. (2013). Child and family characteristics influencing intervention choices in autism spectrum disorders. *Focus on Autism and Other Developmental Disabilites, 28*, 138–146. https://doi.org/10.1177/1088357612468028

Paynter, J., Sulek, R., Luskin-Saxby, S., Trembath, D., & Keen, D. (2018). Allied health professionals' knowledge and ase of ASD intervention practices. *Journal of Autism and Developmental Disorders, 48*, 2335. https://doi.org/10.1007/s10803-018-3505-1. Advance online publication.

Paynter, J. M., Ferguson, S., Fordyce, K., Joosten, A., Paku, S., Stephens, M., … Keen, D. (2017). Utilisation of evidence-based practices by ASD early intervention service providers. *Autism, 21*, 167–180. https://doi.org/10.1177/1362361316633032

Paynter, J. M., & Keen, D. (2015). Knowledge and use of intervention practices by community-based early intervention service providers. *Journal of Autism and Developmental Disorders, 45*, 1614–1623.

Prussing, E., Sobo, E. J., Walker, E., & Kurtin, P. S. (2005). Between 'desperation' and disability rights: A narrative analysis of complementary/alternative medicine use by parents for children with Down syndrome. *Social Science & Medicine, 60*, 587–598. https://doi.org/10.1016/j.socscimed.2004.05.020

Raising Children Network. (2006-2018). Parent guide to therapies. Retrieved April 9, 2018, from http://raisingchildren.net.au/parents_guide_to_therapies/parents_guide_to_therapies.html/context/1534

Rajkovic, M., Thompson, D., & Valentine, K. (2010). *Post-diagnosis support for children with autism spectrum disorder, their families and carers: Older children and young people*. Canberra: Commonwealth of Australia.

Reyes, N. M., Lindly, O. J., Chavez, A. E., Folan, A., Macias, K., Smith, K. A., … Zuckerman, K. (2018). Maternal beliefs about autism: A link between intervention services and autism severity in White and Latino mothers. *Research in Autism Spectrum Disorders, 51*, 38–48. https://doi.org/10.1016/j.rasd.2018.04.001

Robert, M., Leblanc, L., & Boyer, T. (2015). When satisfaction is not directly related to the support services received: Understanding parents' varied experiences with specialised services for children with developmental disabilities. *British Journal of Learning Disabilities, 43*, 168–177. https://doi.org/10.1111/bld.12092

Roberts, C. A., Ruppar, A. L., & Olson, A. J. (2018). Perceptions matter: Administrators' vision of instruction for students with severe disabilities. *Research and Practice for Persons with Severe Disabilities, 43*, 3–19. https://doi.org/10.1177/1540796917734931

Robinson, V. M. J. (1998). Methodology and the research-practice gap. *Educational Researchers, 27*, 17–26. https://doi.org/10.3102/0013189X027001017

Robinson, V. M. J., & Donald, R. (2015). On the job decision making: Understanding and evaluating how leaders solve problems. In S. Chitpin & C. W. Evers (Eds.), *Decision making in educational leadership: Principles, policies, and practices* (pp. 93–109). New York: Routledge.

Ruppar, A. L., Gaffney, J. S., & Dymond, S. K. (2015). Influences on teachers' decisions about literacy for secondary students with severe disabilities. *Exceptional Children, 81*, 209–226. https://doi.org/10.1177/0014402914551739

Ruppar, A. L., Roberts, C. A., & Olson, A. J. (2018). Is it all about loving the kids? Perceptions about expertise in special education. *Teaching and Teacher Education, 71*, 319–328. https://doi.org/10.1016/j.tate.2018.02.001

Salomone, E., Beranova, S., Bonnet-Brilhault, F., Briciet Lauritsen, M., Budisteanu, M., Buitelaar, J., … Charman, T. (2016). Use of early intervention for young children with autism spectrum disorder across Europe. *Autism, 20*, 233–249. https://doi.org/10.1177/1362361315577218

Salomone, E., Charman, T., McConachie, H., & Warreyn, P. (2015). Prevalence and correlates of use of complementary and alternative medicine in children with autism spectrum disorder in Europe. *European Journal of Pediatrics, 174*, 1277–1285. https://doi.org/10.1007/s00431-015-2531-7

Sansosti, F. J., Lavik, K. B., & Sansosti, J. M. (2012). Family experiences through the autism diagnostic process. *Focus on Autism and Other Developmental Disabilities, 27*, 81–92. https://doi.org/10.1177/1088357612446860

Sheehan, R., Kimona, K., Giles, A., Cooper, V., & Hassiotis, A. (2018). Findings from an online survey of family carer experience of the management of challenging behaviour in people with intellectual disabilities, with a focus on the use of psychotropic medication. *British Journal of Learning Disabilities, 46*, 82. https://doi.org/10.1111/bld.12216

Shepherd, D., Csako, R., Landon, J., Goedeke, S., & Ty, K. (2018). Documenting and understanding parent's intervention choices for their child with autism spectrum disorder. *Jouranl of Autism and Developmental Disorders, 48*, 988–1001. https://doi.org/10.1007/s10803-017-3395-7

Shyu, Y. I., Tsai, J. L., & Tsai, W. C. (2010). Explaining and selecting treatments for autism: Parental explanatory models in Taiwan. *Journal of Autism and Developmental Disorders, 40*, 1323–1331. https://doi.org/10.1007/s10803-010-0991-1

Stahmer, A. C., Collings, N. M., & Palinkas, L. A. (2005). Early intervention practices for children with autism: Descriptions from community providers. *Focus on Autism and Other Developmental Disabilities, 20*, 66–79. https://doi.org/10.1177/10883576050200020301

Stephenson, J. (2004). A teacher's guide to controversial practices. *Special Education Perspectives, 13*, 66–74.

Stormont, M., Reinke, W., & Herman, K. (2011). Teacher characteristics and ratings for evidence-based behavioral interventions. *Behavioral Disorders, 37*, 19–29. https://doi.org/10.1177/019874291103700103

Taber, C. S., & Lodge, M. (2006). Motivated skepticism in the evaluation of political beliefs. *American Journal of Political Science, 50*, 755–769. https://doi.org/10.1111/j.1540-5907.2006.00214.x

Test, D. W., Kemp-Inman, A., Diegelmann, K., Hitt, S. B., & Bethune, L. (2015). Are online sources for identifying evidence-based practices trustworthy? An evaluation. *Exceptional Children, 82*, 58–80. https://doi.org/10.1177/0014402915585477

The National Autistic Society. (2018). Our evaluations of autism interventions, treatments and therapies. Retrieved April 9, 2018, from http://researchautism.net/autism-interventions/our-evaluations-interventions

Thomas, K. C., Ellis, A. R., McLaurin, C., Daniels, J., & Morrissey, J. P. (2007). Access to care for autism-related services. *Journal of Autism and Developmental Disorders, 37*, 1902–1912. https://doi.org/10.1007/s10803-006-0323-7

Timberlake, M. T. (2016). The path to academic access for students with significant cognitive disabilities. *The Journal of Special Education, 49*, 199–208. https://doi.org/10.1177/0022466914554296

Travers, J. C. (2016). Evaluating claims to avoid pseudoscientific and unproven practices in special education. *Intervention in School and Clinic, 52*, 195–203. https://doi.org/10.1177/1053451216659466

Trembath, D., Hawtree, R., Arciuli, J., & Caithness, T. (2016). What do speech-language pathologists think parents expect when treating their children with autism spectrum disorder? *International Journal of Speech-Language Pathology, 18*, 250–258. https://doi.org/10.3109/17549507.2016.1139625

Tsiouris, J. A., Kim, S., Brown, W. T., Pettinger, J., & Cohen, I. L. (2013). Prevalence of psychotropic drug use in adults with intellectual disability: Positive and negative findings from a large scale study. *Journal of Autism and Developmental Disorders, 43*, 719–731. https://doi.org/10.1007/s10803-012-1617-6

Tzanakaki, P., Grindle, C., Hastings, R. P., Hughes, C. J., Kovshoff, H., & Remington, B. (2012). How and why do parents choose early intensive behavioral intervention for their young child with autism? *Education and Training in Autism and Developmental Disabilities, 47*, 58–71.

Valentine, K. (2010). A consideration of medicalisation: Choice, engagement and other responsibilities of parents of children with autism spectrum disorder. *Social Science & Medicine, 71*, 950–957. https://doi.org/10.1016/j.socscimed.2010.06.010

Valentine, K., Rajkovic, M., Dinning, B., & Thompson, D. (2010). Post-diagnosis support for children with autism spectrum disorder, their families and carers. Canberra: Australian Government Department of Families, Housing, Community Services and Indigenous Affairs.

West, E. A., McCollow, M., Kidwell, J., Umbarger, G., & Cote, D. L. (2013). Current status of evidence-based practice for students with intellectual disability and autism spectrum disorders. *Education and Training in Autism and Developmental Disabilities, 48*, 443–455.

Whinnery, K., Fuchs, L. S., & Fuchs, D. (1991). General, special, and remedial teachers' acceptance of behavioural and instructional strategies for mainstreaming students with mild handicaps. *Remedial and Special Education, 12*, 6–17. https://doi.org/10.1177/074193259101200403

Wilkins, A., Leonard, H., Jacoby, P., Mackinnon, E., Clohessy, P., Forouhgi, S., & Slack-Smith, L. (2010). Evaluation of the processes of family-centred care for young children with intellectual disability in Western Australia. *Child: Care, Health and Development, 36*, 709–718. https://doi.org/10.1111/j.1365-2214.2010.01104.x

Williams, R. W., Roberts, G. W., Irvine, F. E., & Hastings, R. P. (2010). Exploring decision making in intellectual disability nursing practice: A qualitative study. *Journal of Intellectual Disabilities, 14*, 197–220. https://doi.org/10.1177/1744629510386289

Wilson, M., Hamilton, D., Whelan, T., & Pilkington, P. (2018). A systematic review of factors related to parents' treatment decisions for their children with autism spectrum disorders. *Research in Autism Spectrum Disorders, 48*, 17–35. https://doi.org/10.1016/j.rasd.2018.01.004

Witwer, A., & Lecavalier, L. (2005). Treatment incidence and patterns in children and adolescents with autism spectrum disorders. *Journal of Child and Adolescent Psychopharmacology, 15*, 671–681. https://doi.org/10.1089/cap.2005.15.671

Wodehouse, G., & McGill, P. (2009). Support for family carers of children and young people with developmental disabilities and challenging behaviour: What stops it being helpful? *Journal of Intellectual Disability Research, 53*, 644–653. https://doi.org/10.1111/j.1365-2788.2009.01163.x

Wong, H. H., & Smith, R. G. (2006). Patterns of complementary and alternative medical therapy use in children diagnosed with autism spectrum disorders. *Journal of Autism and Developmental Disorders, 36*, 901–909. https://doi.org/10.1007/s10803-006-0131-0

Wong, V. C. (2009). Use of complementary and alternative medicine (CAM) in autism spectrum disorder (ASD): Comparison of Chinese and western culture

(Part A). *Journal of Autism and Developmental Disorders, 39*, 454–463. https://doi.org/10.1007/s10803-008-0644-9

Yingling, M. E., Hock, R. M., Cohen, A. P., & McCaslin, E. M. (2017). Parent perceived challenges to treatment utilization in a publicly funded early intensive behavioral intervention program for children with autism spectrum disorder. *International Journal of Developmental Disabilities, 64*, 271. https://doi.org/10.1080/20473869.2017.1324352. Advance online publication.

Zablotsky, B., Pringle, B. A., Colpe, L. J., Kogan, M. D., Rice, C., & Blumberg, S. J. (2015). Service and treatment use among children diagnosed with autism spectrum disorders. *Journal of Developmental and Behavioral Pediatrics, 36*, 98–105. https://doi.org/10.1097/DBP.0000000000000127

Zuckerman, K., Lindly, O. J., & Chavez, A. E. (2017). Timeliness of autism spectrum disorder diagnosis and use of services among U.S. elementary school-aged children. *Psychiatric Services, 68*, 33–40. https://doi.org/10.1176/appi.ps.201500549

Part VIII

Treatment of Problem Behaviors

30

Behavioural Strategies to Address Noncompliance

Brittany Cook, Jessica Duris, Toby L. Martin,
and Lindsay McCombe

Introduction

In the following chapter, we will define noncompliance and discuss its importance, based on the understanding that noncompliance should necessarily be regarded as both a behavioural deficit and a behavioural excess. We'll describe general behavioural approaches to remediating both the deficit and the excess and will review specific empirically supported interventions that address each problem. This chapter is intended for readers who provide care or instruction to persons with intellectual/developmental disabilities (PWIDD) or autism spectrum disorders, either professionally or as family caregivers. A basic understanding of behavioural psychology (including principles such as positive reinforcement, extinction, and stimulus control) will be beneficial.

What Is Noncompliance?

An instance of noncompliance is a failure to behave as instructed within a brief interval following the instruction delivery. Noncompliance therefore presumes an instruction or request, that is, a verbal discriminative stimulus that specifies a desired behaviour or that has at least been correlated with consequences for the behaviour. "Noncompliance" may also refer to a history or pattern of instances of noncompliance. The term "noncompliance" is sometimes used in other ways, such as a failure to adhere to medical procedures or exercise regimes; our usage is confined to the definition provided above.

As a problem behaviour, noncompliance clearly represents a behaviour deficit: in the context of the instruction, a desired behaviour occurs too infrequently or too late. Noncompliance is also a behaviour excess, in that any behaviour other than what was instructed is likely to be unwanted. Often the behaviour that occurs in place of compliance would be undesirable in any context. That is, noncompliance often includes challenging behaviour (CB) that occurs in response to the instruction. Other excesses may be less troublesome, such as simply walking away or sitting quietly. The distinction between the deficit and excess implied by noncompliance guides the organization of the following sections.

B. Cook · J. Duris
Department of Psychology, University of Manitoba, Winnipeg, MB, Canada

T. L. Martin (✉)
Department of Psychology, University of Manitoba, Winnipeg, MB, Canada

St. Amant Research Centre, Winnipeg, MB, Canada
e-mail: toby.martin@umanitoba.ca

L. McCombe
St. Amant Research Centre, Winnipeg, MB, Canada

J. L. Matson (ed.), *Handbook of Intellectual Disabilities*, Autism and Child Psychopathology Series, https://doi.org/10.1007/978-3-030-20843-1_30

General Causes: Deficit

What might cause a deficit in compliance following an instruction? The problem is likely to be one of stimulus control and/or of inadequate repertoire. Instructions will tend to be effective (i.e. to have good stimulus control) when the probability of reinforcement for compliance has historically been greater than the probability of reinforcement for noncompliance. A fundamental strategy to reduce noncompliance is therefore to ensure appropriate reinforcement when compliance occurs and to try to reduce reinforcement for noncompliance. Reinforcement for compliance should be immediate, potent, and reliable. It can be faded and ultimately replaced by natural contingencies once desired behaviour is occurring more reliably.

When providing instructions, it's important to consider that behaviours have intrinsic consequences as well as socially mediated consequences. Some behaviours feel good to do; others are tiring or uncomfortable. Requesting behaviour that is especially effortful may require more extrinsic reinforcement to maintain.

Instructors should also ensure that instructions are distinct (i.e. the instruction sounds different from anything else that the person typically hears) and consistent, especially at first. Once compliance occurs more regularly, the form of the instruction may be varied to promote generality.

Even if attention is paid to stimulus control strategies (i.e. salient instructions and differential reinforcement), PWIDD may not comply with instructions if they lack the skill to perform the instructed behaviour. In such cases it would be important to develop the necessary topographies, for example, through a shaping or chaining procedure.

It may sometimes seem to caregivers that a person engages in noncompliance "on purpose". That is, the caregiver believes that the person can perform the requested behaviour and understands the instruction, but he or she simply chooses not to do it. In many such cases, the caregiver may be correct, but that doesn't mean the strategies recommended here are inapplicable. In behavioural terms, the purpose of a behaviour should be understood as the typical reinforcer that maintains it. In the case of wilful refusal to comply, a plausible reinforcer is the reaction (social attention) of the person who gave the instructions. (The caregiver may in fact recognize this by saying, e.g. "He's just refusing to do it to wind me up".) In such cases it would be best to reduce social attention for noncompliance and improve the reinforcement contingency for compliance.

General Causes: Excess

Like deficits, excesses following instructions are a stimulus control problem. Behaviours other than what was instructed will tend to occur when the probability of reinforcement for them has historically been greater than the probability of reinforcement for compliance.

CBs are those that can seriously impact the well-being and social opportunities of the behaving person and/or the people nearby. These behaviours include self-injury, aggression, property damage, and stereotypy. PWIDD are more likely to engage in these behaviours than the general population; studies show prevalence rates for PWIDD between 5% and 15% (e.g. Holden & Gitlesen, 2006; Rojahn, Matson, Lott, Esbensen, & Smalls, 2001).

CBs can severely degrade quality of life. They may have health impacts on the individual and his or her caregivers, both directly (i.e. through injury) and indirectly (i.e. by making it difficult to access health care). Severe CB is often treated by restrictive procedures including pharmacological interventions which may have negative side-effects. CBs are socially aberrant and may therefore contribute to exclusion from learning environments and other social contexts.

As noted above, CBs by PWIDD are often occasioned by an instruction or request. This relationship is so common that noncompliance is sometimes described as a type of CB or is defined as CB that follows an instruction. However, the relation between instructions and CB is better understood in the context of functional assessment.

Functional Assessment

The general recommendations and specific interventions described in this chapter should be guided by a functional assessment of the actual causes of a given individual's behaviour. Functional assessment is a systematic effort to identify the contingencies (arrangements of antecedents and consequences of behaviour) that maintain a given behaviour. Functional assessment may be performed descriptively by carefully observing and recording the events that precede and follow a problem behaviour. It can also be performed experimentally (it is then often called functional analysis), by manipulating antecedents and consequences in order to temporarily increase the behaviour's frequency. Decades of research on functional assessment procedures have shown that most problem behaviours are maintained by one or more of several common contingencies, including social attention, escape from demand, and direct (nonsocial) stimulation, sometimes called automatic reinforcement.

Escape from demand is especially relevant to noncompliance. In this contingency, a demand is presented that, perhaps through association with the activity requested, is aversive to the individual. Engaging in CB may cause the caregiver to "back off" and remove the request. The CB is thereby strengthened through negative reinforcement. A review of 173 studies (Matson et al., 2011) of functional assessment found that escape from demand was the most commonly determined cause of CB in three of the most frequently reported CB types (self-injury, aggression, property destruction).

Importance

It is important for caregivers to be prepared to deal with noncompliance in part because it is prevalent among PWIDD. Two total population studies in the United Kingdom (Emerson et al., 2001; Lowe et al., 2007) have found that serious CB occurs in approximately 10% of the PWIDD population. Among those who showed CB, noncompliance was described by Emerson et al. (2001) as the most prevalent form, occurring in over 80% of persons with serious CB. Lowe et al. (2007) found that generalized noncompliance occurred in approximately 70% (87% for children; 55% for adults) of PWIDD who display CB. Both studies reported that prevalence of noncompliance was even higher in children than in adults.

CB such as self-injury and aggression can have serious negative effects on the health and well-being of the individual performing them and on the people around him or her. If noncompliance is accompanied by these types of CB, appropriate intervention becomes all the most important. This is particularly true when the noncompliance and CB are functionally related (e.g. in the case of the "escape from demand" function where CB is evoked by an instruction or request and maintained by negative reinforcement).

Improving the behaviour deficit in cases of noncompliance may have multiple benefits. First, simply increasing the desirable behaviour may, independent of any other intervention, reduce the frequency of undesirable behaviour (Ducharme & Ng, 2012; Russo, Cataldo, & Cushing, 1981). Second, the behaviours specified by an instruction may be not merely desired or convenient but critical to the individual's safety or health; street safety and fire response are notable examples (Dixon, Bergstrom, Smith, & Tarbox, 2010; Wright & Wolery, 2011). Finally, promoting compliance may facilitate expanding a person's repertoire in other ways. For example, an individual who follows instructions may more easily come into contact with natural reinforcement contingencies. Complex skills (e.g. behaviour chains that are common in home or work settings) may be easier to build if the component behaviours can be reliably evoked using instructions. There is evidence that generalized compliance can be efficiently taught (Walters, Holborn, & Ediger, 2007), as well as presumably emerging through the teaching of multiple exemplars.

Search Method

We systematically searched relevant databases (CINAHL, Medline, PsycINFO, and Scopus) to gather peer-reviewed evidence for interventions that were aimed at either decreasing noncompliance or increasing compliant behaviours. We used search terms related to intellectual or developmental disabilities or learning disabilities, as this term is commonly used in the United Kingdom, and combined those results with the "AND" operator with "noncomplian∗" "OR" "complian∗".

Initially, 1799 records were identified and upon removal of duplicates, a total of 1149 articles were screened by two of the authors. A preliminary screen of articles to identify original research describing intervention studies and exclusion of compliance studies to medical interventions or policies further reduced our sample to 722 articles. A second strategized review of our sample focused on articles in which noncompliance was treated as a problem behaviour, which left us with a more manageable sample of 140 articles to consider. Ultimately, we primarily focused our attention on studies published in the twenty-first century so as to be mindful of current ethical practices and perceived social acceptability of the interventions presented. Interventions from 23 articles are described in the following sections of this chapter.

Interventions to Remediate Behavioural Deficits

Form of Instruction

The form of an instruction and the way in which it is delivered can influence the likelihood of compliance. Effective instruction delivery has been reported by caregivers as an acceptable addition to compliance training interventions with no perceived disadvantages (Bellipanni, Tingstrom, Olmi, & Roberts, 2013; Benoit, Edwards, Olmi, Wilczynski, & Mandal, 2001; Mandal, Olmi, Edwards, Tingstrom, & Benoit, 2000; Speights Roberts, Tingstrom, Olmi, & Bellipanni, 2008). Guidelines for effective instruction delivery include (a) being in close proximity to the individual; (b) obtaining eye contact prior to making a request and providing praise for doing so; (c) presenting the request as a command instead of a question; (d) waiting 5–10 s for compliance; and (e) delivering praise contingent on compliance (Mandal et al., 2000). As a whole, the strategy has successfully improved compliance in typically developing children as well as PWIDD (Bellipanni et al., 2013; Benoit et al., 2001; Mandal et al., 2000; Speights Roberts et al., 2008). Considerable, but less pronounced increases in compliance have also been observed when contingent praise for compliance was not included (Bellipanni et al., 2013; Speights Roberts et al., 2008). This indicates that while contingent praise is beneficial, the form of the instruction and its delivery alone can sufficiently improve levels of compliance.

The remaining components of effective instruction delivery have not been examined in isolation with PWIDD, but their inclusion is recommended. Firstly, being in close proximity to the individual and obtaining an attending response such as eye contact help to ensure that the individual is aware of the request being made. A single, clear directive is also critical given the nature of this population's disability. Multicomponent requests may be too complex and may thereby increase the likelihood of noncompliance. This includes refraining from providing a rationale for compliance, which may distract the individual and has been reported as ineffective even in typically developing children (Lipschultz & Wilder, 2017). Lastly, the person must be given an appropriate amount of time to process and comply with the instruction. For PWIDD, 5–10 s has consistently been used.

Presenting instructions in an affirmative rather than a negative format can result in greater compliance for PWIDD (Ducharme & Worling, 1994). These authors used high-probability command sequences (HPCS) comparing affirmative "do" and negative "don't" requests with two PWIDD. While compliance with "do" requests readily increased following the intervention, compliance with "don't" requests decreased for one individual and was negligible for the other. When symmetrical requests, that is, requests that require the same responses as the “don’t” requests but are presented in a “do” format, were implemented for the latter individual, levels of compliance substantially increased. For example, the symmetrical request “sit in a chair” was complied with more often than its negative counterpart “don’t lie on the floor”.

Presenting an instruction in a “do” format has also been shown to result in fewer CBs that are incompatible with compliance (Adelinis & Hagopian, 1999). Finally, criterion levels of compliance can be achieved in fewer sessions using “do” as compared to “don’t” requests (Montgomery & Ayllon, 1993).

Based on these findings, caregivers and practitioners should always consider the form and delivery of an instruction during compliance training. Instructions are phrased as “do” requests whenever possible, especially when noncompliance co-occurs with other forms of CB. Formatting instructions as “do” requests, although potentially beneficial on its own, should be used in conjunction with other compliance training methods. It is also important to note that compliance with one request form does not necessarily generalize to the other (Neef, Shafer, Egel, Cataldo, & Parrish, 1983). This suggests that training with “do” requests exclusively is unlikely to increase compliance with “don’t” requests. Alternative strategies are therefore required if caregivers wish to improve compliance with instructions phrased as “don’t” requests specifically.

High-Probability Command Sequences

High-probability command sequence (HPCS) is an intervention commonly used to promote compliance in PWIDD as well as those without intellectual disabilities. This method is also referred to as high-probability instructional sequence, high-probability request sequence, or high-p sequence (Lipschultz & Wilder, 2017). HPCS is frequently used in academic and home environments to promote compliance with related skills and activities (Belfiore, Basile, & Lee, 2008; Humm, Blampied, & Liberty, 2005; Lee, Belfiore, Scheeler, Hua, & Smith, 2004). HPCS has been shown to be an effective intervention method for increasing compliance among PWIDD, including those with autism spectrum disorders (ASD), Down syndrome (DS), and attention deficit hyperactivity disorder (ADHD) (Belfiore et al., 2008; Radley & Dart, 2016).

HPCS is the presentation of a series of instructions or requests for which compliance is highly likely followed by an instruction with a lower probability of compliance (Mace et al., 1988). The result is increased likelihood of compliance for the low-probability request. This method operates on the analogy of behavioural momentum: behaviour reinforced at higher rates will have greater resistance to changes in reinforcement conditions (Nevin, Mandell, & Atak, 1983). In the case of HPCS, the behavioural momentum of compliance with high-probability instructions transfers to the low-probability instruction, making it more probable that the person will comply with the low-probability instruction.

To implement a HPCS intervention, a practitioner, behaviour analyst, or parent must identify the setting where the relevant instructions or requests will be administered. After the applicable setting is determined, a list of instructions and/or requests that are important for the individual to comply with can be created. The probability of responding in the absence of reinforcement must then be determined for each instruction or request to develop a hierarchy of high- to low-probability requests. Prior to implementation, a preference assessment should be administered with a variety of items to determine which reinforcers are the most effective for the individual. During implementation of HPCS, a series of high-probability requests are made prior to introduction of a low-probability request to consequently increase compliance with the low-probability request. Reinforcers are presented after each instance of compliance and noncompliance is ignored. The presentation of high- and low-probability requests can be gradually randomized to promote generalization.

A large body of research supports HPCS as an antecedent intervention for promoting compliance in PWIDD and non-IDD populations, beginning with a seminal study by Mace et al. (1988). HPCS has been included in several reviews including most recently Lipschultz and Wilder (2017) and Radley and Dart (2016). Lipschultz and Wilder (2017) made several evidence-based recommendations for application of HPCS. First, HPCS interventions should consist of at least three high-probability instructions or requests before one low-probability instruction or request is presented. Additionally, high-probability instructions should be presented within 5 s of the previous trial, and low-priority requests should always be preceded by a successfully complied high-probability instruction (i.e. a low-probability request should not be given following noncompliance or an error).

An example of successful HPCS is given by Belfiore et al. (2008), who increased compliance with low-probability requests in a 7-year-old male with Down syndrome and moderate intellectual disability in a classroom setting. An ABAB experimental design was used to evaluate the teacher-designed and teacher-delivered intervention. A hierarchy of requests was constructed prior to phase 1, where high-probability requests were complied with 80% or more of the time and low-probability requests were complied with less than 40% of the time. Requests that were complied with 50–70% of the time were not included. Compliance for both low- and high-probability requests were reinforced with praise, and noncompliance was ignored.

The first baseline phase presented low-probability requests without HPCS. In the second phase, three to five high-probability requests preceded one low-probability request. During the return to baseline and subsequent intervention phases, the participant began taking Ritalin. A fading procedure occurred after the fourth phase where one high-probability request was then followed by one low-probability request to promote generalization of the conditions that typically occur in the classroom environment.

The intervention increased compliance with low-probability requests to an average of 78% and removal of the intervention (phase 3) resulted in a return to baseline levels of compliance for low-probability requests (M = 17%). When the intervention was reintroduced, there was a jump to 85% compliance with low-probability requests. When the participant began taking Ritalin and return to baseline occurred, compliance with low-probability requests averaged 25% and improved to 70% in phase 4 when the intervention was reintroduced. Compliance to low-probability requests remained high (M = 77%) when the fading procedure was implemented.

Medium-Probability Command Sequences (MPCS)

For an instruction or request to be considered high probability, it should be complied with or responded to correctly at least 80% of the time. This requirement may create a gap for lower functioning individuals that do not have high probability responding in their repertoire. In such cases, medium-probability requests with compliance 50–70% of the time can be used when high-probability requests do not exist for an individual.

The effectiveness of medium-probability requests was compared to high-probability requests by Romano and Roll (2000) using a simultaneous treatment design with a reversal component for three PWIDD. For each participant, a list of requests was compiled, and each one was requested ten times in order to compute a compliance percentage. Requests complied with less than 40% of the time were considered low-probability, medium-probability were 50–70%, and high-probability requests were 80% or higher.

Each participant started with a baseline phase where the experimenter presented low-probability requests on a fixed interval 1 min schedule. Compliance was praised and noncompliance was ignored. After stability was observed in the baseline phase, the intervention was implemented in which three to four medium- or high-probability requests were presented at 10 s intervals and preceded by one low-probability request.

The researchers found that both high- and medium-probability request sequences increased compliance to low-probability requests that followed for all three participants and there was no difference in effectiveness between the high- and medium-probability request sequences. Romano and Roll's (2000) study demonstrated the principles of behavioural momentum can be extended to medium-probability requests, and thus the authors deemed their experiment a successful replication of Mace et al. (1988).

Errorless Compliance Training

Errorless compliance training (ECT) is a compliance training intervention method developed by Ducharme and Popynick (1993). It is an approach that resembles HPCS but differs in that requests or commands of varied probabilities (not only 80% or greater) are used to fade in lower-probability requests. ECT has been shown to increase compliance in home and education settings. Additionally, it has been shown to be effective in many PWIDD and autism spectrum disorders (Radley & Dart, 2016).

ECT may involve parents, education professionals, behaviour analysts, and/or practitioner support when working with the student. These figures aid in determining a list of requests or commands that the student should comply with and then assist in observing the frequency in which the student complies with each request or command. The frequency of compliance for each request can then be used to build a hierarchy of the requests. Requests that are complied with at a high frequency (i.e. 80% or more of the time) would be level 1 requests. Subsequent levels would have lower frequencies relative to each other; for example, level 2 could have 51–79% compliance, level 3 could have 30–50% compliance, and level 4 could have <30% compliance. Generally, there are four levels used in a program with level 1 having the highest-probability requests and level 4 having the lowest-probability requests (Ducharme & Ng, 2012). By ranking the requests by highest to lowest probability, practitioners can arrange these levels appropriately for each student. The ability to arrange the probabilities in a hierarchy is beneficial as it allows flexibility when determining level cut-offs. This would be beneficial for individuals who may not have high-probability requests as the hierarchy could be constructed relative to the individual's highest probability request.

Initially during ECT, level 1 requests are made, and reinforcement is provided for compliance with the high-probability requests. Once high levels of compliance are established for these requests, lower level requests can slowly be faded in with the level 1 requests (e.g. once all level 1 requests are consistently complied with, level 2 requests can be faded in, etc.). The key is that the lower level requests are integrated with higher level requests at a slow enough rate to increase the compliance of low level requests as a consequence of the high level requests already being complied with. Once all level requests are integrated together, the student should have a high probability of complying with all requests even the requests first deemed low probability.

In an article by Ducharme and Ng (2012), ECT was used to increase compliance with academic-related requests (hence, errorless academic compliance training, or EACT) in three PWIDD in a special education classroom setting. One teacher and two support staff administered the interventions which were tailored to meet each student's needs. Trained administrators conducted all baseline and treatment sessions in the classroom where each administrator was paired with one participant.

A multiple baseline across subject design was used. Baseline rates of compliance and on/off task behaviour were recorded with high interobserver agreement. Prior to implementation, staff completed an EACT introductory workshop, an academic compliance probability checklist, a reinforcer checklist, and a group training during a second workshop. They then developed a hierarchy of requests and demands for each participant and made baseline observations. In this experiment, level 1 requests ranged from 76% to 100% compliance, level 2 ranged from 51% to 75% compliance, level 3 ranged from 26% to 50% compliance, and level 4 ranged from 0% to 25%

compliance, for all but one participant. A third workshop was held to train the administrators for the individual participant treatment procedures. The treatment phase began following this third workshop.

Treatment phase 1 involved reinforcing compliance of level 1 requests, using prompts as needed and ignoring noncompliance. A phase change occurred when a participant complied with at least 80% of requests. Advancement to the next phase involved transition sessions in which a combination of requests from the preceding phase was combined with the new level. Phases 2, 3, and 4 were procedurally the same as phase 1. Follow-up sessions were conducted after phase 4.

Implementation of EACT resulted in an increase in compliance for all four phases of the experiment where the least probable requests showed the greatest increases in compliance. These improvements were maintained at follow-up. Additionally, there were also increases in on-task behaviour and reductions in off-task and disruptive behaviour. This suggests that EACT was effective in improving compliance of all targeted requests and also improved covarying behaviours such as aggression, disruptive, and off-task behaviour.

Radley and Dart (2016) summarized the evidentiary support for ECT and found that it appears to consistently increase compliance of PWIDD. This conclusion is consistent with the fact that ECT is an extension of traditional HPCS, which has an even larger body of evidentiary support. A potential disadvantage of ECT is that it takes time to be implemented, which may be problematic for time-sensitive situations.

Response Effort Manipulation

To date, Fischetti et al. (2012) is the only study to have evaluated the effects of response effort manipulation in relation to compliance training. During their study, three PWIDD were instructed to put a highly preferred toy into a toy bin. Response effort was manipulated by decreasing the distance between the individual and the toy bin from 3 m to 0.3 m. Results indicate that the intervention was only effective for one of three children. Despite using gradual increments, it appears as though the distance between the child and the toy bin was unable to be increased without commensurate decreases in compliance. Complying with the instruction when the bin was 3 m away was only achieved using other compliance training strategies. These results suggest that response effort manipulation can increase compliance for some individuals but its application is limited. In general, tasks should always be in accordance with the skill level of the individual and should be made more manageable when appropriate.

Guided Compliance

Guided compliance is a procedure that involves the presentation of increasingly intrusive prompts contingent on noncompliance (Lipschultz & Wilder, 2017). Typically, it is carried out in accordance with Horner and Keilitz's (1975) least-to-most prompting procedure. The first step of the procedure is the delivery of a verbal prompt (e.g. an instruction). If the individual does not comply within a given time frame, a gestural or modelling prompt is subsequently provided. Continued noncompliance results in a final physical prompt in which the individual is physically guided through the task using the least amount of force necessary. Reinforcement in the form of social praise is usually delivered following compliance with verbal, gestural, and modelling prompts only.

Research evaluating the efficacy of guided compliance in the absence of other types of CB is scarce, but Smith and Lerman (1999) found it could be used successfully. In their study, caregivers of children with developmental disabilities were taught to use the three-step process of guided compliance. Following the initial instruction, caregivers waited 5 s for child compliance before presenting each successive prompt. Levels of compliance for each child improved from 0% to 70% and 23% to 71% respectively. When guided compliance was briefly removed for one individual and both compliance and noncompliance were ignored, compliance immediately decreased below baseline levels.

A multi-element design also allowed researchers to compare guided compliance and HPCS. The former was found to be substantially more effective, and caregivers were equally competent in their implementation of the two procedures.

Results from the Smith and Lerman (1999) study suggest that guided compliance may be particularly useful when noncompliance is due to a skill deficit since each trial inevitably results in the completion of the desired task. Unlike many other forms of compliance training, individuals learn what the appropriate response is as well as how to execute it. Furthermore, guided compliance can be considered a form of escape extinction wherein demands are continually presented regardless of the individual's behaviour. In this way any behaviours that are interfering with the individual's ability to comply with requests are no longer being reinforced by an escape from task demands. Consequently, there is a decrease in the frequency of escape-maintained behaviours and an increase in compliance. Guided compliance with the aim of simultaneously reducing CB and increasing compliance has been widely adopted (Lipschultz & Wilder, 2017).

Guided compliance can be an effective intervention, but it is important that the function of noncompliance is first identified. If the function of noncompliant behaviour is to gain access to social attention, guided compliance may reinforce noncompliance (Kern, Delaney, Hilt, Bailin, & Elliot, 2002). Wilder, Saulnier, Beavers, and Zonneveld (2008) have also reported instances where guided compliance has led to an increase in CB. This may be indicative of the potentially aversive properties of physical guidance and suggests that caregivers must be cautious in its application. Caregivers must also be aware that dependence on gestural, modelling, or physical prompts can develop and that additional procedures such as prompt fading may be required (MacDuff, Krantz, & McClannahan, 2001).

Functional Communication Training

Functional communication training (FCT) is an antecedent intervention method that can promote compliance in PWIDD. FCT is an intervention that teaches PWIDD to make appropriate verbal or nonverbal requests or statements to gain access to a reinforcer (Carr & Durand, 1985). Understanding the function of an individual's behaviour is fundamental for FCT, as it can be applied to both attention- and escape-maintained behaviours like noncompliance. If an individual's noncompliance is maintained by attention, then the therapist should teach the person to verbally or nonverbally ask for attention. For example, the individual should be taught to ask for positive attention instead of not complying with a request in order to receive negative attention. Or if a child does not comply with instructions in order to escape from demands in a school environment, FCT can be used to teach the child to ask for breaks instead of storming out and disrupting the class when they are asked to do their schoolwork.

According to Zangrillo, Fisher, Greer, Owen, and De Souza (2016), FCT should include four procedures. The first is that a qualified professional like a Board-Certified Behaviour Analyst conducts a functional assessment to identify the function of the noncompliance. Second, appropriate alternative responses that the individual is capable of emitting need to be identified. These behaviours will vary depending on the individual's physical, physiological, and cognitive abilities. For example, alternative responses may be written, verbal, ASL, pointing to a picture, etc. (Zangrillo et al., 2016). Third, the individual is reinforced for engaging in the alternative response. Lastly, reinforcement for the target response (i.e. noncompliance) is withheld in order to extinguish the behaviour. When first establishing the alternative response, a small fixed-ratio schedule of reinforcement is used, and as the behaviour develops, the reinforcement schedule is increased (Zangrillo et al., 2016).

FCT was first described by Carr and Durand (1985) and continues to be an effective intervention (Luiselli, 2009). FCT has been and still is very popular with many practitioners as it addresses a primary concern for PWIDD, that is, increasing verbal communication skills (Luiselli, 2009). Once the FCT repertoire is established, it can also be generalized to other requests which increase an individual's adaptive behaviour skills.

Self-Monitoring

Self-monitoring, sometimes referred to as self-recording or self-management, involves regulating one's own behaviours according to specified rules (Martin & Pear, 2015).

Self-monitoring strategies are most effective when they are simple and response effort is manageable relative to reinforcement contingencies (Bialas & Boon, 2010). Noncompliant behaviours in the classroom not only interfere with learning opportunities for the students themselves, but they also disrupt the learning of their peers. Self-monitoring is one method for addressing noncompliance that is especially advantageous in classroom settings as students record their own data, thereby freeing up teacher and educational aide resources (Bialas & Boon, 2010). Self-monitoring also improves student engagement in instructional tasks (Ennis, Lane, & Oakes, 2018). Limited research exists looking at the self-monitoring of compliance for PWIDD, but two studies have shown promise.

Bialas and Boon (2010) used a multiple baseline design across participants to evaluate a self-monitoring procedure during reading, language arts, and mathematics instruction. Participants were three male kindergarten students, aged 5.5–5.7 years, at risk for developmental disabilities in an inclusive classroom setting. The self-monitoring procedure involved a checklist the students used to record their behaviour and colour pictures taped to student desks to serve as visual prompts. Students also graphed their progress after each session. Teachers were instructed to provide no more than two verbal prompts per target behaviour request, and the visual prompts were used as needed. Reinforcers were those typically used in the classroom, such as stickers. Three cubes were placed in front of students at the start of each session. If noncompliant behaviours occurred, a cube was removed, and the opportunity for reinforcement was lost. If all three cubes were removed, regular classroom rules were applied (e.g. a 5 min timeout). During the baseline and maintenance phases, compliance was observed; meanwhile, the students did not use their checklists. The dependent variable was percentage of compliance for two target behaviours: (1) "listening to directions" by sitting quietly and attending to the teacher and (2) "repeating directions" to the teacher verbally.

Results revealed an almost immediate effect of the intervention on the dependent variable. All three participants achieved 100% compliance during the intervention phase, as well as during both of their maintenance assessments. Additionally, a social validity assessment demonstrated that the students liked the self-monitoring procedure and it helped them with their classroom behaviours.

Wadsworth, Hansen, and Wills (2015) evaluated teacher-monitoring and self-monitoring procedures targeting compliance in a special education classroom. Participants were two 9-year-old males and a 7-year-old female, all with an intellectual disability (either moderate or unspecified). Functional behavioural assessments established escape from demands as the function of noncompliance for all three students, and access to preferred items was an additional function for one of the students. A teacher prompting routine was followed during baseline and intervention phases. Four space token boards with pictures or words of the reward to be exchanged after the board was filled with tokens were used. Students earned tokens at a 1:1 ratio for each occurrence of compliance, operationalized as completing or initiating responses to teacher instructions or requests within 5 s. During the teacher-monitoring phase, the teacher added tokens for each instruction complied with. After 80% compliance was achieved for two sessions, teachers prompted students to self-monitor their behaviour and add their own tokens to the board for the self-monitoring phase. During this phase, spaces were added to the token board to thin the reinforcement schedules for token exchanges, and teachers had to agree with at least 75% of the self-administered tokens before students earned breaks or tangible rewards.

The introduction of the teacher-monitoring intervention improved mean student compliance between 29% and 54% as compared to baseline levels. Compliance was generally maintained for all students once the self-monitoring intervention was introduced and remained quite stable even when the reinforcement schedules were thinned. Additionally, social validity data suggested the interventions were effective and produced important results, and that teachers were highly likely to use the procedures in the future.

Self-monitoring is a fundamental adaptive skill that aides in promoting independence, motivation, and student engagement in the classroom (Bialas & Boon, 2010). Studies by Bialas and Boon (2010) and Wadsworth et al. (2015) demonstrated that elementary students with mild or moderate disabilities or who are at risk for intellectual disabilities can successfully self-monitor their compliant behaviours in classroom settings. This was an important practical implication as self-monitoring interventions require minimal teacher resources to implement. Furthermore, Wadsworth et al. (2015) showed self-monitoring to be comparable to the effectiveness of the teacher-monitoring intervention.

Although these studies demonstrated effectiveness of self-monitoring interventions for compliance, the generality of the results is limited. First, long-term maintenance data was not collected in either study, so the persistence of compliant behaviours is unknown. Second, small samples sizes for a narrow age range of participants were studied. The effectiveness with older populations remains unknown. Lastly, only individuals with mild or moderate intellectual disabilities were included in these studies, and it would appear inappropriate to use self-monitoring interventions with lower functioning individuals. However, self-monitoring procedures are relatively simple to implement and offer practical benefits to teachers.

Differential Reinforcement

Differential reinforcement is a schedule or procedure of reinforcement, whereby an individual's compliant behaviour is reinforced at a higher rate than his or her noncompliant behaviour; typically, the latter behaviour is on extinction. Three common procedures for addressing noncompliant behaviour in PWIDD are (1) differential reinforcement of low rates (DRL); (2) differential reinforcement of zero responding (DRO); and (3) differential reinforcement of alternative (DRA) behaviour (Martin & Pear, 2015).

DRL is "a schedule of reinforcement in which a reinforcer is presented *only* if a particular response occurs at a low rate" (Martin & Pear, 2015, p.114). To carry out a DRL procedure, a behaviour analyst in conjunction with an individual and/or their family will set a criterion that describes a low rate of noncompliant behaviour, and once that criterion is set, reinforcement is only provided when noncompliant behaviour does not surpass the criterion. Limited-responding DRL involves specifying an interval of time, for example, 30 min, and setting a limit of how many responses (noncompliant ones) will be acceptable during the time period.

DRO is "a schedule in which a reinforcer is presented *only* if a specified response does *not* occur during a specified period of time" (Martin & Pear, 2015, p. 115). Specific time intervals are set where the undesirable behaviour (e.g. noncompliance) cannot occur. If an instance of noncompliance occurs during that interval, then the specified time interval will restart. Reinforcement is only provided when no instances of noncompliance occur during the specified period of time (i.e. compliance is observed for the entire time interval). As reinforcement increases (i.e. the individual does not perform the undesirable behaviour), the time interval can be increased. This method may be particularly useful for instances where noncompliance coincides with problem behaviours, such as self-injurious behaviour, where instances of self-injury can be easily measured, and where a decrease in self-injury may also produce a decrease in noncompliance.

DRA "is a procedure that involves the extinction of a problem behaviour combined with reinforcing a behaviour that is topographically dissimilar to, but not necessarily incompatible with, the problem behaviour" (Martin & Pear, 2015, p.116). DRA is applicable when a problem behaviour occurs in conjunction with noncompliance, and compliance can be reinforced while the problem behaviour is ignored.

An important consideration when implementing interventions is to ensure that reinforcement in any form is only provided as a consequence to desired behaviours. Conducting a functional assessment to determine the contingencies maintaining noncompliance as well as preference assessments to determine effective reinforcers will provide important information when designing interventions targeting compliance (Lipschultz & Wilder, 2017).

Donohue, Casey, Bicard, and Bicard (2012) examined the effects of differential reinforcement on noncompliance in a 17-year-old male with autism spectrum disorder. The study used differential reinforcement of short latencies in a changing criterion design. Response latency was measured as the number of seconds between the end of the instruction and the response by the participant. Accuracy was measured as the proportion of requests followed by correct responses.

Baseline sessions required the participant to answer instructions before the timer beeped (10 s) in order to receive reinforcement. The intervention phase was similar to baseline with the exception that differential reinforcement of short latencies was used with praise and a preferred reinforcer. The response criterion decreased by 1 s after the participant responded correctly for three consecutive sessions. Incorrect responses received constructive feedback.

Results indicated that the mean response latency decreased each time that the intervention was implemented or the criterion was reduced. The authors concluded that differential reinforcement of short latencies was effective for decreasing response latency because the participant only responded to requests after long periods of time prior to the intervention. The intervention also increased the proportion of correct responses and decreased the proportion of incorrect and no-response requests.

DRL, DRO, and DRA are all well-supported and frequently used methods of consequence interventions for improving compliance in PWIDD. They can also be used in combination with ratio and interval schedules of reinforcement to develop desirable behaviours. Differential reinforcement has been mentioned in several recent reviews including Lipschultz and Wilder (2017), Luiselli (2009), and Scheithauer, Cariveau, Call, Ormand, and Clark (2016).

Interventions to Remediate Behavioural Excesses

Extinction

Extinction procedures can be used to reduce the frequency of behaviours that are incompatible with compliance. While the procedures may vary in their presentation (e.g. social extinction, escape extinction, etc.), the underlying processes remain the same. Reinforcers that have been maintaining noncompliant behaviour are identified and withheld contingent on noncompliance (Martin & Pear, 2015). The result is a decrease in the probability that the individual will engage in the noncompliant behaviour. This creates a situation where compliance is more likely to occur.

Extinction procedures are rarely used in isolation during compliance training (Lipschultz & Wilder, 2017). The majority of its applications are instead supplementary but are often a necessary addition for PWIDD whose noncompliance is accompanied by other CBs (Piazza et al., 1997; Zarcone, Iwata, Hughes, & Vollmer, 1993; Zarcone, Iwata, Mazaleski, & Smith, 1994). One study found that the use of a high-probability instructional sequence was unable to produce an improvement in compliance or self-injurious behaviour when self-injury was being maintained by an escape from task demands (Zarcone et al., 1993). However, when extinction was included such that self-injury no longer provided a break from task demands, levels of compliance and latency to self-injurious behaviour increased. This was demonstrated even when extinction was implemented alone. Piazza et al. (1997) also found that extinction procedures were required but only for one of three individuals. Other studies have found its addition to be redundant (Carter, 2010; Lalli et al., 1999).

Extinction should be considered as a means to reduce problem behaviour so long as those conducting the procedure are appropriately trained and prepared. For example, an immediate but temporary increase in the undesirable behaviour (i.e. an extinction burst) can occur following the implementation of an extinction procedure (Martin & Pear, 2015). If these consequences pose a risk to the person's or another's safety, then extinction should be removed from the treatment package. The correct application of an extinction procedure also requires that a functional analysis be conducted beforehand; otherwise, the contingency that is maintaining the undesirable behaviour may remain intact.

Non-contingent Reinforcement and Non-contingent Escape

Noncompliance is often associated with other forms of CB, such as aggression or self-injury. The function of these behaviours may be to escape from task demands, thereby resulting in noncompliance. If the behaviours are instead being maintained by positive reinforcement (i.e. access to social attention and tangibles or are automatically reinforcing), then engaging in such behaviours may distract the individual and prevent them from adequately complying with requests. One way to promote compliance then may be to use interventions that target CBs that accompany noncompliance.

A relatively simple intervention used for the purpose of reducing CB and increasing compliance in PWIDD is non-contingent reinforcement (NCR). NCR involves the delivery of reinforcement regardless of the behaviour that has just occurred (Martin & Pear, 2015). This is usually done according to a fixed or variable time schedule, where the reinforcer is consistently presented to the individual following a specified amount of time. It is believed that the non-contingent presentation of a functionally relevant reinforcer decreases an individual's motivation to engage in CBs that were being maintained by the same reinforcer. In circumstances where a brief break from task demands or requests is delivered non-contingently, the terminology non-contingent escape (NCE) has been used (Kodak, Miltenberger, & Romaniuk, 2003).

Kodak et al. (2003) demonstrated the effectiveness of NCE for two children with developmental disabilities. Prior assessments had indicated that CB such as aggression was being maintained by escape from task demands. It was found that providing a brief break from instructional task demands every 10 s reduced CB and increased compliance relative to baseline for both individuals. Treatment gains were also maintained when the interval between breaks was increased gradually from 10 s to 2 min. Furthermore, an alternating treatment design allowed researchers to conclude that NCE was equally effective in reducing CB and increasing compliance compared to an alternative intervention (i.e. differential negative reinforcement of other behaviours).

Similar results have also been reported by Ingvarsson, Kahng, and Hausman (2008), who found that providing NCR in the form of preferred edible items decreased CB that was being maintained by access to tangible items and escape from task demands. Considerable increases in compliance were also observed using both dense (i.e. an edible every trial) and lean (i.e. an edible once every fourth trial) schedules of NCR. Again, positive behaviour change was achieved even though a 30 s break from task demands remained contingent on CB. A clear demonstration of the utility of NCR was shown when treatment gains were reversed and subsequently regained during a return to baseline followed by reinstatement of NCR, respectively.

Although there has been limited research documenting the use of NCR methods for increasing compliance specifically, results from the aforementioned studies indicate that NCE and NCR can be an effective form of compliance training in some situations. Its widespread success in treating CB often associated with noncompliance in PWIDD further highlights its potential as a compliance training strategy (Carr, Severtson, & Lepper, 2009). For caregivers with no previous experience, correct implementation of NCR methods can be achieved following a single, 1-hour training session (Noel & Getch, 2016). Unfortunately, NCE and NCR do not teach an individual what the appropriate behaviour is (i.e.

compliance), and so it is best used in combination with other strategies. Accordingly, both Kodak et al. and Ingvarsson et al. incorporated a least-to-most prompting strategy during baseline and treatment sessions. This ensured that the individuals received the assistance necessary to complete the instructional tasks. In order for NCR interventions to be most effective, functionally relevant reinforcers should be utilized, and NCR or NCE should be delivered at a rate equal to the rate of CB during baseline (Lalli, Casey, & Kates, 1997). Once acceptable rates of compliance have been established, gradual reductions in the frequency of NCR or NCE delivery can be made.

Punishment

Punishment interventions may be considered for some individuals whose noncompliance cannot be managed effectively with non-aversive procedures and when noncompliance is accompanied with self-injury or aggression towards others. Punishment should only be used under the guidance and supervision of a Board-Certified Behaviour Analyst or licensed mental health professional and then only as a last resort. When a behaviour analyst uses punishment procedures, special attention must be paid to the ethical code 4.08, which discusses guidelines for using punishment as a behavioural intervention and how it should be a last resort option (Bailey & Burch, 2016, p. 132–135). Failing to abide by the appropriate ethical codes when using punishment can result in suspension or revocation of certification. Additionally, strict policies with regard to the use of punishment procedures should exist at the organization level. Punishment must be used cautiously and with safeguards in place to ensure that appropriate training is provided to individuals implementing interventions to prevent misapplication of the procedure and to ensure the safety of everyone involved.

Timeout. According to Martin and Pear (2015), a timeout is “a period of time immediately following a particular behaviour during which an individual loses the opportunity to earn reinforcers” (p. 122). Timeouts may be exclusionary or non-exclusionary (Martin & Pear, 2015). An exclusionary timeout involves removal of a client from the reinforcing situation and relocation to a different room or area immediately following an instance of the target behaviour, (Martin & Pear, 2015). Non-exclusionary timeouts involve adding a stimulus to the situation following an instance of noncompliance that is associated with less reinforcement (Martin & Pear, 2015), for example, having a child sit on a chair during playtime after an instance of noncompliance or removing a reinforcing toy when noncompliant behaviours are observed. Generally, exclusionary timeouts are more commonly used by professionals and are often used to supplement reinforcement procedures (Lipschultz & Wilder, 2017).

To implement an exclusionary timeout, the target behaviour must be operationally defined, and an appropriate timeout duration and location must be determined. For example, the behaviour may be noncompliant in the form of a child’s tantrum when he is asked to clean up his toys. A parent might escort the child to a chair in the home office where they must remain for 1 min. After the 1 min duration is complete, the child is allowed to resume what they were doing prior to the timeout.

In a study by Handen, Parrish, McClung, Kerwin, and Evans (1992), a timeout intervention was compared to guided compliance as a method of decreasing noncompliance in five children with developmental disabilities, three of whom had a mild intellectual disability. An ABAC experimental design was used, and the order of treatment conditions was counterbalanced across participants. During baseline sessions, compliance was praised, and noncompliance was ignored. During guided compliance sessions, the participant had to respond to requests within 10 s. Praise and uninterrupted play were provided for compliant responses, whereas noncompliance resulted in the adult administering the requests to guide the child to complete the task by placing their hand over the participants hand and guiding the desired response. Once the task was completed, the child was allowed to continue playing.

During timeout sessions compliance was treated the same as in the guided compliance sessions, and noncompliance resulted in a 30 s timeout in a chair facing a corner in the training room. After the 30 s timeout, participants were allowed to return to play. Handen et al. found that mean compliance during baseline was 41% which increased to 85% and 59% during timeout and guided compliance training sessions, respectively. Further examination of the intra-subject data demonstrated that timeouts were more effective at increasing compliance for four of five participants; however, both interventions increased compliance compared to baseline scores.

Determining the function of noncompliance is important when implementing timeouts because if the function is escape from demands, timeouts actually reinforce noncompliance by removing them from the situation and demand. Timeouts are more appropriate when noncompliance is a function of attention since timeouts away from others remove opportunities for reinforcement in the form of attention. Furthermore, if the function of noncompliance is attention, care must be taken to ensure that not too much time is spent putting clients in timeout as the interaction provided by placing the client in timeout may lead to the timeout being reinforcing.

Conclusion

Noncompliance is a prevalent and important problem among PWIDD. It is best understood as both a behavioural deficit and a behavioural excess. Defining it this way (a) clarifies how basic behavioural principles such as reinforcement and stimulus control can be applied to increase compliance and (b) may help caregivers to choose appropriate and effective interventions. Fortunately, a variety of such interventions have been reported in the research literature; many have considerable support.

In general, caregivers should seek to increase compliance by providing simple, clear, affirmative instructions for behaviours that are in the person's repertoire. Positive reinforcement should immediately follow compliance and should be more likely following compliance than noncompliance. Preference assessments can identify effective reinforcers for interventions designed to increase compliance. Various antecedent strategies may be used to increase the likelihood of compliance with a given instruction; one well-tested method is to provide the difficult instruction in the context of several other, easier-to-follow instructions.

When noncompliance includes CB, effective reinforcement-based strategies to increase compliance may be a sufficient strategy—by definition compliance and noncompliance are incompatible and thus covary. Reinforcement-based strategies such as NCE and differential reinforcement may be indicated but should be guided by a functional assessment. Knowing the function of the behaviour helps to ensure noncompliance is not inadvertently being maintained. Other strategies for reducing noncompliance with CB such as extinction and punishment may be effective but should be applied only by qualified professionals.

References

Adelinis, J. D., & Hagopian, L. P. (1999). The use of symmetrical 'do' and 'don't' requests to interrupt ongoing activities. *Journal of Applied Behavior Analysis, 32*(4), 519–523. https://doi.org/10.1901/jaba.1999.32-519

Bailey, J., & Burch, M. (2016). *Ethics for behavior analysts*. Routledge: Taylor & Francis Group. https://doi.org/10.4324/9781315669212

Belfiore, P. J., Basile, S. P., & Lee, D. L. (2008). Using a high probability command sequence to increase classroom compliance: The role of behavioral momentum. *Journal of Behavioral Education, 17*(2), 160–171. https://doi.org/10.1007/s10864-007-9054-x

Bellipanni, K. D., Tingstrom, D. H., Olmi, D. J., & Roberts, D. S. (2013). The sequential introduction of positive antecedent and consequent components in a compliance training package with elementary students. *Behavior Modification, 37*(6), 768–789. https://doi.org/10.1177/0145445513501959

Benoit, D. A., Edwards, R. P., Olmi, D. J., Wilczynski, S. M., & Mandal, R. L. (2001). Generalization of a positive treatment package for child noncompliance. *Child and Family Behavior Therapy, 23*(2), 19–32. https://doi.org/10.1300/J019v23n02_02

Bialas, J. B., & Boon, R. (2010). Effects of self-monitoring on the classroom preparedness skills of kindergarten students at-risk for developmental disabilities. *Australian Journal of Early Childhood, 35*(4), 40–52.

Carr, E. G., & Durand, V. M. (1985). Reducing behavior problems through functional communication training. *Journal of Applied Behavior Analysis, 18*(2), 111–126. https://doi.org/10.1901/jaba.1985.18-111

Carr, J. E., Severtson, J. M., & Lepper, T. L. (2009). Noncontingent reinforcement is an empirically supported treatment for problem behavior exhibited by individuals with developmental disabilities. *Research in Developmental Disabilities, 30*(1), 44–57. https://doi.org/10.1016/j.ridd.2008.03.002

Carter, S. L. (2010). A comparison of various forms of reinforcement with and without extinction as treatment for escape-maintained problem behavior. *Journal of Applied Behavior Analysis, 43*(3), 543–546. https://doi.org/10.1901/jaba.2010.43-543

Dixon, D. R., Bergstrom, R., Smith, M. N., & Tarbox, J. (2010). A review of research on procedures for teaching safety skills to persons with developmental disabilities. *Research in Developmental Disabilities, 31*(5), 985–994. https://doi.org/10.1016/j.ridd.2010.03.007

Donohue, M. M., Casey, L. B., Bicard, D. F., & Bicard, S. E. (2012). Effects of differential reinforcement of short latencies on response latency, task completion, and accuracy of an adolescent with autism. *Education and Training in Autism and Developmental Disabilities, 47*(1), 97–108.

Ducharme, J. M., & Ng, O. (2012). Errorless academic compliance training: A school-based application for young students with autism. *Behavior Modification, 36*(5), 650–669. https://doi.org/10.1177/0145445511436006

Ducharme, J. M., & Popynick, M. (1993). Errorless compliance to parental requests: Treatment effects and generalization. *Behavior Therapy, 24*(2), 209–226. https://doi.org/10.1016/S0005-7894(05)80264-3

Ducharme, J. M., & Worling, D. E. (1994). Behavioral momentum and stimulus fading in the acquisition and maintenance of child compliance in the home. *Journal of Applied Behavior Analysis, 27*(4), 639–647. https://doi.org/10.1901/jaba.1994.27-639

Emerson, E., Kiernan, C., Alborz, A., Reeves, D., Mason, H., Swarbrick, R., … Hatton, C. (2001). The prevalence of challenging behaviors: A total population study. *Research in Developmental Disabilities, 22*(1), 77–93. https://doi.org/10.1016/S0891-4222(00)00061-5

Ennis, R. P., Lane, K. L., & Oakes, W. P. (2018). Empowering teachers with low-intensity strategies to support instruction: Self-monitoring in an elementary resource classroom. *Preventing School Failure: Alternative Education for Children and Youth, 62*(3), 176–189. https://doi.org/10.1080/1045988X.2017.1408055

Fischetti, A. T., Wilder, D. A., Myers, K., Leon-Enriquez, Y., Sinn, S., & Rodriguez, R. (2012). An evaluation of evidence-based interventions to increase compliance among children with autism. *Journal of Applied Behavior Analysis, 45*(4), 859–863. https://doi.org/10.1901/jaba.2012.45-859

Handen, B. L., Parrish, J. M., McClung, T. J., Kerwin, M. E., & Evans, L. D. (1992). Using guided compliance versus time out to promote child compliance: A preliminary comparative analysis in an analogue context. *Research in Developmental Disabilities, 13*(2), 157–170. https://doi.org/10.1016/0891-4222(92)90022-X

Holden, B., & Gitlesen, J. P. (2006). A total population study of challenging behaviour in the county of Hedmark, Norway: Prevalence, and risk markers. *Research in Developmental Disabilities, 27*(4), 456–465. https://doi.org/10.1016/j.ridd.2005.06.001

Horner, R. D., & Keilitz, I. (1975). Training mentally retarded adolescents to brush their teeth. *Journal of Applied Behavior Analysis, 8*(3), 301–309. https://doi.org/10.1901/jaba.1975.8-301

Humm, S. P., Blampied, N. M., & Liberty, K. A. (2005). Effects of parent-administered, home-based, high-probability request sequences on compliance by children with developmental disabilities. *Child and Family Behavior Therapy, 27*(3), 27–45. https://doi.org/10.1300/J019v27n03_03

Ingvarsson, E. T., Kahng, S., & Hausman, N. L. (2008). Some effects of noncontingent positive reinforcement on multiply controlled problem behavior and compliance in a demand context. *Journal of Applied Behavior Analysis, 41*(3), 435–440. https://doi.org/10.1901/jaba.2008.41-435

Kern, L., Delaney, B. A., Hilt, A., Bailin, D. E., & Elliot, C. (2002). An analysis of physical guidance as reinforcement for noncompliance. *Behavior Modification, 26*(4), 516–536. https://doi.org/10.1177/0145445502026004005

Kodak, T., Miltenberger, R. G., & Romaniuk, C. (2003). The effects of differential negative reinforcement of other behavior and noncontingent escape on compliance. *Journal of Applied Behavior Analysis, 36*(3), 379–382. https://doi.org/10.1901/jaba.2003.36-379

Lalli, J. S., Casey, S. D., & Kates, K. (1997). Noncontingent reinforcement as treatment for severe problem behavior: Some procedural variations. *Journal of Applied Behavior Analysis, 30*(1), 127–137. https://doi.org/10.1901/jaba.1997.30-127

Lalli, J. S., Vollmer, T. R., Progar, P. R., Wright, C., Borrero, J., Daniel, D., … May, W. (1999). Competition between positive and negative reinforcement in the treatment of escape behavior. *Journal of Applied Behavior Analysis, 32*(3), 285–296. https://doi.org/10.1901/jaba.1999.32-285

Lee, D. L., Belfiore, P. J., Scheeler, M. C., Hua, Y., & Smith, R. (2004). Behavioral momentum in academics: Using embedded high-p sequences to increase academic productivity. *Psychology in the Schools, 41*(7), 789–801. https://doi.org/10.1002/pits.20014

Lipschultz, J. L., & Wilder, D. A. (2017). Behavioral assessment and treatment of noncompliance: A review of the literature. *Education and Treatment of Children, 40*(2), 263–297. https://doi.org/10.1353/etc.2017.0012

Lowe, K., Allen, D., Jones, E., Brophy, S., Moore, K., & James, W. (2007). Challenging behaviours: Prevalence and topographies. *Journal of Intellectual Disability Research, 51*(8), 625–636. https://doi.org/10.1111/j.1365-2788.2006.00948.x

Luiselli, J. K. (2009). Aggression and noncompliance. In *Applied behavior analysis for children with autism spectrum disorders* (pp. 175–187). New York, NY: Springer. Retrieved from https://link.springer.com/chapter/10.1007/978-1-4419-0088-3_10

MacDuff, G. S., Krantz, P. J., & McClannahan, L. E. (2001). Prompts and prompt-fading strategies for people with autism. In *Making a difference: Behavioural intervention for autism* (pp. 37–50). Austin, TX: Pro-Ed Publishers. Retrieved from http://psycnet.apa.org.uml.idm.oclc.org/record/2002-01560-004

Mace, F. C., Hock, M. L., Lalli, J. S., West, B. J., Belfiore, P., Pinter, E., & Brown, D. K. (1988). Behavioral momentum in the treatment of noncompliance. *Journal of Applied Behavior Analysis, 21*(2), 123–141. https://doi.org/10.1901/jaba.1988.21-123

Mandal, R. L., Olmi, D. J., Edwards, R. P., Tingstrom, D. H., & Benoit, D. A. (2000). Effective instruction delivery and time-in: Positive procedures for achieving child compliance. *Child and Family Behavior Therapy, 22*(4), 1–12. https://doi.org/10.1300/J019v22n04_01

Martin, G., & Pear, J. J. (2015). Behavior modification: What it is and how to do it (10th ed). New York: Psychology Press. Retrieved from https://www.taylorfrancis.com/books/9781315663340

Matson, J. L., Sipes, M., Horovitz, M., Worley, J. A., Shoemaker, M. E., & Kozlowski, A. M. (2011). Behaviors and corresponding functions addressed via functional assessment. *Research in Developmental Disabilities, 32*(2), 625–629. https://doi.org/10.1016/j.ridd.2010.12.011

Montgomery, R. W., & Ayllon, T. (1993). The effects of 'do,' 'don't' and 'wait' requests training on compliance behavior and stimulus generalization. *Journal of Behavior Therapy and Experimental Psychiatry, 24*(2), 171–180. https://doi.org/10.1016/0005-7916(93)90046-Y

Neef, N. A., Shafer, M. S., Egel, A. L., Cataldo, M. F., & Parrish, J. M. (1983). The class specific effects of compliance training with 'do' and 'don't' requests: Analogue analysis and classroom application. *Journal of Applied Behavior Analysis, 16*(1), 81–99. https://doi.org/10.1901/jaba.1983.16-81

Nevin, J. A., Mandell, C., & Atak, J. R. (1983). The analysis of behavioral momentum. *Journal of the Experimental Analysis of Behavior, 39*(1), 49–59. https://doi.org/10.1901/jeab.1983.39-49

Noel, C. R., & Getch, Y. Q. (2016). Noncontingent reinforcement in after-school settings to decrease classroom disruptive behavior for students with autism spectrum disorder. *Behavior Analysis in Practice, 9*(3), 261–265. https://doi.org/10.1007/s40617-016-0117-0

Piazza, C. C., Fisher, W. W., Hanley, G. P., Remick, M. L., Contrucci, S. A., & Aitken, T. L. (1997). The use of positive and negative reinforcement in the treatment of escape-maintained destructive behavior. *Journal of Applied Behavior Analysis, 30*(2), 279–298.

Radley, K. C., & Dart, E. H. (2016). Antecedent strategies to promote children's and adolescents' compliance with adult requests: A review of the literature. *Clinical Child and Family Psychology Review, 19*(1), 39–54. https://doi.org/10.1007/s10567-015-0197-3

Rojahn, J., Matson, J. L., Lott, D., Esbensen, A. J., & Smalls, Y. (2001). The behavior problems inventory: An instrument for the assessment of self-injury, stereotyped behavior, and aggression/destruction in individuals with developmental disabilities. *Journal of Autism and Developmental Disorders, 31*(6), 577–588. https://doi.org/10.1023/A:1013299028321

Romano, J. P., & Roll, D. (2000). Expanding the utility of behavioral momentum for youth with developmental disabilities. *Behavioral Interventions, 15*(2), 99–111. https://doi.org/10.1002/(SICI)1099-078X(200004/06)15:2<99::AID-BIN48>3.0.CO;2-K

Russo, D. C., Cataldo, M. F., & Cushing, P. J. (1981). Compliance training and behavioral covariation in the treatment of multiple behavior problems. *Journal of Applied Behavior Analysis, 14*(3), 209–222. https://doi.org/10.1901/jaba.1981.14-209

Scheithauer, M., Cariveau, T., Call, N. A., Ormand, H., & Clark, S. (2016). A consecutive case review of token systems used to reduce socially maintained challenging behavior in individuals with intellectual and developmental delays. *International Journal of Developmental Disabilities, 62*(3), 157–166. https://doi.org/10.1080/20473869.2016.1177925

Smith, M. R., & Lerman, D. C. (1999). A preliminary comparison of guided compliance and high-probability instructional sequences as treatment for noncompliance in children with developmental disabilities. *Research in Developmental Disabilities, 20*(3), 183–195. https://doi.org/10.1016/S0891-4222(99)00002-5

Speights Roberts, D., Tingstrom, D. H., Olmi, D. J., & Bellipanni, K. D. (2008). Positive antecedent and consequent components in child compliance training. *Behavior Modification, 32*(1), 21–38. https://doi.org/10.1177/0145445507303838

Walters, K. L., Holborn, S. W., & Ediger, J. (2007). General case programing of compliance to instruction for individuals with developmental disabilities. *Journal of Developmental and Physical Disabilities, 19*(1), 51–64. https://doi.org/10.1007/s10882-006-9031-7

Wilder, D. A., Saulnier, R., Beavers, G., & Zonneveld, K. (2008). Contingent access to preferred items versus a guided compliance procedure to increase compliance among preschoolers. *Education and Treatment of Children, 31*(3), 297–305.

Wright, T., & Wolery, M. (2011). The effects of instructional interventions related to street crossing and individuals with disabilities. *Research in Developmental Disabilities, 32*(5), 1455–1463. https://doi.org/10.1016/j.ridd.2011.03.019

Zangrillo, A. N., Fisher, W. W., Greer, B. D., Owen, T. M., & De Souza, A. A. (2016). Treatment of escape-maintained challenging behavior using chained schedules: An evaluation of the effects of thinning positive plus negative reinforcement during functional communication training. *International Journal of Developmental Disabilities, 62*(3), 147–156. https://doi.org/10.1080/20473869.2016.1176308

Zarcone, J. R., Iwata, B. A., Hughes, C. E., & Vollmer, T. R. (1993). Momentum versus extinction effects in the treatment of self-injurious escape behavior. *Journal of Applied Behavior Analysis, 26*(1), 135–136. https://doi.org/10.1901/jaba.1993.26-135

Zarcone, J. R., Iwata, B. A., Mazaleski, J. L., & Smith, R. G. (1994). Momentum and extinction effects on self-injurious escape behavior and noncompliance. *Journal of Applied Behavior Analysis, 27*(4), 649–658. https://doi.org/10.1901/jaba.1994.27-649

Wadsworth, J. P., Hansen, B. D., & Wills, S. B. (2015). Increasing compliance in students with intellectual disabilities using functional behavioral assessment and self-monitoring. *Remedial and Special Education, 36*(4), 195–207. https://doi.org/10.1177/0741932514554102

Stereotypy and Repetitive Behaviors

31

Jessica Akers, Tonya Davis, Stephanie Gerow,
Abby Hodges, and Regan Weston

A category of inappropriate behaviors in which individuals with intellectual disabilities engage that do not fit into the typical classifications of aggressive, disruptive, or self-injurious behavior exists. These behaviors include stereotypy, ritualistic behavior, and perseverative behavior related to unusual interests. Stereotypic behaviors are defined as repetitive, coordinated, seemingly purposeless, rhythmic behaviors that are most often maintained by automatic reinforcement (Chebli, Martin, & Lanovaz, 2016; Rapp & Vollmer, 2005; Singer, Mink, Gilbert, & Jankovic, 2016). Common topographies of stereotypy fall into the following categories: whole body (e.g., body rocking), head (e.g., head nodding), hand/finger (e.g., hand flapping), locomotion (e.g., jumping), object (e.g., object spinning), and sensory (e.g., covering ears; Lam & Aman, 2007). In addition to motor stereotypy, individuals with intellectual disabilities also engage in vocal stereotypy (e.g., repetitive sounds or words; Lanovaz & Sladeczek, 2012).

Stereotypy is not exclusive to individuals with intellectual disabilities; all individuals engage in some form of stereotypy (e.g., tapping feet, nail-biting, humming). When exhibited by individuals with intellectual disabilities, stereotypy is distinguished by the lack of appropriateness and should be considered within the context of developmental and social norms (Cunningham & Shriebman, 2008; Rapp & Vollmer, 2005). Stereotypic movements are present during the first few years of life in as many as two thirds of typically developing infants and in 20–50% of children overall (Tröster, 1994). Stereotypies causing concern, however, are likely to persist beyond infancy and childhood, occur at high rates, appear atypical compared to stereotypy exhibited by same age peers, and interfere with an individual's daily functioning (American Psychiatric Association, 2013; Didden et al., 2012).

Individuals with intellectual and multiple disabilities are at an increased risk of exhibiting such complex stereotypies (Lancioni, Singh, O'Reilly, & Sigafoos, 2009). Much of the research and disseminated information on stereotypy focuses on individuals with autism spectrum disorder (ASD). This is likely due, in part, to the inclusion of repetitive behavior within the diagnostic criteria for ASD. Further, individuals with ASD tend to display more severe and varied topographies of stereotypy (Bodfish, Symons, Parker, & Lewis, 2000; Matson & Dempsey, 2008). Chebli et al. (2016) conducted a systematic review of studies reporting prevalence of stereotypy in individuals with developmental disabilities and found that the median (Mdn)

J. Akers (✉) · T. Davis · S. Gerow · A. Hodges
R. Weston
Department of Educational Psychology, Baylor University, Waco, TX, USA
e-mail: jessica_akers@baylor.edu;
tonya_davis@baylor.edu;
Stephanie_gerow@baylor.edu;
abby_hodges@baylor.edu; regan_weston@baylor.edu

J. L. Matson (ed.), *Handbook of Intellectual Disabilities*, Autism and Child Psychopathology Series, https://doi.org/10.1007/978-3-030-20843-1_31

prevalence of stereotypy was 61% among individuals with developmental disabilities. They also noted that stereotypy is most common in children with specific diagnoses such as ASD (Mdn = 88%), followed by fragile X syndrome (Mdn = 69%), and Down syndrome (Mdn = 63%). Finally, they reported that hand/finger, locomotion, object, sensory, and vocal stereotypies were the most common forms of stereotypy aggregated across studies (Mdn range = 46–49%).

Stereotypy and repetitive behaviors can range from low frequency and nondisruptive to high frequency and highly disruptive. According to the Repetitive Behavior Scale-Revised (RBS-R), a measure developed to evaluate the severity of repetitive behaviors, severity is measured by the following: (a) frequency of the behavior, (b) the extent to which interruption of the behavior leads to emotional responding, and (c) the extent to which the behavior interferes with ongoing activities (Lam & Aman, 2007). Many researchers assessing and treating stereotypy report severity in terms of missed educational and social opportunities experienced by the individual (Miguel, Clark, Tereshko, & Ahearn, 2009; Saylor, Sidener, Reeve, Fetherston, & Progar, 2012; Taylor, Hoch, & Weissman, 2005). These missed learning opportunities are especially detrimental for individuals with intellectual disabilities as they are already at an academic disadvantage. In addition, stereotypy and repetitive behaviors can be extremely stigmatizing, thus increasing the likelihood of the individual being excluded from social interactions.

Research has identified several risk factors associated with stereotypy, including intellectual functioning, communication abilities, diagnosis, and environmental factors. The degree of intellectual disability is correlated with stereotypy, with individuals with severe or profound intellectual disability being much more likely to engage in stereotypy than individuals with mild or moderate intellectual disability (McClintock, Hall, & Oliver, 2003). The degree of intellectual disability can also predict the topography, frequency, and severity of stereotypy. Individuals with lower intellectual functioning are more likely to engage in motor stereotypies such as hand flapping or body rocking, while individuals with higher intellectual functioning are more likely to engage in nonfunctional rituals (Leekam, Prior, & Uljarevic, 2011; Turner, 1999). Similarly, among individuals with ASD, those with a lower IQ engage in more severe topographies of stereotypy at a higher frequency compared to those with a higher IQ (McTiernan, Leader, Healy, & Mannion, 2011).

Communication ability may also be correlated with higher risk for stereotypy. While a great deal of evidence indicates the inverse correlation between communication ability and challenging behavior in general (e.g., Emerson et al., 2001; Emerson & Bromley, 1995; Jones et al., 2008), the same may be true for stereotypy specifically. In fact, Bowring, Totsika, Hastings, Toogood, and Griffith (2017) found that individuals with intellectual disabilities who were nonverbal or lacked clear speech were at a higher risk of engaging in stereotypy.

Finally, several diagnoses are associated with unique stereotypy risk factors. First, stereotypy occurs at higher rates among individuals with ASD than any other developmental disability, with frequency and intensity of stereotypy correlated with severity of the ASD diagnosis (Bodfish et al., 2000; Chebli et al., 2016; Matson, Dempsey, & Fodstad, 2009). Other diagnoses associated with high prevalence of stereotypy include Cornelia de Lange, Cri du Chat, Angelman syndrome, fragile X syndrome, Rett syndrome, and Smith-Magenis syndrome (Cornish & Pigram, 1996; Downs et al., 2014; Dykens & Smith, 1998; Hagerman & Lampe, 1999; Hyman, Oliver, & Hall, 2002; Summers, Allison, Lynch, & Sandler, 1995). Some diagnoses are associated with specific topographies of stereotypy. For example, a defining feature of Rett syndrome is hand stereotypy often manifested as hands joining, wringing, clasping, or mouthing (Downs et al., 2014; Olson & Rett, 1985). Similarly, fragile X syndrome is also associated with hand stereotypy often manifested as hand flapping (Lachiewicz, Spiridigliozzi, Gullion, Ransford, & Rao, 1994; Symons, Clark, Hatton, Skinner, & Bailey, 2003). Last, Cornelia de Lange syndrome is associated with body rocking among other topographies of stereotypy (Oliver, Arron, Sloneem, & Hall, 2008; Sarimski, 1997).

In addition to specific diagnoses, visual impairments among individuals with intellectual disabilities are associated with a higher risk of stereotypy (Bowring et al., 2017). Specifically, increased vision loss is correlated with increase prevalence of stereotypic behaviors (Bak, 1999). Not only are individuals with visual impairment and intellectual disability at a higher risk for displaying stereotypy, but specific topographies are common among this population, including body rocking and eye rolling (Abang, 1985).

Some research has identified certain environmental risk factors of stereotypic behavior. Specifically, individuals in congregate or paid care (e.g., group homes or assisted living centers) are at a higher risk of exhibiting stereotypic behaviors (Bowring et al., 2017). Other studies have produced similar findings of an association of those who live in congregate care with higher rates of challenging behavior, which often manifests in the form of stereotypic behaviors (Holden & Gitlesen, 2006; Jones et al., 2008; Lowe et al., 2007).

When individuals experience a lack of daytime activities, they are more likely to engage in stereotypic behaviors (Bowring et al., 2017). Similarly, Schlaggar and Mink (2003) suggest that individuals are more likely to engage in stereotypic behaviors during times of boredom. As a result, it may be beneficial to increase levels of daytime engagement for individuals who are at risk for exhibiting stereotypic behaviors (Bowring et al., 2017; Lowe et al., 2007). Other environmental arrangements that have been associated with increased stereotypy are instances in which high social or emotional demands are in place (LeMonda, Holtzer, & Goldman, 2012; Schlaggar & Mink, 2003). For example, when an individual experiences times of excitement, he or she may be more likely to exhibit stereotypic behaviors (Schlaggar & Mink, 2003).

Assessment of Stereotypy

To efficiently and effectively reduce stereotypy, a functional assessment is required to determine the variables maintaining stereotypy. Typical pre-intervention assessments of stereotypy include interviews, observations, functional analyses, and competing items assessments.

Interviews and Observations

The purpose of initial interviews is to gather information from key stakeholders such as caregivers, teachers, other professionals, and the individual who engages in stereotypy. Interviews typically consist of questions related to (a) the topography of the behavior; (b) the severity of the behavior; (c) key stakeholders' preferences, resources, and priorities related to possible treatment options; (d) the individual's current strengths and skills, (e) typical situations in which the behavior occurs; and (f) common reactions to the behavior.

This information can be gathered using a combination of open-ended interviews and formal questionnaires. Interviews will cover topics such as typical situations (i.e., antecedents) associated with stereotypy and typical reactions of other people to the stereotypy (i.e., consequences). Questions from an open-ended interview provide the interviewer the opportunity to build rapport through conversation and obtain general information related to the target behavior. Formal questionnaires (e.g., Functional Analysis Screening Tool; Iwata, DeLeon, & Roscoe, 2013) provide the interviewer with a guide for obtaining specific information related to possible functions of the target behavior. The information obtained from the interviews aides in the development of an initial hypothesis regarding the function of stereotypy and the identification of antecedents and consequences to include in subsequent assessments. Unstructured observations can further assist in the identification of the topography of stereotypy, the refinement of an operational definition, and the selection of antecedents and consequences for subsequent assessments.

As previously discussed, the severity of stereotypy can be determined based on the extent to which the behavior negatively impacts the individual's day-to-day functioning or long-term goals. Prior to introducing a formal intervention for stereotypy, the severity of the behavior must be assessed to determine whether the behavior warrants treatment. For example, an individual with intellectual disability may engage in body rocking during unstructured play but not during instructional activities; thus, the behavior does not impede the individual's educational goals. If the individual's social interactions are not negatively impacted, it may be determined that although the individual engages in atypical behavior, the severity does not warrant formal treatment. If after gathering information via interviews and observations the severity is determined to be at a level which warrants intervention, a functional analysis should be conducted.

Functional Analysis

The purpose of a functional analysis is to identify the function of a behavior by systematically evaluating the effect of antecedents and consequences on the occurrences of stereotypy. Typically, the results of the functional analysis suggest a social function or an automatic function. Social functions include sources of reinforcement from the environment (e.g., attention, escape from demands; Cooper, Heron, & Heward, 2007). Automatically maintained behavior is behavior that results in reinforcement independent of changes in the environment.

Traditional functional analyses consist of conditions to assess positive reinforcement (e.g., tangible, attention), negative reinforcement (e.g., escape), and automatic reinforcement (e.g., alone, ignore) as possible functions of the target behavior (Hanley, Iwata, & McCord, 2003; Iwata, Dorsey, Slifer, Bauman, & Richman, 1982/1994). In addition, a control condition, often the play condition, is included in the functional analysis to serve as a comparison condition for the remaining test conditions.

The number of conditions and sessions per condition and the length of sessions have varied widely in the literature. Functional analysis sessions typically last 5–15 min (Hanley et al., 2003). The length of the session should depend on the rate of the behavior, the severity of the behavior, and the social validity of the suggested session length (e.g., presenting pre-academic demands to a 3-year-old for an extended period of time may be inappropriate). Traditional functional analyses often include three to five conditions, with five or more sessions per condition (Hanley et al., 2003; Iwata et al., 1982/1994). However, a number of variations including conducting one test and one control condition (i.e., a pairwise functional analysis; Iwata, Duncan, Zarcone, Lerman, & Shore, 1994) and decreasing the number of sessions (i.e., a brief functional analysis; Northup et al., 1991) have been used in the extant literature.

An additional modification to the functional analysis can be employed in cases in which the interviews and/or observations suggest that the behavior is maintained by automatic reinforcement. The extended alone or ignore assessment consists of consecutive alone or ignore conditions, with no other functional analysis conditions (Querim et al., 2013). This assessment can be a more efficient method to identify the function of the individual's stereotypy, in cases in which the stereotypy is maintained by automatic reinforcement. However, it is important to only use this assessment in circumstances in which previous data suggest automatic reinforcement because this assessment will not lead to the identification of social positive or negative reinforcement as primary or secondary functions of stereotypy.

Interpreting functional analysis outcomes. Functional analysis results are typically displayed in a line graph. To identify the function of stereotypy, the occurrences of stereotypy during each test condition is compared to those during the control condition (e.g., play). The functional analysis results suggest a socially maintained function (e.g., access to attention, escape from demands) if stereotypy occurs more frequently or at a higher rate during one or more test condition as compared to the control condition. The results indicate that the behavior is not sensitive to social contingencies (i.e., automatically maintained behavior) if the individual engages in stereotypy at similar rates across all conditions or if the individual engages in more stereotypy during the alone or ignore condition as compared to the remaining conditions (Hagopian, Rooker, & Zarcone, 2015; Iwata et al., 1982/1994). For the extended alone or ignore assessment, the data suggest an automatic function if the child continues to engage in the behavior throughout the sessions or if the rate of the behavior increases across sessions.

Competing Items Assessment

For individuals who engage in automatically maintained stereotypy, identifying activities or items associated with lower rates of stereotypy can lead to the development of a more effective intervention. Previous research has described the use of a competing items assessment to identify items associated with lower rates of stereotypy (e.g., Groskreutz, Groskreutz, & Higbee, 2011; Nuernberger, Vargo, & Ringdahl, 2013). In a competing-item assessment, the interventionist presents an item or activity during each session and records the duration of stereotypy and item engagement. The items or activities associated with lower rates of stereotypy and higher item engagement can be used during the intervention to reduce stereotypy.

Treatment for Stereotypy

Following the identification of the function of stereotypy, an individualized intervention must be developed. If stereotypy is maintained by social reinforcement, the intervention strategies will follow the same procedures as with other topographies of challenging behavior. If the behavior is not socially maintained, as is the case with most stereotypic behaviors, a different set of procedures will be necessary. In the literature, effective interventions for automatically maintained stereotypy can be divided into three categories: (a) antecedent manipulations, (b) reinforcement-based interventions, and (c) punishment procedures. The most common and well-researched interventions within these categories will be described in detail, including considerations for implementation.

Antecedent Manipulations

Automatically maintained behavior poses a unique challenge for implementing reinforcement-based interventions because the functional reinforcer cannot be manipulated by the interventionist. Due to this difficulty, several interventions targeting the reduction of stereotypy incorporate antecedent manipulations. These manipulations include bringing stereotypy under stimulus control, environmental enrichment, and manipulating motivating operations.

Establishing stimulus control. Stimulus control is demonstrated when a behavior occurs in the presence of one stimulus and does not occur in the presence of another, for example, driving through an intersection when the light is green and not when it is red. Stimuli come to exert control based on a history with specific consequences (Cooper et al., 2007). Interventions which incorporate stimulus control procedures introduce different stimuli (e.g., colored cards) and pair the stimuli with different contingencies. For example, the presence of a green poster emitting a specific behavior will result in reinforcement, whereas in the presence of a red poster, the same behavior will contact extinction. Such arrangements are commonly implemented in conjunction with functional communication training as a means to decrease excessive requesting without observing an increase in challenging behavior due to mands contacting extinction (Saini, Miller, & Fisher, 2016). Discriminated responding is achieved when the individual requests the functional reinforcer in the presence of the stimulus associated with reinforcement and refrains from requesting in the presence of the stimulus associated with extinction.

In regard to establishing stimulus control to reduce stereotypy, the interventionist uses stimuli to signal times in which engaging in stereotypy is appropriate (i.e., will not be interrupted) and inappropriate (i.e., will be interrupted and/or will contact specific consequences). For example, in the presence of the green card, stereotypy will not be blocked, and in the presence of the red card, stereotypy will be blocked.

In contrast to destructive behavior (e.g., aggression, self-injury), it is not imperative for stereotypy to be reduced to zero levels. Some level of stereotypy can be accepted as long as it does not negatively disrupt the individual's day-to-day functioning. Most individuals have certain behaviors which they emit only within the privacy of their own home. Thus, it seems reasonable to provide individuals with intellectual disabilities the same courtesy in the case of stereotypy. In addition, the sensory stimulation accessed by engaging in the stereotypic behavior likely serves as a potent reinforcer for the individual. Rather than denying an individual access to this source of reinforcement, the goal of establishing stimulus control is to reduce the duration of engagement in stereotypy and/or number of settings in which the individual engages in stereotypy. Previous researchers have suggested that this is not only more practical but also more ethical as individuals with intellectual disabilities may have a limited number of available reinforcers (Tiger, Wierzba, Fisher, & Benitez, 2017).

Researchers have evaluated the use of punishment in conjunction with the establishment of stimulus control (e.g., Anderson, Doughty, Doughty, Williams, & Saunders, 2010; Martinez, Betz, Liddon, & Werle, 2016), such that in the presence of the stimulus which signals stereotypy is inappropriate, stereotypy results in a punishment procedure (e.g., hands-down procedure). Punishment rather than extinction may be necessary in these cases because the reinforcing properties of stereotypy cannot be manipulated by the interventionist.

After discriminated responding is achieved (i.e., stereotypy decreases in the presence of the stimulus signaling punishment), the number of implementations of the punishment procedure will decrease, and there is some evidence to suggest that the actual punishment contingency may no longer be necessary in order to maintain suppression of stereotypy. Tiger et al. (2017) demonstrated that after introducing response blocking in the presence of a stimulus for a given amount of time, the presence of the stimulus resulted in suppression of ritualistic behavior even when the punishment contingency was no longer in place. Due to ethical considerations regarding the use of punishment, establishing stimulus control may be an ideal option for including punishment within an intervention without excessive implementation of the punishment procedure.

Bringing stereotypy under stimulus control may also be ideal because it does not require implementation of the intervention 100% of the time. This may be desirable due to the labor-intensive nature of many of the interventions for stereotypy. Once stimulus control is established, interventionists have the option to present the stimulus signaling stereotypy is appropriate in situations in which they are unable to implement the punishment procedure (e.g., caregiver attending to another child) without risking a deterioration of intervention effectiveness. In addition, there is some evidence to suggest that introducing a punishment procedure within a stimulus control arrangement may reduce some of the undesirable side effects of punishment, including the interventionist becoming an aversive stimulus and nonselective response suppression (Doughty, Anderson, Doughty, Williams, & Saunders, 2007).

Prior to introducing stimulus control procedures, the interventionist should consider the type of stimuli to include within the intervention. There is some evidence to suggest including therapist-arranged stimuli (e.g., colored cards, wristbands) may be more effective for the acquisition of discriminated responding as compared to naturally occurring stimuli (Shamlian et al., 2016). However, the selection of stimuli should be thoughtfully considered to ensure the achievement of discriminated responding while avoiding overly stigmatizing stimuli. One reason for decreasing stereotypy is to decrease stigmatization; therefore, the stimuli included should not be unduly stigmatizing.

Environmental enrichment. Environmental enrichment is an antecedent manipulation that has been used to reduce stereotypy by introducing competing sources of noncontingent reinforcement into an individual's environment (Rapp & Vollmer, 2005). The stimuli are considered noncontingent because they remain freely accessible to the individual regardless of his or her engagement in specific responses. Research has shown that increased levels of sensory stimulation can lead to reductions in stereotypic behavior due to heightened levels of stimulation in the immediate environment (Hill, Trusler, Furniss, & Lancioni, 2012; Sidener, Carr, & Firth, 2005). Variations exist regarding the characteristics of stimuli introduced to establish an enriched environment including preferred stimuli (Higbee, Chang, & Endicott, 2005), matched stimuli (e.g., Rapp, Cook, McHugh, & Mann, 2017), and competing stimuli (e.g., Groskreutz et al., 2011). Matched stimuli are those that provide similar sensory stimulation as the stimulation hypothesized to maintain the stereotypy (e.g., music for vocal stereotypy; Ahearn, Clark, MacDonald, & Chung, 2007) and competing stimuli are items associated with lower rates of stereotypy identified using a competing items assessment.

Previous research evaluating the effects of an enriched environment using preferred and nonpreferred stimuli indicates that using a highly preferred stimulus alone does not always result in subsequent reductions of stereotypic behaviors (Higbee et al., 2005; Lanovaz, Sladeczek, & Rapp, 2011). Conversely, researchers have evaluated the effects of matched stimulation on the occurrence of stereotypic behaviors and found that introducing noncontingent access to highly preferred stimuli that produce consequences similar to the hypothesized maintaining consequences of stereotypic behaviors may result in decreases of the targeted behavior (Higbee et al., 2005; Love, Miguel, Fernand, & LaBrie, 2012; Rapp, 2006; Rapp et al., 2017; Saylor et al., 2012). Although identifying highly preferred stimuli that match stimulation produced by stereotypy has been shown to reduce targeted topographies of stereotypy, there are mixed results of the long-term effects of highly preferred matched stimuli on nontargeted topographies of stereotypy (Rapp et al., 2017). Finally, introducing preferred items that serve as competing stimuli for the occurrence of stereotypy has also been shown to be a promising strategy for the reduction of stereotypy in some instances (Groskreutz et al., 2011). Interventions using enriched environments often include multiple intervention components such as enriched environment in combination with response cost (Watkins, Paananen, Rudrud, & Rapp, 2011; Watkins & Rapp, 2014), presession free access to stereotypy (Lang et al., 2009; Lang et al., 2010; Rispoli et al., 2014), or response interruption and redirection (RIRD; Love et al., 2012).

Factors to consider prior to using environmental enrichment strategies include (a) selection of methods for identifying preferred, matched, or competing stimuli, (b) documentation of subsequent reductions in targeted and nontargeted stereotypic behaviors as well as increases in appropriate behaviors, (c) the extent to which prompting is required to promote interaction with stimuli in the environment, and (d) the extent to which additional reinforcement is necessary to increase appropriate behaviors (Rapp & Vollmer, 2005). This type of antecedent manipulation may be ideal as it is relatively simple to implement because there is no need to closely observe behavior due to the noncontingent nature of the intervention. Thus, the extent to which additional resources (e.g., training and man power) are necessary is greatly reduced. Another benefit of environmental enrichment is related to challenging behavior associated with blocking stereotypic behavior. Because response blocking is not a component of environmental enrichment, an increase in other topographies of challenging behavior related to blocked stereotypy is unlikely. Despite the effectiveness of this antecedent manipulation, there is the possibility that the presence of the stimuli (preferred, matched, or competing) may in some cases be more disruptive than the actual stereotypy (Watkins et al., 2011). In addition, there is little evidence regarding the extent to which these stimuli remain effective in reducing stereotypy over time.

Manipulating motivating operations. In situations in which noncontingent access to competing stimuli is impractical (e.g., classroom instruction), it may be beneficial to abolish decrease the individual's motivation to engage in stereotypy. An abolishing operation refers to any event that reduces the value of a specific reinforcer and thereby leads to a decrease in behavior associated with the reinforcer (Laraway, Snycerski, Michael, & Poling, 2003). For example, after eating an entire bag of chips, the value of chips is reduced, and thus, behaviors associated with obtaining chips decrease. In the case of stereotypy, prior to the target activity (e.g., math lesson), the individual is provided with noncontingent access to matched and/or unmatched stimuli (Rapp, 2006), or the individual is able to freely engage in stereotypy (Lang et al., 2010) for a specified duration. Researchers have found this method to be effective for decreasing stereotypy during subsequent research sessions for many individuals. In the cases which it has not been shown to be effective, it has been suggested that perhaps the amount of time allotted for presession exposure was not sufficient to abate subsequent stereotypy. Therefore, researchers have suggested strategies to identify behavioral indicators which suggest that the value of sensory reinforcement has adequately reduced such that stereotypy will decrease (Lang et al., 2010; Rispoli et al., 2014). For example, Rispoli and colleagues used three rejections of the object associated with stereotypy as a criterion for determining the effectiveness of the presession exposure.

There are several benefits to manipulating motivating operations as a strategy to decrease stereotypy. The first two align with benefits of environmental enrichment. First, the intervention is relatively simple to implement and thus would not require extensive staff training (Rispoli et al., 2014). Second, the exclusion of response blocking within the procedures will reduce engagement in challenging behavior related to stereotypy being blocked. Third, it has been suggested that decreasing the value of sensory stimulation may lead to a subsequent increase in the value of other more socially appropriate reinforcers such as social interactions. Finally, because the items are not present during the "intervention period," the distractions related to noncontingent access to the items are eliminated.

Despite the benefits of this antecedent manipulation, it is important to consider the possibility of extensive time being required to achieve presession sensory habituation. In the Rispoli study, one participant required 26 min of presession access before displaying the behavioral indicators associated with habituation. It is also unclear how long the effects of manipulating the motivating operations will remain effective due to relatively short session lengths (e.g., 10 min) reported within the research.

Reinforcement-Based Interventions

The extant literature provides ample evidence that reinforcement-based interventions can be used to increase socially appropriate behavior and decrease stereotypy (e.g., Gehrman, Wilder, Forton, & Albert, 2017; Greenberg, Lau, & Lau, 2016; Lancioni et al., 2008). Previous reviews of the literature indicate that research supports the use of reinforcement-based interventions to reduce inappropriate behavior in individuals with intellectual disability (Brosnan & Healy, 2011; Chowdhury & Benson, 2011). Furthermore, professional organizations and leaders in the field recommend the use of reinforcement-based interventions for the reduction of inappropriate behavior (BACB, 2014; Horner et al., 2005).

When developing reinforcement-based interventions for stereotypy maintained by access to automatic reinforcement, highly preferred items or activities can be introduced as reinforcers. Alternatively, the reinforcer can be an item or activity that is associated with low rates of stereotypy, based on the findings from a competing items assessment. The use of competing items in the previous section referred to noncontingent access to these stimuli, whereas within reinforcement-based interventions, these items are provided contingent on the individual engaging in specific responses or refraining from engaging in stereotypy. Each of the interventions described below includes the use of reinforcement to decrease stereotypy but differ based on the behaviors in which the individual must engage to access reinforcement.

Differential reinforcement of alternative behaviors (DRA). DRA involves providing reinforcement contingent on the occurrence of an alternative, appropriate behavior (Cooper et al., 2007; Tiger, Hanley, & Bruzek, 2008). Although

DRA is one of the most common function-based interventions for socially maintained challenging behavior, researchers have also evaluated the efficacy of DRA to reduce automatically maintained stereotypy (e.g., Bennett, Reichow, & Wolery, 2011; Lancioni et al., 2008). Prior to implementing DRA, an alternative behavior must be identified, the source of reinforcement must be selected, and the schedule of reinforcement for the alternative behavior (e.g., fixed ratio 1, fixed ratio 5, variable ratio 5) must be established. It is also important to include teaching strategies for the alterative behavior (e.g., modeling), especially in cases in which the alternative behavior is not part of the individual's current skill repertoire. To implement the intervention, reinforcement should be provided contingent on the occurrence of the alternative behavior.

Differential reinforcement of incompatible behaviors (DRI). DRI is a variation of DRA, in which reinforcement is provided contingent on a behavior that is incompatible with the topography of stereotypy (Cooper et al., 2007). To implement DRI, a behavior that cannot occur simultaneously with the stereotypy (i.e., the incompatible behavior) must be identified, the source of reinforcement must be selected, and the schedule of reinforcement for the incompatible behavior must be established. During the intervention, the interventionist provides reinforcement contingent on the individual engaging in the incompatible behavior. The same considerations related to teaching strategies discussed in the DRA section are also applicable for DRI.

One of the relative strengths of DRI interventions is that they involve teaching the individual a response that cannot occur at the same time as the stereotypy. Thus, as the frequency of the incompatible behavior increases, the frequency of stereotypy decreases. For example, Colón, Ahearn, Clark, and Masalsky (2012) demonstrated the efficacy of teaching appropriate verbal behavior to decrease vocal stereotypy for three children with ASD. This intervention, along with RIRD, resulted in decreases in vocal stereotypy. As was the case in this study, DRI is recommended when the interventionist is able to identify a feasible and appropriate incompatible behavior based on the topography of the stereotypy.

Differential reinforcement of other behaviors (DRO). Several studies have evaluated the use of DRO to reduce stereotypy (e.g., Gehrman et al., 2017; Nuernberger et al., 2013). DRO involves providing reinforcement contingent on the absence of stereotypy (Cooper et al., 2007). Prior to implementation, the length of the interval (i.e., length of time the individual is required refrain from engaging in stereotypy) and the source of reinforcement are selected. Often, the interval length is based on the baseline rate of stereotypy (e.g., Gehrman et al., 2017; Nuernberger et al., 2013). Reinforcers may be identified with a preference assessment or competing items assessment. The intervention consists of providing reinforcement if the individual does not engage in stereotypy for the predetermined length of time.

DRO may be especially well-suited to treat stereotypy as it does not require the identification of appropriate incompatible or alternative behaviors. It may be difficult to identify appropriate incompatible behaviors that the individual can engage in for extended periods of time. As an example, placing hands in lap would function as an incompatible response for an individual who engages in repetitive hand flapping; however, keeping hands in lap may impede academic engagement. In this case, it may be more practical to reinforce the absence of hand flapping than to reinforce keeping hands in lap. Similarly, it can be difficult to identify and teach an alternative behavior to replace automatically maintained stereotypy. In the majority of DRA interventions, the appropriate alternative behavior persists because the behavior results in access to the same reinforcer that the inappropriate behavior produced. However, in the case of automatically maintained stereotypy, it may be difficult to identify an appropriate alternative behavior that results in the same form of reinforcement produced by the stereotypy. If a highly preferred item or activity is designated as the reinforcer (i.e., not function-based), the alternative behavior may not occur at high rates because it is not functionally related to the stereotypy. For these reasons, it may be more feasible to reinforce the absence of stereotypy than to reinforce particular behaviors that will replace stereotypy.

When deciding whether to implement environmental enrichment, in which reinforcers are freely available, or a DRO procedure, the interventionist must consider the benefits and disadvantages of each. The benefit of introducing reinforcers noncontingently, as is the case with environmental enrichment, is the response effort for the interventionist is extremely low; however, because the reinforcer is freely available, there is a chance that stereotypy could be adventitiously reinforced. Within a DRO procedure, adventitious reinforcement would be eliminated because the reinforcer is only delivered in the absence of stereotypy. While this is ideal, implementing DRO also requires additional resources as constant observation is required to determine whether the individual engaged in stereotypy during the interval.

Punishment Procedures

By definition, punishment is the immediate presentation or removal of a stimulus after the occurrence of a behavior that reduces the future frequency of that behavior (Azrin & Holtz, 1966). Punishment procedures remain a controversial topic among behavior analysts, with controversy existing regarding both ethics and efficacy of punishment procedures (DiGennaro Reed & Lovett, 2008). From an ethical perspective, behavior analysts must adhere to the responsibilities associated with all human service providers: to do no harm, provide clients with the least intrusive procedures, and provide effective treatment (Cooper et al., 2007). Unfortunately, adhering to these responsibilities does not always provide a clear decision-making model regarding the use of punishment. In some cases, procedures involving punishment may be less intrusive and/or more effective than procedures without punishment; therefore, in some cases use of punishment may be ethical (DiGennaro Reed & Lovett, 2008; Foxx, 2005). The Association for Behavior Analysis established a task force on the right to effective treatment. In this seminal work, Van Houten et al. (1988) point out that specific behavior change procedures are not inherently *good* or *bad*, but instead treatment selection should be based on a myriad of information including risks the behavior poses, controlling variables, treatment effectiveness, and treatment risks. In the case of stereotypy, in which the controlling variables are often unknown or cannot be controlled by the interventionist, punishment procedures may be necessary for effective treatment. Review of the literature indicates in the cases in which punishment has been implemented to treat stereotypy, the punishment procedures are considered relatively less intrusive in that they are combined with reinforcement-based procedures and do not involve delivery of physical pain. With interventionists called to select effective and ethical treatment, it stands to reason that continued empirical investigations of the effectiveness punishment procedures are warranted (DiGennaro Reed & Lovett, 2008; Lerman & Vorndran, 2002).

A variety of punishment procedures to reduce stereotypy have been evaluated across the literature. The most common punishment procedures include RIRD, response blocking, hands-down, verbal interruption and reprimand, and response cost.

Response interruption and redirection. RIRD involves interrupting the targeted stereotypy and redirecting the individual to emit several socially acceptable responses. RIRD was introduced by Ahearn et al. (2007) as a procedural variation to response blocking because it is physically impossible to block vocal stereotypy. In this initial evaluation of RIRD, contingent on stereotypy, the interventionist prompted attendance and delivered prompts for appropriate vocalizations until the participant emitted three consecutive correct responses in the absence of vocal stereotypy.

RIRD is the most frequently evaluated punishment procedure to treat stereotypic behavior, with the vast majority of published studies reporting successful reductions of stereotypy. While several studies have indicated the success of RIRD to reduce motor stereotypy (e.g., Ahrens, Lerman, Kodak, Worsdell, & Keegan, 2011), the majority of literature has evaluated the effectiveness of RIRD on vocal stereotypy. This may be a result of relatively few punishment procedures (e.g., response blocking) being compatible to treat vocal stereotypy.

Several procedural variations have been evaluated across the literature. One such variation is the number of redirection demands delivered. The most common approach involves the interventionist prompting the participant until he or she emits three consecutive correct responses; however, at least one study found RIRD to be effective with the completion of only one redirection demand (Saini, Gregory, Uran, & Fantetti, 2015). This variation was not only effective but also resulted in shorter implementation time. Another procedural variation involves the topography of redirection demands. In most studies, the redirection tasks match the topography of the targeted stereotypy. For example, if aiming to reduce motor stereotypy, the interventionist interrupts motor stereotypy and redirects the individual to complete socially acceptable motor tasks such as motor imitation. Similarly, if aiming to reduce vocal stereotypy, the interventionist interrupts vocal stereotypy and redirects the individual to complete socially acceptable vocal responses such as answering questions. However, initial evidence indicates that the similarity between stereotypy and redirection topographies is not necessary for effective treatment. In fact, several studies have found RIRD involving redirection to appropriate motor tasks has successfully reduced vocal stereotypy, and redirection to appropriate vocalizations has successfully reduced motor stereotypy (e.g., Ahrens et al., 2011; Cassella, Sidener, Sidener, & Progar, 2011; Shawler & Miguel, 2015). These variations are particularly assuring because RIRD should not be limited to treat stereotypy only among individuals capable of emitting a variety of socially acceptable responses that match the topography of the targeted stereotypy. In other words, RIRD may be successful in reducing vocal stereotypy among individuals who can emit no appropriate vocal responses and only a few appropriate motor responses.

One of the many strengths of RIRD is the fact that it is successful in reducing both vocal and motor stereotypy. Additionally, although the primary purpose of RIRD is to reduce stereotypy, several studies have also found an increase in pro-social behaviors, such as appropriate vocalizations (e.g., Shawler & Miguel, 2015). This could be attributed to the fact that in the literature, RIRD often involves reinforcement procedures such as praise for appropriate vocalization and compliance with redirection demands.

Response blocking. Response blocking involves the interventionist physically preventing or redirecting a targeted response. For example, response blocking for hand mouthing could involve physically blocking a hand moving toward the mouth or physically removing a hand from the mouth. Response blocking has been shown to be an effective procedure to reduce motor stereotypy across several studies; however, the exact behavioral mechanisms which contribute to this success, punishment, sensory extinction, or prompting of alternative behaviors, have yet to be determined (Langone, Luiselli, & Hamill, 2013).

While effective, several factors should be considered when choosing to implement response blocking. First, response blocking may increase other forms of challenging behavior. Second, response blocking can be time intensive in that the interventionist must be in close proximity to the client at all times to block targeted stereotypy. Langone et al. (2013) addressed these two drawbacks by implementing punishment within a stimulus control arrangement. Specifically, the interventionist wore a tennis wristband during initial response blocking sessions. Later the tennis wristband alone, without implementation of response blocking, maintained low levels of motor stereotypy. More research is needed to determine the success of such procedures. The last consideration of response blocking involves evaluating the targeted stereotypy immediately following response blocking sessions. Rapp (2006) found response blocking to be effective at reducing stereotypy during intervention sessions; however, in the 5 min after response blocking sessions, the targeted stereotypy increased to levels above those observed immediately before intervention sessions.

Although response blocking is associated with some drawbacks, initial evidence supports the success of response blocking to reduce motor stereotypy. Needless to say, response blocking is physically incompatible with vocal stereotypy, as vocal responses cannot be physically blocked or redirected. With careful considerations, response blocking may be an effective treatment to use individually or as a component of a treatment package.

Hands-down. A hands-down procedure is somewhat of an extension of a response blocking procedure. While response blocking is typically terminated at the prevention or redirection of a targeted stereotypic response, the hands-down procedure involves extending this prevention or redirection by placing the individual's hand down, typically at his or her side or on a table, for a specific amount of time. Hands are manually kept in a specific location for a short period of time, typically spanning 1–10 s. Interventionists only use enough force to prevent occurrence of targeted stereotypy rather than attempt to completely immobilize an individual's hands.

The hands-down procedure is associated with the same drawbacks and considerations associated with the response blocking procedure. Prior to implementing this procedure, less intrusive procedures should have been introduced and shown to be ineffective as this is the most intrusive punisher reported in this chapter. If it is determined that this procedure is necessary, interventionists should also consider beginning the intervention with a shorter duration of hands-down (e.g., 1 s) and progress to longer hands-down durations if necessary to reach desirable treatment effects (e.g., Doughty et al., 2007).

Verbal interruption and reprimand. In some cases, stereotypy may be effectively treated with a mild verbal interruption and reprimand. This procedure involves interrupting the occurrence of stereotypy by delivering a reprimand. Reprimands typically involve a short statement indicating the targeted behavior should be terminated (e.g., "time for hands-down" or "no movie talk"). What differentiates verbal interruption and reprimand from response blocking is that the targeted stereotypy is not physically blocked from occurring; instead, the interventionist is simply providing redirection instructions. As a result, a strength of this procedure is that it can be used to reduce both vocal and motor stereotypy.

Although ease of implementation and the mild and unobtrusive nature of verbal interruption and reprimand make this procedure appealing, relatively few studies have evaluated the effectiveness of verbal interruption and reprimand in isolation. This procedure is most often introduced as one component of a treatment package (e.g., Rapp et al., 2017), and the results of studies in which it has been implemented in isolation have been mixed for effectiveness. Rapp, Patel, Ghezzi, O'Flaherty, and Titterington (2009) found verbal reprimands to be effective for reducing vocal stereotypy for two of three participants, but a response cost procedure was required to reduce vocal stereotypy for the third participant.

Response cost. Response cost is a negative punishment procedure in which a specific amount of reinforcement is lost contingent on the targeted behavior (Cooper et al., 2007). Because the procedure is predicated on the removal of reinforcement, the individual must initially have access to reinforcement which can be removed.

Several studies have evaluated the effectiveness of response cost to reduce both vocal and motor stereotypy. In the majority of these studies, response cost has been evaluated as part of a multicomponent treatment package. For example, Shillingsburg, Lomas, and Bradley (2012) evaluated the effectiveness of response cost first with noncontingent reinforcement and then with DRO. In the first phase of the study, the participant had noncontingent access to a preferred computer, but the computer was turned off for 20 s contingent on vocal stereotypy. In the second phase of the study, the participant earned tokens within a DRO contingency. Earning a specified number of tokens resulted in access to the preferred computer, but tokens were lost if he engaged in vocal stereotypy. Both treatment packages successfully reduced stereotypy.

With several studies indicating its potential effectiveness, response cost should be considered as an intervention to reduce levels of both vocal and motor stereotypy. However, rather than implemented as a treatment in isolation, response cost should be used as a component of a treatment package. Not only have most studies evaluated response cost as part of a treatment package, but also response cost would be impossible to implement if the individual did not have access to preferred stimuli which could be removed contingent on targeted stereotypy. Such preferred stimuli may be provided as part of a noncontingent reinforcement procedure, token economy, or other reinforcement-based procedure.

Conclusion

Within this chapter several research-based interventions for automatically maintained stereotypy have been described. These interventions include antecedent manipulations, reinforcement-based interventions, and punishment procedures. One of the more promising interventions appears to be establishing stimulus control with the inclusion of a punishment procedure. The benefits and initial positive findings in the literature make it a prime target for additional research. Overall, many of the studies discussed within the chapter included multicomponent treatment packages which should be a primary consideration for interventionists planning to address stereotypy. While there is sufficient evidence suggesting the interventions presented in the chapter are effective in reducing stereotypy for short durations, it is unclear to what extent these interventions remain effective over time. Future research should evaluate the effectiveness of interventions across longer periods of time (e.g., days, weeks, months) and provide recommendations for recapturing previously observed reductions in stereotypy following a collapse in treatment effectiveness (e.g., how and when to identify novel competing or matched stimuli).

References

Abang, T. B. (1985). Blindism: Likely causes and preventative measures. *Journal of Visual Impairment and Blindness, 79*, 400–401.

Ahearn, W. H., Clark, K. M., MacDonald, R. F., & Chung, B. I. (2007). Assessing and treating vocal stereotypy in children with autism. *Journal of Applied Behavior Analysis, 40*, 263–275. https://doi.org/10.1901/jaba.2007.30-06

Ahrens, E. N., Lerman, D. C., Kodak, T., Worsdell, A. S., & Keegan, C. (2011). Further evaluation of response interruption and redirection as treatment for stereotypy. *Journal of Applied Behavior Analysis, 44*, 95–108. https://doi.org/10.1901/jaba.2011.44-95

American Psychiatric Association. (2013). *Diagnostic and statistical manual of mental disorders* (5th ed.). Washington, DC: Author.

Anderson, C. M., Doughty, S. S., Doughty, A. H., Williams, D. C., & Saunders, K. J. (2010). Evaluation of stimulus control over a communication response as an intervention for stereotypical responding. *Journal of Applied Behavior Analysis, 43*, 333–339. https://doi.org/10.1901/jaba.2010.43-333

Azrin, N. H., & Holtz, W. C. (1966). Punishment. In W. K. Honig (Ed.), *Operant behavior: Areas of research and application* (pp. 380–447). New York: Appleton-Century Crofts.

Bak, S. (1999). Relationships between inappropriate behaviours and other factors in young children with visual impairments. *Review, 31*, 84–91.

Behavior Analyst Certification Board® (BACB®). (2014). Professional and ethical compliance code for behavior analysts. Retrieved from https://www.bacb.com/ethics/ethics-code/.

Bennett, K., Reichow, B., & Wolery, M. (2011). Effects of structured teaching on the behavior of young children with disabilities. *Focus on Autism And Other Developmental Disabilities, 26*, 143–152. https://doi.org/10.1177/1088357611405040

Bodfish, J., Symons, F., Parker, D., & Lewis, M. (2000). Varieties of repetitive behavior in autism: Comparisons to mental retardation. *Journal of Autism and Developmental Disorders, 30*, 237–243. https://doi.org/10.1023/A:1005596502855

Bowring, D. L., Totsika, V., Hastings, R. P., Toogood, S., & Griffith, M. (2017). Challenging behaviours in adults with an intellectual disability: A total population study and exploration of risk indices. *British Journal of Clinical Psychology, 56*, 16–32. https://doi.org/10.1111/bjc.12118

Brosnan, J., & Healy, O. (2011). A review of behavioral interventions for the treatment of aggression in individuals with developmental disabilities. *Research in Developmental Disabilities, 32*, 437–446. https://doi.org/10.1016/j.ridd.2010.12.023

Cassella, M. D., Sidener, T. M., Sidener, D. W., & Progar, P. R. (2011). Response interruption and redirection for vocal stereotypy in children with autism: A systematic replication. *Journal of Applied Behavior Analysis, 44*, 169–173. https://doi.org/10.1901/jaba.2011.44-169

Chebli, S., Martin, V., & Lanovaz, M. (2016). Prevalence of stereotypy in individuals with developmental disabilities: A systematic review. *Review Journal of Autism and Developmental Disorders, 3*, 107–118. https://doi.org/10.1007/s40489-016-0069-x

Chowdhury, M., & Benson, B. A. (2011). Use of differential reinforcement to reduce behavior problems in adults with intellectual disabilities: A methodological review. *Research in Developmental Disabilities, 32*, 383–394. https://doi.org/10.1016/j.ridd.2010.11.015

Colón, C. L., Ahearn, W. H., Clark, K. M., & Masalsky, J. (2012). The effects of verbal operant training and response interruption and redirection on appropriate and inappropriate vocalizations. *Journal of Applied Behavior Analysis, 45*, 107–120. https://doi.org/10.1901/jaba.2012.45-107

Cooper, J. O., Heron, T. E., & Heward, W. L. (2007). *Applied behavior analysis* (2nd ed.). Upper Saddle River, NJ: Pearson Education, Inc.

Cornish, K., & Pigram, J. (1996). Developmental and behavioural characteristics of Cri du Chat syndrome. *Archives of Disease in Childhood, 75*, 448–450. https://doi.org/10.1136/adc.75.5.448

Cunningham, A., & Shreibman, L. (2008). Stereotypy in autism: The importance of function. *Research in Autism Spectrum Disorders, 2*, 469–479. https://doi.org/10.1016/j.rasd.2007.09.006

Didden, R., Sturmey, P., Sigafoos, J., Lang, R., O'Reilly, M. F., & Lancioni, G. E. (2012). Nature, prevalence, and characteristics of challenging behavior. In J. L. Matson (Ed.), *Functional assessment for challenging behaviors* (pp. 25–44). New York: Springer New York.

DiGennaro Reed, F. D., & Lovett, B. J. (2008). Views on the efficacy and ethics of punishment: Results from a national survey. *International Journal of Behavioral Consultation and Therapy, 4*, 61–67. https://doi.org/10.1037/h0100832

Doughty, S. S., Anderson, C. M., Doughty, A. H., Williams, D. C., & Saunders, K. J. (2007). Discriminative control of punished stereotyped behavior in humans. *Journal of TheExperimental Analysis of Behavior, 87*, 325–336. https://doi.org/10.1901/jeab.2007.39-05

Downs, J., Parkinson, S., Ranelli, S., Leonard, H., Diener, P., & Lotan, M. (2014). Perspectives on hand function in girls and women with Rett syndrome. *Developmental Neurorehabilitation, 17*, 210–217. https://doi.org/10.3109/17518423.2012.758183

Dykens, E. M., & Smith, A. C. M. (1998). Distinctiveness and correlates of maladaptive behaviour in children and adolescents with Smith-Magenis syndrome. *Journal of Intellectual Disability Research, 42*, 481–489. https://doi.org/10.1046/j.1365-2788.1998.4260481.x

Emerson, E., & Bromley, J. (1995). The form and function of challenging behaviours. *Journal of Intellectual Disability Research, 39*, 388–398. https://doi.org/10.1111/j.1365-2788.1995.tb00543.x

Emerson, E., Kiernan, C., Alborz, A., Reeves, D., Mason, H., Swarbrick, R., … Hatton, C. (2001). The prevalence of challenging behaviors: A total population study. *Research in Developmental Disabilities, 22*, 77–93. https://doi.org/10.1016/S0891-4222(00)00061-5

Foxx, R. M. (2005). Severe aggressive and self-destructive behavior: The myth of the nonaversive treatment of severe behavior. In J. W. Jacobson, R. M. Foxx, & J. A. Mulick (Eds.), *Controversial therapies for developmental disabilities: Fad, fashion, and science in professional science* (pp. 295–310). Mahwah, NJ: Erlbaum.

Gehrman, C., Wilder, D. A., Forton, A. P., & Albert, K. (2017). Comparing resetting to non-resetting DRO procedures to reduce stereotypy in a child with autism. *Behavioral Interventions, 32*, 242–247. https://doi.org/10.1002/bin.1486

Greenberg, J. H., Lau, W., & Lau, S. (2016). Teaching appropriate play to replace stereotypy using a treatment package with students having autism. *Global Education Review, 3*, 94–104.

Groskreutz, M. P., Groskreutz, N. C., & Higbee, T. S. (2011). Response competition and stimulus preference in the treatment of automatically reinforced behavior: A comparison. *Journal of Applied Behavior Analysis, 44*, 211–215. https://doi.org/10.1901/jaba.2011.44-211

Hagerman, R. J., & Lampe, M. E. (1999). Fragile X syndrome. In S. Goldstein & C. R. Reynolds (Eds.), *Handbook of neurodevelopmental and genetic disorders in children*. New York: Guilford Press.

Hagopian, L. P., Rooker, G. W., & Zarcone, J. R. (2015). Delineating subtypes of self-injurious behavior maintained by automatic reinforcement. *Journal of Applied Behavior Analysis, 48*, 523–543. https://doi.org/10.1002/jaba.236

Hanley, G. P., Iwata, B. A., & McCord, B. E. (2003). Functional analysis of problem behavior: A review. *Journal of Applied Behavior Analysis, 36*, 147–185. https://doi.org/10.1901/jaba.2003.36-147

Higbee, T. S., Chang, S., & Endicott, K. (2005). Noncontingent access to preferred sensory stimuli as a treatment for automatically reinforced stereotypy. *Behavioral Interventions, 20*, 177–184. https://doi.org/10.1002/bin.190

Hill, L., Trusler, K., Furniss, F., & Lancioni, G. (2012). Effects of multisensory environments on stereotyped behaviours assessed as maintained by automatic reinforcement. *Journal of Applied Research in Intellectual Disabilities, 25*, 509–521. https://doi.org/10.1111/j.1468-3148.2012.00697.x

Holden, B., & Gitlesen, J. P. (2006). A total population study of challenging behaviour in the county of Hedmark, Norway: Prevalence and risk markers. *Research in Developmental Disabilities, 27*, 456–465. https://doi.org/10.1016/j.ridd.2005.06.001

Horner, R. H., Dunlap, G., Koegel, R. L., Carr, E. G., Sailor, W., Anderson, J., … O'Neill, R. E. (2005). Toward a technology of "nonaversive" behavioral support. Research and Practice for Persons with Severe Disabilities, 30, 3-10. doi: https://doi.org/10.2511/rpsd.30.1.3

Hyman, P., Oliver, C., & Hall, S. (2002). Self-injurious behavior, self-restraint, and compulsive behaviors in Cornelia de Lange syndrome. *American Journal of Mental Deficiency, 107*, 146–154. https://doi.org/10.1352/0895-8017(2002)107<0146:SIBSRA>2.0.CO;2

Iwata, B. A., DeLeon, I. G., & Roscoe, E. M. (2013). Reliability and validity of the functional analysis screening tool. *Journal of Applied Behavior Analysis, 46*, 271–284. https://doi.org/10.1002/jaba.31

Iwata, B. A., Dorsey, M. F., Slifer, K. J., Bauman, K. E., & Richman, G. S. (1994). Toward a functional analysis of self-injury. *Journal of Applied Behavior Analysis, 27*, 197–209. https://doi.org/10.1901/jaba.1994.27-197. (Reprinted from *Analysis and Intervention in Developmental Disabilities*, *2,* 3-20, 1982).

Iwata, B. A., Duncan, B. A., Zarcone, J. R., Lerman, D. C., & Shore, B. A. (1994). A sequential, test control methodology for conducting functional analyses of self-injurious behavior. *Behavior Modification, 18*, 289–306. https://doi.org/10.1177/01454455940183003

Jones, S., Cooper, S.-A., Smiley, E., Allen, L., Williamson, A., & Morrison, J. (2008). Prevalence of, and factors associated with, problem behaviors in adults with intellectual disabilities. *The Journal of Nervous and Mental Disease, 196*, 678–686. https://doi.org/10.1097/NMD.0b013e318183f85c

Lachiewicz, A. M., Spiridigliozzi, G. A., Gullion, C. M., Ransford, S. N., & Rao, K. (1994). Aberrant behavior of young boys with fragile X syndrome. *American Journal on Mental Retardation, 98*, 567–579.

Lam, K. L., & Aman, M. G. (2007). The repetitive behavior scale-revised: Independent validation in individuals with autism spectrum disorders. *Journal of Autism and Developmental Disorders, 37*, 855–866. https://doi.org/10.1007/s10803-006-0213-z

Lancioni, G. E., Singh, N. N., O'Reilly, M. F., & Sigafoos, J. (2009). An overview of behavioral strategies for reducing hand-related stereotypies of persons with severe to profound intellectual and multiple disabilities: 1995-2007. *Research in Developmental Disabilities, 30*, 20–43. https://doi.org/10.1016/j.ridd.2008.02.002

Lancioni, G. E., Singh, N. N., O'Reilly, M. F., Sigafoos, J., Didden, R., Smaldone, A., & Oliva, D. (2008). Helping a man with multiple disabilities increase object-contact responses and reduce hand stereotypy via a microswitch cluster program. *Journal of Intellectual and Developmental Disability, 33*, 349–353. https://doi.org/10.1080/13668250802441664

Lang, R., O'Reilly, M., Sigafoos, J., Lancioni, G. E., Machalicek, W., Rispoli, M., & White, P. (2009). Enhancing the effectiveness of a play intervention by abolishing the reinforcing value of stereotypy: A pilot study. *Journal of Applied Behavior Analysis, 42*, 889–894. https://doi.org/10.1901/jaba.2009.42-889

Lang, R., O'Reilly, M., Sigafoos, J., Machalicek, W., Rispoli, M., Lancioni, G. E., … Fragale, C. (2010). The effects of an abolishing operation intervention component on play skills, challenging behavior, and stereotypy. *Behavior Modification, 34*(267), 289. https://doi.org/10.1177/0145445510370713

Langone, S. R., Luiselli, J. K., & Hamill, J. (2013). Effects of response blocking and programmed stimulus control on motor stereotypy: A pilot study. *Child and Family Behavior Therapy, 35*, 249–255. https://doi.org/10.1080/07317107.2013.818906

Lanovaz, M. J., & Sladeczek, I. E. (2012). Vocal stereotypy in individuals with autism spectrum disorders: A review of behavioral interventions. *Behavior Modification, 36*, 146–164. https://doi.org/10.1177/0145445511427192

Lanovaz, M. J., Sladeczek, I. E., & Rapp, J. T. (2011). Effects of music on vocal stereotypy in children with autism. *Journal of Applied Behavior Analysis, 44*, 647–651. https://doi.org/10.1901/jaba.2011.44-647

Laraway, S., Snycerski, S., Michael, J., & Poling, A. (2003). Motivating operations and terms to describe them: Some further refinements. *Journal of Applied Behavior Analysis, 36*, 407–414. https://doi.org/10.1901/jaba.2003.36-407

Leekam, S. R., Prior, M. R., & Uljarevic, M. (2011). Restricted and repetitive behaviors in autism spectrum disorders: A review of research in the last decade. *Psychological Bulletin, 137*, 562–593. https://doi.org/10.1037/a0023341

LeMonda, B. C., Holtzer, R., & Goldman, S. (2012). Relationship between executive functions and children with Autistic Disorder. *Research in Autism Spectrum Disorders, 6*, 1099–1106. https://doi.org/10.1016/j.rasd.2012.03.001

Lerman, D. C., & Vorndran, C. M. (2002). On the status of knowledge for using punishment: Implications for treating behavior disorders. *Journal of Applied Behavior Analysis, 35*, 431–464. https://doi.org/10.1901/jaba.2002.35-431

Love, J. J., Miguel, C. F., Fernand, J. K., & LaBrie, J. K. (2012). The effects of matched stimulation and response interruption and redirection on vocal stereotypy. *Journal of Applied Behavior Analysis, 45*, 549–564. https://doi.org/10.1901/jaba.2012.45-549

Lowe, K., Allen, D., Jones, E., Brophy, S., Moore, K., & Jones, W. (2007). Challenging behaviours: Prevalence and topographies. *Journal of Intellectual Disability Research, 51*, 625–636. https://doi.org/10.1111/j.1365-2788.2006.00948.x

Martinez, C. K., Betz, A. M., Liddon, C. J., & Werle, R. L. (2016). A progression to transfer RIRD to the natural environment. *Behavioral Interventions, 31*, 144–162. https://doi.org/10.1002/bin.1444

Matson, J. L., & Dempsey, T. (2008). Stereotypy in adults with autism spectrum disorders: Relationship and diagnostic fidelity. *Journal of Developmental and Physical Disabilities, 20*, 15–165. https://doi.org/10.1007/s10882-007-9086-0

Matson, J. L., Dempsey, T., & Fodstad, J. C. (2009). Stereotypies and repetitive/restrictive behaviours in infants with autism and pervasive developmental disorder. *Developmental Neurorehabilitation, 12*, 122–127. https://doi.org/10.1080/17518420902936730

McClintock, K., Hall, S., & Oliver, C. (2003). Risk markers associated with challenging behaviours in people with intellectual disabilities: A meta-analytic study. *Journal of Intellectual Disability Research, 47*, 405–416.

McTiernan, A., Leader, G., Healy, O., & Mannion, A. (2011). Analysis of risk factors and early predictors of challenging behavior for children with autism spectrum disorder. *Research in Autism Spectrum Disorder, 5*, 1215–1222. https://doi.org/10.1016/j.rasd.2011.01.009

Miguel, C. F., Clark, K., Tereshko, L., & Ahearn, W. H. (2009). The effects of response interruption and redirection and sertraline on vocal stereotypy. *Journal of Applied Behavior Analysis, 42*, 883–888. https://doi.org/10.1901/jaba.2009.42-883

Northup, J., Wacker, D., Sasso, G., Steege, M., Cigrand, K., Cook, J., & DeRaad, A. (1991). A brief functional analysis of aggressive and alternative behavior in an outclinic setting. *Journal of Applied Behavior Analysis, 24*, 509–522. https://doi.org/10.1901/jaba.1991.24-509

Nuernberger, J. E., Vargo, K. K., & Ringdahl, J. E. (2013). An application of differential reinforcement of other behavior and self-monitoring to address repetitive behavior. *Journal of Developmental and Physical Disabilities, 25*, 105–117. https://doi.org/10.1007/s10882-012-9309-x

Oliver, C., Arron, K., Sloneem, J., & Hall, S. (2008). Behavioural phenotype of Cornelia de Lange syndrome: Case–control study. *The British Journal of Psychiatry, 193*, 466–470. https://doi.org/10.1192/bjp.bp.107.044370

Olson, B., & Rett, A. (1985). Behavioral observations concerning differential diagnosis between Rett syndrome and autism. *Brain and Development, 6*, 281–289. https://doi.org/10.1016/S0387-7604(85)80029-2

Querim, A. C., Iwata, B. A., Roscoe, E. M., Schlichenmeyer, K. J., Virgues Ortego, J., & Hurl, K. E. (2013). Fictional analysis screening for problem behavior maintained by automatic reinforcement. *Journal of Applied Behavior Analysis, 46*, 47–60. https://doi.org/10.1002/jaba.26

Rapp, J. T. (2006). Toward an empirical method for identifying matched stimulation for automatically reinforced behavior: A preliminary investigation. *Journal of Applied Behavior Analysis, 39*, 137–140. https://doi.org/10.1901/jaba.2006.37-05

Rapp, J. T., Cook, J. L., McHugh, C., & Mann, K. R. (2017). Decreasing stereotypy using NCR and DRO with functionally matched stimulation: Effects on targeted and non-targeted stereotypy. *Behavior Modification, 41*, 45–83. https://doi.org/10.1177/0145445516652370

Rapp, J. T., Patel, M. R., Ghezzi, P. M., O'Flaherty, C. H., & Titterington, C. J. (2009). Establishing stimulus control of vocal stereotypy displayed by young children with autism. *Behavioral Interventions, 24*, 85–105. https://doi.org/10.1002/bin.276

Rapp, J. T., & Vollmer, T. R. (2005). Stereotypy I: A review of behavioral assessment and treatment. *Research in Developmental Disabilities, 26*, 527–547. https://doi.org/10.1016/j.ridd.2004.11.005

Rispoli, M., Camargo, S. H., Neely, L., Gerow, S., Lang, R., Goodwyn, F., & Ninci, J. (2014). Pre-session satiation as a treatment for stereotypy during group activities. *Behavior Modification, 38*, 392–411. https://doi.org/10.1177/0145445513511631

Saini, V., Gregory, M. K., Uran, K. J., & Fantetti, M. A. (2015). Parametric analysis of response interruption and redirection as treatment for stereotypy. *Journal of Applied Behavior Analysis, 48*, 96–106. https://doi.org/10.1002/jaba.186

Saini, V., Miller, S. A., & Fisher, W. W. (2016). Multiple schedules in practical application: Research trends and implications for future investigation. *Journal of Applied Behavior Analysis, 49*, 421–444. https://doi.org/10.1002/jaba.300

Sarimski, K. (1997). Communication, social-emotional development and parenting stress in Cornelia de Lange Syndrome. *Journal of Intellectual Disability Research, 41*, 70–75. https://doi.org/10.1111/j.1365-2788.1997.tb00678.x

Saylor, S., Sidener, T. M., Reeve, S. A., Fetherston, A., & Progar, P. R. (2012). Effects of three types of noncontingent auditory stimulation on vocal stereotypy in children with autism. *Journal of Applied Behavior Analysis, 45*, 185–190. https://doi.org/10.1901/jaba.2012.45-185

Schlaggar, B. L., & Mink, J. W. (2003). Movement disorders in children. *Pediatrics in Review, 24*, 39–51. https://doi.org/10.1542/pir.24-2-39

Shamlian, K. D., Fisher, W. W., Steege, M. W., Cavanaugh, B. M., Samour, K., & Querim, A. C. (2016). Evaluation of multiple schedules with naturally occurring and therapist arranged discriminative stimuli following functional communication training. *Journal of Applied Behavior Analysis, 49*, 228–250. https://doi.org/10.1002/jaba.293

Shawler, L. A., & Miguel, C. F. (2015). The effects of motor and vocal response interruption and redirection on vocal stereotypy and appropriate vocalizations. *Behavioral Interventions, 30*, 112–134. https://doi.org/10.1002/bin.1407

Shillingsburg, M. A., Lomas, J. E., & Bradley, D. (2012). Treatment of vocal stereotypy in an analogue and classroom setting. *Behavioral Interventions, 27*, 151–163. https://doi.org/10.1002/bin.1340

Sidener, T. M., Carr, J. E., & Firth, A. M. (2005). Superimposition and withholding of edible consequences as treatment for automatically reinforced stereotypy. *Journal of Applied Behavior Analysis, 38*, 121–124. https://doi.org/10.1901/jaba.2005.58-04

Singer, H. S., Mink, J. W., Gilbert, D. L., & Jankovic, J. (2016). *Movement disorders in childhood* (2nd ed.). San Diego, CA: Elsevier Academic Press.

Summers, J. A., Allison, D. B., Lynch, P. S., & Sandler, L. (1995). Behaviour problems in Angelman syndrome. *Journal of Intellectual Disability Research, 39*, 97–106. https://doi.org/10.1111/j.1365-2788.1995.tb00477.x

Symons, F. J., Clark, R. D., Hatton, D. D., Skinner, M., & Bailey, D. B. (2003). Self-injurious behavior in young boys with fragile X syndrome. *American Journal of Medical Genetics, 118A*, 115–121. https://doi.org/10.1111/j.1365-2788.2004.00632.x

Taylor, B. A., Hoch, H., & Weissman, M. (2005). The analysis and treatment of vocal stereotypy in a child with autism. *Behavioral Interventions, 20*, 239–253. https://doi.org/10.1002/bin.200

Tiger, J. H., Hanley, G. P., & Bruzek, J. (2008). Functional communication training: A review and practical guide. *Behavior Analysis in Practice, 1*, 16–23. https://doi.org/10.1007/BF03391716

Tiger, J. H., Wierzba, B. C., Fisher, W. W., & Benitez, B. B. (2017). Developing and demonstrating inhibitory stimulus control over repetitive behavior. *Behavioral Interventions, 32*, 160–174. https://doi.org/10.1002/bin.1472

Tröster, H. (1994). Prevalence and functions of stereotyped behaviors in nonhandicapped children in residential care. *Journal of Abnormal Child Psychology, 1*, 79–97. https://doi.org/10.1007/BF02169257

Turner, M. (1999). Annotation: Repetitive behaviour in autism: A review of psychological research. *The Journal of Child Psychology and Psychiatry, 40*, 839–849. https://doi.org/10.1111/1469-7610.00502

Van Houten, R., Axelrod, S., Bailey, J. S., Favell, J. E., Foxx, R. M., Iwata, B. A., & Lovaas, O. I. (1988). The right to effective behavioral treatment. *Journal of Applied Behavior Analysis, 21*, 381–384. https://doi.org/10.1901/jaba.1988.21-381

Watkins, N., Paananen, L., Rudrud, E., & Rapp, J. T. (2011). Treating vocal stereotypy with environmental enrichment and response cost. *Clinical Case Studies, 10*, 440–448. https://doi.org/10.1177/1534650111429377

Watkins, N., & Rapp, J. T. (2014). Environmental enrichment and response cost: Immediate and subsequent effects on stereotypy. *Journal of Applied Behavior Analysis, 47*, 186–191. https://doi.org/10.1002/jaba.97

Self-Injurious Behavior

32

Mindy Scheithauer, Colin Muething,
Kristina R. Gerencser, and Nathan A. Call

Introduction

Given that the term self-injurious behavior (SIB) may seem self-explanatory, some may be surprised at how the literature struggles to reach a consensus definition (Rojahn, Schroeder, & Hoch, 2007). Some discussions on the topic even point out that certain behaviors that cause tissue damage to the person who emits them are common in most cultures, such as picking a scab, or even socially acceptable, such as piercing one's ears. Even a cursory review of the topic brings one in contact with a host of definitions and potential synonyms, such as non-suicidal self-injury and self-harm behaviors (Jacobson & Gould, 2007). Such inconsistency in terminology is an indicator of how different disciplines focus on specific forms of the behavior or specific subgroups of individuals who engage in this type of behavior (Rojahn et al., 2007). For example, behaviors such as cutting exhibited by individuals without intellectual and/or developmental disabilities (IDD) have been related to diagnoses of anxiety and depression (Jacobson & Gould, 2007) and have therefore been studied in the psychiatry and psychology fields by individuals with training in these disorders. That literature often makes mention of the intentionality of these self-harm behaviors, with converging evidence suggesting that it may serve to aid in the regulation of negative affect or arousal (Klonsky, 2007).

SIB exhibited by individuals with IDD is less likely to be covert and more likely to have repetitive qualities than self-harm behaviors occurring in populations without IDD (Whitlock, Eckenrode, & Silverman, 2006). Although it is possible that individuals with IDD may engage in self-harm of the type more commonly seen in individuals without IDD, the literature on these different types of SIB generally do not overlap. This makes it difficult to ascertain whether, or to what degree, this may occur. The specific forms of SIB, or the *topographies*, most often emitted by individuals with IDD also tend to differ from those without IDD and include head or body hitting, banging the head or other extremities on hard surfaces, self-biting, skin picking, eye-gouging, etc. This type of SIB is generally studied by psychologists with training in IDD, behavior analysts, and special educators (Rojahn et al., 2007). It is also important to distinguish between SIB exhibited by individuals with IDD from other behaviors prevalent in this population

---

M. Scheithauer (✉) · C. Muething · K. R. Gerencser
N. A. Call
Department of Pediatrics, Division of Autism and Related Disorders, Emory University School of Medicine, Atlanta, GA, USA

Marcus Autism Center, Atlanta, GA, USA
e-mail: Mindy.Scheithauer@choa.org; Colin.Muething@choa.org; Krstina.Gerencser@choa.org; Nathan.Call@choa.org

J. L. Matson (ed.), *Handbook of Intellectual Disabilities*, Autism and Child Psychopathology Series, https://doi.org/10.1007/978-3-030-20843-1_32

that pose significant risk of harm, such as pica or elopement. In contrast to these behaviors, the SIB directly produces tissue damage, whereas these other dangerous behaviors may produce damage only as an indirect consequence.

Prevalence and Risk Factors

Estimating the prevalence of SIB is complicated, as much of the research on the topic employs differing definitions, instruments, and sampling strategies (Rojahn et al., 2007). For example, a point prevalence study found that 14% of individuals with IDD engaged in SIB (Griffin, Williams, Stark, Altmeyer, & Mason, 1986). However, all of the individuals evaluated resided in facilities in the same state, making it challenging to ascertain how well such a finding can be extended to the broader population of individuals with IDD. In contrast, a study of all of the individuals with IDD in Sweden found that 31% engaged in SIB (Lundqvist, 2013). Yet a similar study in England found that only 4% engaged in SIB (Emerson et al., 2001a). Thus, it is most appropriate to assume that the prevalence of SIB ranges between 4% and 30%, depending on the particular study.

Aside from overall incidence, establishing prevalence of SIB among certain subgroups can be helpful in identifying risk factors and developmental trajectories. For example, researchers have examined gender disparities in the prevalence of certain challenging behaviors, finding that certain topographies are more commonly observed in males, such as aggression (McClintock, Hall, & Oliver, 2003). However, such a gender difference does not seem to appear for SIB (Baghdadli, Pascal, Grisi, & Aussilloux, 2003), with similar rates found in males and females.

One of the variables most strongly correlated with SIB is the level of intellectual disability (Borthwick, Meyers, & Eyman, 1981; Griffin et al., 1986). In a meta-analysis of 22 prior studies, the authors found a highly significant association between the degree of intellectual disability and SIB, with SIB more likely to occur in individuals with a severe or profound degree of intellectual disability compared to those with a mild or moderate degree of intellectual disability (McClintock et al., 2003). The same study demonstrated that SIB was more likely to occur in individuals with deficits in receptive and expressive language than those without such language deficits.

The prevalence of SIB also varies based on the etiology of the individual's IDD, with higher prevalence among individuals with IDD associated with specific genetic disorders. For example, Arron, Oliver, Berg, Moss, and Burbidge (2011) identified prevalence rates of SIB of 45% for Angelman syndrome, 77% for Cri du Chat syndrome, 70% in Cornelia de Lange syndrome, 51% in Fragile X syndrome, 64% in Lowe syndrome, 52% in Prader-Willi syndrome, and 93% in Smith-Magenis syndrome. Self-injury is also highly prevalent in Lesch-Nyhan syndrome, with over 90% of individuals engaging in SIB (Anderson & Ernst, 1994). Yet, for other syndromic forms of IDD (e.g., Down syndrome), SIB is less common except in those with profound intellectual disabilities (Cooper et al., 2009). Additionally, certain syndromes are associated with increased prevalence of specific topographies of SIB. For example, of individuals with Prader-Willi who engage in SIB, 75% exhibit skin picking (Neidert, 2007). Individuals with Smith-Magenis and Cri du Chat syndromes are far more likely to engage in pulling on themselves (e.g., pulling out hair or nails) compared to individuals with idiopathic IDD (Arron et al., 2011; Finucane, Haines Dirrigl, & Simon, 2001; Smith, Dykens, & Greenberg, 1998). Similarly, individuals with Lesch-Nyhan who are self-injurious are likely to engage in self-biting (93%; Anderson & Ernst).

Of the developmental disabilities associated with intellectual disabilities, perhaps the most research on factors associated with increased risk of SIB has occurred with autism spectrum disorder (ASD). Therefore, we turn to this literature to identify other potential risk factors. Estimates of the prevalence of SIB in individuals with ASD range from 35% to 60% (Baghdadli et al., 2003; Rattaz, Michelon, & Baghdadli, 2015; Richards, Oliver, Moss, & Nelson, 2012; Richman et al., 2013;

Shattuck et al., 2007). Individuals with co-occurring ASD and intellectual disability are more likely to engage in SIB than those with ASD or intellectual disability alone (Lundqvist, 2013; McClintock et al., 2003; Rojahn, Wilkins, Matson, & Boisjoli, 2010). However, in at least one study, developmental regression was not found to be associated with increased probability of SIB (Lance, York, Lee, & Zimmerman, 2014). Severity of ASD symptomatology (Baghdadli et al., 2003, 2008), early speech deficits (Baghdadli et al., 2008), stereotypy (Richman et al., 2013), atypical sensory processing (Duerden et al., 2012), impulsivity (Duerden et al., 2012; Richman et al., 2013), and limited daily living skills (Baghdadli et al., 2003, 2008) have also all been found as related to SIB.

Development of Self-Injurious Behavior

It is important to note that SIB can occur as part of typical development, with up to 15% of typically developing children engaging in SIB (Romanczyk, Kistner, & Plienis, 1982). The age of onset of SIB for typically developing peers is similar to those found in retrospective studies of individuals with IDD who engaged in SIB, with most children beginning to engage in SIB prior to the age of 5 (Schneider, Bijam-Schulte, Janssen, & Stolk, 1996) and an average age of onset of 17 months (Kurtz et al., 2003). Kurtz, Chin, Huete, & Cataldo (2012) followed 32 children under the age of 5 who were at risk for IDD and engaged in SIB and found that more than half had an onset of SIB before 1 year of age.

Although the age of onset of SIB may be similar between typically developing children and those with IDD, the prevalence rates among individuals with IDD described above are much higher than the estimated 15% of typically developing children. To better understand the higher prevalence of SIB among children with IDD, it is important to understand the course of emerging SIB. To study this, Richman and Lindauer (2005) conducted a longitudinal study, tracking SIB over the course of about a year using direct observation of 12 children (age 12–32 months). They found that for some children who began engaging in SIB, the topography was similar to a past stereotypic behavior that did not cause damage or physical harm. For example, some of the participants who engaged in head hitting previously engaged in arm-waving in the head and shoulder region. In another study, Oliver, Petty, Ruddick, and Bacarese-Hamilton (2012) found that, in a sample of 943 children with severe intellectual disability, those who engaged in high-frequency repetitive behaviors were more likely to engage in SIB than those who did not engage in high-frequency repetitive behavior. However, increased odds for other topographies of challenging behavior were also associated with repetitive behaviors, making the specific relationship between repetitive behaviors and SIB unclear. Alternatively, it is possible that repetitive behaviors are related to challenging behaviors more broadly or that some alternative variable is causing both repetitive behaviors and challenging behaviors to occur. There is a need for additional research to evaluate the relationship between repetitive behaviors and the development of SIB specifically.

A major difference in SIB between typically developing individuals and those with IDD is the persistence of the behavior. In typical development, SIB usually resolves by age 5 (Romanczyk et al., 1982). For individuals with IDD, the course of SIB is more heterogeneous, as there are examples in the literature of SIB remitting, as well as several examples of SIB occurring well into adulthood (e.g., Cooper et al., 2009). Cooper and colleagues sought to identify the course of SIB in adults with IDD by following a cohort of participants for a 2-year period. Some individuals exhibited occasional SIB followed by a remission period (39% of individuals with SIB). However, SIB persisted for the majority of individuals (62%). Other research has also supported the tendency toward persistence of SIB among individuals with IDD, with estimates of 71% of individuals engaging in SIB continuing to do so over 7 years (Emerson et al., 2001b) and 84% persistence over 20 years in another study (Taylor, Oliver, & Murphy, 2011).

Impact of Self-Injurious Behavior

SIB can have a profound negative impact on the individuals who emit the behavior, with the most obvious negative consequence being physical injury. In less severe cases, most topographies of SIB have the potential to cause bruising, redness, and lacerations. In severe cases, many topographies, such as head hitting, head banging, self-biting, or eye-gouging, have the potential to cause permanent physical injury (Anderson & Ernst, 1994) and may cause wounds that increase the risk of infection or loss of sensory function (e.g., Barrera, Violo, & Graver, 2007).

In addition to the detrimental effect of causing physical harm, SIB also has the potential to produce significant negative collateral outcomes. For one, SIB increases the risk of restrictive placements, including hospitalization (Mandell, 2008) and residential care (Allen, Lowe, Moore, & Brophy, 2007; Lakin, Hill, Hauber, Bruininks, & Heal, 1983). Such out-of-home placements may further impede quality of life by limiting access to educational, employment, or leisure opportunities (Crossland, Burns, Leach, & Quinn, 2005). Individuals with IDD who engage in SIB are also more prone to experience manual or mechanical restraints (Jones, Allen, Moore, Phillips, & Lowe, 2007).

The presence of SIB can also be a substantial stressor for caregivers. Parents of children with IDD who exhibit challenging behavior in general are often significantly more stressed than caregivers of children without behavioral concerns (Davis & Gavidia-Payne, 2009). The presence of challenging behavior, including SIB, can negatively affect caregiver's quality of life through outcomes such as decreased marital satisfaction (Hall, Bouldin, Andresen, & Ali, 2012) and challenges with employment (Hartley, Barker, Baker, Seltzer, & Greenberg, 2012). For instance, it is not uncommon for the parents of children with severe SIB to receive calls from their child's school to retrieve their child when school personnel find it challenging to keep the student safe. In addition, when at home, children with severe SIB often require increased supervision to prevent injuries. This resulting time and need for vigilance can result in increased callouts from work and decreased time for self-care on the part of the caregiver.

Causes of Self-Injurious Behavior

In a seminal paper, Carr (1977) explored several theoretical explanations of the possible origins of SIB. These included an operant behavior hypothesis, which conceptualized SIB as a learned operant behavior (maintained by positive or negative social reinforcement); a self-stimulatory hypothesis in which SIB is thought to produce sensory stimulation that maintains the behavior; an organic hypothesis in which SIB is the result of abnormal physiological processes; and a psychodynamic hypothesis, in which SIB is meant to reduce guilt or assist with the ego. Carr reviewed the literature supporting each of these theories. He found several examples in the literature to support the operant and self-stimulatory hypotheses, based on changes in SIB that occurred following environmental manipulations. He also found support for the organic theory from studies demonstrating an association between certain topographies of SIB and specific genetic syndromes, changes in SIB related to medication changes, and increased rates of SIB among individuals experiencing potentially painful physiological conditions (e.g., ear infections). Carr dismissed the psychodynamic theory of SIB as unsubstantiated, likely as a result of the constructs being difficult to operationally define in individuals with IDD.

In summary, Carr (1977) identified a few potential hypotheses for causes of SIB that have since been studied in more depth over the past 40 years. The organic hypothesis has been further studied through potential neurobiological models of SIB, including the underlying changes in neurotransmitters related to the occurrence of SIB. Additional evidence has also emerged related to how SIB may be associated with pain and sensory experiences. In today's literature, these theories are often referred to as sensory models of SIB. Carr's operant and self-stimulatory models have also continued to receive attention, specifically in the domain of applied behavior analysis and behavioral models of SIB. These various models are described in more detail in this section.

Neurobiological Models of Self-Injurious Behavior

Several studies have attempted to identify underlying neurological causes and correlates of SIB exhibited by individuals with IDD. At the present time, there is no overarching consensus on a single neurobiological model of SIB, but numerous hypotheses have been proposed in the literature. It is important to note that several neurobiological models have acquired significant support from preclinical work with animals but remain unstudied in humans. A review of this literature is beyond the scope of this chapter (for an overview of this work, see Devine [2012]). Instead, we present a few of the most prominent neurobiological models and support for them within human research.

Endorphin dysregulation and opioid release are related neurobiological mechanisms that may be associated with SIB. Specifically, endorphins serve as inhibitory neuropeptides that affect pain perception. Some theories suggest that individuals who engage in SIB do so to access the release of endogenous opiates that occur as a result of the behavior (Richardson & Zaleski, 1983; Sandman & Kemp, 2011). In this model, SIB may be considered an addictive behavior for which the subsequent release of opiates acts as reinforcement for the behavior. Support for this theory comes from research identifying higher levels of endorphins among individuals who engage in SIB compared to control groups (Sandman & Kemp, 2011).

There is also varying support for the role of dopamine in SIB. Dopamine has a heavy concentration in the caudate nucleus. This is relevant to SIB because evidence exists for a relationship between diminished caudate nuclei volumes and diagnoses of Lesch-Nyhan syndrome, which is characterized by high prevalence of SIB (Wong et al., 1996). MRI studies also indicate decreased caudate nuclei volumes associated with SIB with individuals with ASD and Fragile X syndrome (Wolff, Hazlett, Lightbody, Reiss, & Piven, 2013). Given the high concentration of dopamine in the caudate nuclei and the diminished size of this area in individuals who engage in high rates of SIB, researchers have suggested differences in dopamine levels as potentially contributing to SIB in the neurobiological model of SIB.

A relationship between SIB and the neurotransmitters glutamate and gamma-aminobutyric acid (GABA) has also been proposed, as elevated rates of glutamate in the plasma levels of individuals with ASD and related disorders have been consistently noted (e.g., Rojas, 2014; Shinohe et al., 2006). The roles of serotonin (5-HT) in SIB have also been discussed, as there are consistent findings of elevated blood levels of 5-HT in ASD (e.g., Adamsen et al., 2014; Muller, Anacker, & Veenstra-VanderWeele, 2016). Given the high prevalence of SIB in ASD, some researchers have proposed a potential connection worthy of additional study.

Additional support for several of these theories comes from research on the effects of drugs that impact these various biological processes and the impact that they have on SIB. A brief summary of some of the more commonly studied drugs, and how they may impact and subsequently lend support to some of the above theories, can be found in the treatment section below.

Sensory Models of Self-Injurious Behavior

Although less systematic research has been conducted on these models, a common proposed hypothesis for the occurrence of SIB relates to sensory abnormalities. Specifically, the concept of pain has received significant attention in the literature. In typically developing individuals, pain is mediated by nociceptors, which are peripheral sensory neurons that have a high threshold for activation. The sensitivity of nociceptors can be altered based on repeated exposure to physical trauma or most relevant to this subject, SIB. Specifically, unlike the opioid model proposed above, this framework suggests that individuals who engage in SIB may have nociceptors that are more sensitive to pain and therefore actually have a diminished pain threshold (Symons, Harper, McGrath, Breau, & Bodfish, 2009; Symons, Shinde, Clary, Harper, & Bodfish, 2010). This decreased threshold may then create abnormal sensory experiences that are related to the causes of SIB.

A related sensory theory is that SIB occurs as a means of promoting immune response in reaction to illness or other physiological issues. For instance, SIB frequently causes inflammation, which subsequently can result in the body releasing an immune response or an instinctive response to guard that area from future damage (Peebles & Price, 2012; Symons, 2011). This theory may partially explain studies identifying increased rates of SIB during times when the individual is likely experiencing physiological discomfort from an underlying medical condition (Bosch, Van Dyke, Smith, & Poulton, 1997). For example, SIB has been shown to be related to allergy symptoms (Kennedy & Meyer, 1996), menses (Taylor, Rush, Hetrick, & Sandman, 1993), and ear infections (Carr & Smith, 1995).

Behavioral Models of Self-Injurious Behavior

In addition to biological factors that may increase the risk of SIB, environmental factors can lead to the emergence and maintenance of SIB. The behavioral model views all behaviors, including those that are problematic like SIB, as a function of the consequences that maintain it and the antecedents or environmental events that evoke it (Hanley, Iwata, & McCord, 2003; Iwata, Dorsey, Slifer, Bauman, & Richman, 1982/1994). This model views SIB as learned (i.e., operant) behavior. For example, a young child may engage in a tantrum when asked to brush his or her teeth. During the course of the tantrum, the child may drop to the ground and end up hitting his head on the floor. Given that most caregivers' first concern is their child's safety, it is likely that the child's caregivers respond to the head hitting by shifting their focus from brushing teeth to ensuring the child is safe and preventing further injury. Some may even soothe the child or offer preferred items in an attempt to calm him (Thompson & Iwata, 2007). Although this strategy is logical in terms of ensuring the safety of the child, some children with IDD may subsequently learn that in the past this behavior of head banging has resulted in the removal of an aversive demand of teeth brushing or the presentation of attention from a caregiver, which are both likely preferred outcomes (Beavers, Iwata, & Lerman, 2013).

As discussed above, SIB is more likely to occur among individuals with language and communication deficits. The behavioral model views SIB as communicative. In the above example, the child's head banging might be a means of communicating a desire to avoid brushing his teeth (Tiger, Hanley, & Bruzek, 2008). Assuming that SIB has historically resulted in the removal of demands, the child is more likely to engage in SIB in future events when an aversive demand is presented. In this situation, we would say that SIB is maintained by access to escape or that the function of the behavior is escape from demands (Hanley et al., 2003). Similarly, in a situation where a preferred toy is removed, the child engages in SIB, and the toy is subsequently returned; the SIB might be viewed as a request for the toy to be returned. Said another way, if SIB has historically resulted in access to preferred items, in future situations when a toy is removed, the child may be likely to engage in SIB. In this situation, we would say that SIB is maintained by access to tangible items or that the behavior serves a tangible function.

Assessment of Self-Injurious Behavior

Functional behavioral assessments are a category of procedures that are conducted to identify the function of challenging behavior or what is maintaining those behaviors. Functional behavioral assessments are standard practice when treating self-injurious behavior (Emerson, 1992). In fact, they are required by law in public school systems for children with special needs (including ASD) who engage in challenging behaviors such as self-injury (Drasgow & Yell, 2001; Gresham, Watson, & Skinner, 2001). A description of functional behavioral assessments can be found in Chap. 25 of this book. However, there are special considerations when assessing SIB. Specifically, we will discuss the experimental FA, which is the most intensive and experimentally rigorous method of identifying the function of challenging behavior (Vollmer, Marcus, Ringdahl, & Roane, 1995), as it relates specifically to the assessment of SIB.

Overview of Functional Analyses for Self-Injurious Behavior

An approach to analyzing the behavior-environment interaction and identifying the function of challenging behavior is known as a functional analysis (FA). This approach, described by Iwata et al. (1982/1994), has become a standard assessment used to identify the function of challenging behaviors such as SIB. In this assessment, the environmental antecedents and associated consequences are manipulated in isolation in a series of test conditions (e.g., attention, tangible, demand). For example, in an attention test condition, the therapist's attention is diverted at the start of the session, and the therapist provides a brief reprimand following SIB. This condition is analogous to one in which caregivers have to attend to something other than their child but return their attention to their child in the event of SIB in an attempt to calm him/her. In an escape test condition, the therapist provides demands and gives the child a short break following SIB. This condition is analogous to the brushing of teeth example mentioned earlier. The caregiver requests their child to brush his teeth, but then following SIB, the task is terminated and the child is allowed to escape the demand. Test conditions are designated to evaluate if SIB is maintained by positive reinforcement (access to attention or preferred items), negative reinforcement (escape from demands), or automatic reinforcement (meaning the behavior persists in the absence of external social reinforcement).

Professionals directly observe and record instances of SIB and compare rates of the behavior in the test to a control condition where the child has free access to attention and toys and no demands are made. Elevated rates of SIB in one or more test condition(s) compared to the control condition provide evidence that the variable tested in that condition maintains SIB. Beavers et al. (2013) reviewed published FAs across a 30-year period, and SIB was assessed in 55% of the reviewed FAs. SIB was found to be maintained by attention for 21%, maintained by access to preferred tangible items for 11%, and maintained by escape from demands or other aversive stimuli for 27% of participants. In addition, 9% of the studies concluded SIB was multiply maintained (i.e., more than one function).

Special Considerations for Severe Self-Injurious Behavior

In FAs, there are often special considerations when evaluating SIB compared to other topographies. As a result, we focus this discussion on these modifications as they specifically relate to SIB. Depending on the level of intensity, SIB can pose a significant risk to an individual's safety. For example, for an individual who engages in severe eye-gouging, a single instance of SIB could lead to permanent loss of vision. Similarly, in cases of extreme head banging, one head hit to a hard surface could result in a concussion. To assess the variables that maintain SIB using a traditional FA, the behavior must be allowed to occur multiple times. It is important to note that in a recent review of 99 participants who completed FAs for severe challenging behavior in an inpatient setting, injuries during assessment were very low, and overall the assessment was deemed safe so long as appropriate precautions were taken (Kahng et al., 2015). As a result, clinicians should *not* assume that severe SIB precludes the use of FAs. Instead, modifications and safety measures should be considered and incorporated into the procedures.

One approach to mitigating risk of harm during the FA is to develop clear termination criteria. For example, a clinician may decide to stop any session in which 20 instances of SIB occur. Alternatively, the clinician may elect to run short sessions (e.g., 3–5 min sessions as opposed to 10 min which is a common practice). These criteria should be set in conjunction with a medical professional who can assist in identifying the number of instances of behavior that can occur in a short period of time without the risk of injury.

If a single incident of SIB poses a risk of permanent damage, additional precautions should be considered. For example, protective equipment (Dorsey, Iwata, Reid, & Davis, 1982; Moore, Fisher, & Pennington, 2004) may be used during the assessment. A helmet may be worn for an individual who engages in head banging or arm splints or padded gloves for an individual who hits themselves or engages in self-scratching. When the setting allows, therapists may also consider padding walls or hard surfaces to prevent the impact of topographies of SIB that include forceful contact with these kinds of surfaces.

Although the potential of increased safety may make the use of protective equipment appealing, it is important to consider that these devices may interfere with identifying some functions (e.g., Borrero, Vollmer, Wright, Lerman, & Kelley, 2002). For example, it is important that SIB still persists at some level that makes assessment feasible. If the protective equipment was to eliminate SIB completely, then a function of SIB will not be able to be concluded using this assessment. The use of protective equipment may also eliminate SIB that occurs to access automatic reinforcement in the absence of the protective equipment. For example, if a child engages in hand-to-head hitting because the behavior itself automatically produces reinforcement, it is possible that this reinforcement would be eliminated by the addition of a helmet. As a result, you may get low rates of problem behavior in an alone or ignore condition of the FA when the helmet is on. This creates the risk of ruling out an automatic function when in fact the behavior is maintained by automatic reinforcement without the helmet (i.e., a false negative for an automatic function). As such, the potential impact of the protective equipment on problem behavior should always be considered when interpreting results of the FA.

An additional modification is to focus the FA on less dangerous behaviors that are in the same response class as more intense SIB (Borrero & Borrero, 2008). Often, when individuals engage in severe SIB, there are behaviors that commonly precede the SIB that serve the same function. Fritz, Iwata, Hammond, and Bloom (2013) describe a systematic method for identifying these precursor behaviors while observing a minimal number of targeted dangerous behaviors. First, clinicians can interview client's caregivers and conduct unstructured observation to identify hypotheses about behaviors that are likely to precede SIB and serve the same function. Next, these hypothesized precursor behaviors and the more severe behaviors are measured during brief exposure to antecedents that are likely to evoke problem behavior (e.g., removal of preferred items, placing nonpreferred demands). Probability analyses can then be used to compare the occurrence of potential precursor behaviors and target behaviors. In the probability analyses, the clinician identifies the chance of various events occurring given the occurrence of other events. For example, if the primary target is severe SIB and the hypothesized precursor is screaming, the clinician would identify the probability of severe SIB occurring when screaming has occurred. The inverse would also be calculated, determining the probability of screaming during times when severe SIB has occurred. The absence of these behaviors (e.g., the probability of severe SIB occurring in the absence of screaming) is also calculated. Comparing these calculations allows the clinician to identify the likelihood that screaming does in fact commonly precede and serve the same function as SIB.

After identifying precursor behaviors, the function of these less dangerous behaviors can then be identified in subsequent FAs. Assuming the precursor behaviors generally precede SIB, reinforcing these behaviors in the FA should limit the amount of more severe SIB that occurs in assessment. Thus, safety can be maintained, and the function of the targeted topographies of SIB can still be identified, so long as the precursors serve the same function.

Another method of increasing safety for severe SIB in FAs is to only allow a single instance to occur in each session. Typically, multiple instances of the targeted behavior are allowed to occur, and it is recorded in terms of its rate, duration, or the percentage of intervals in which it occurred. However if the target behavior poses a significant risk, as in the case of high-intensity SIB, the latency to a single instance of the severe behavior may serve as an alternative measure that does not require repetition of the behavior. In conducting a latency-based FA, the clinician presents various potential events that may evoke SIB (establishing operations) and measure how long it takes from the onset of this event until the time that SIB occurs (Thomason-Sassi, Iwata, Neidert, & Roscoe, 2011). After SIB occurs, the hypothesized functional reinforcer is delivered, and the session is terminated, meaning that only one instance of the severe behavior is expected to occur in each session.

The use of shorter sessions and protective equipment, assessment of precursor behavior(s), and evaluating latency to SIB are possible modifications to consider when conducting FAs for individuals who engage in SIB. However, in addition to considering these modifications to the standard FA (Iwata et al., 1982/1994), practicing professionals need to ensure proper oversight throughout the assessment process to safeguard the individual from potential injuries. If redness, bruising, or other sign of injury is observed during assessment, it is best to air on the side of caution and terminate the assessment to ensure safety until a new assessment plan can be created. Whenever possible, a medical professional should be consulted regarding the potential impact and injury from SIB. In addition, for severe cases of SIB where the risk of injury is high, it is best to consult with a behavior analyst who specializes in the assessment and treatment of severe challenging behavior or refer to a treatment facility that has the proper facilities and expertise.

Assessment of Automatically Maintained Self-Injurious Behavior. SIB is more likely to be maintained by automatic reinforcement than other topographies of problem behavior commonly exhibited by individuals with IDD. In a review of published FAs, Beavers et al. (2013) found that 25% of cases of SIB were maintained by automatic reinforcement, whereas aggressive and destructive behaviors were maintained by automatic reinforcement for only 2% and 11% of cases, respectively. As such, a further discussion of problem behavior maintained by automatic reinforcement is warranted.

As described above, problem behavior maintained by automatic reinforcement is identified through continued occurrence of the behavior in the absence of social consequences. By ruling out the influence of social consequences, it is assumed that the behavior produces the reinforcement directly. Unfortunately, it is often difficult to identify the specific form of automatic reinforcement that SIB is producing. For instance, SIB might be maintained by positive automatic reinforcement (the behavior is producing a preferred internal experience), negative automatic reinforcement (the behavior is reducing an aversive internal experience), or some combination of the two. Vollmer (1994) suggests an example where an individual who engages in self-scratching to the point of injury may do so because the behavior produces temporary alleviation of an uncomfortable "itchy" sensation or automatic negative reinforcement. For other individuals, SIB may result in the release of endorphins or other physiological effects that are desired and serve as positive reinforcement for the behavior. Note that both of these examples and several of the examples described below directly relate to and may be better explained by the sensory and neurobiological models of SIB. However, they are being discussed here in the context of research on assessment techniques for SIB that has been primarily focused on in the behavioral literature.

There have been several examples in the literature in which researchers have identified the specific type of automatic reinforcement produced by challenging behavior. For example, Piazza, Adelinis, Hanley, Goh, and Delia (2000) conducted competing item assessments with individuals with automatically maintained challenging behavior. This assessment included items that produced stimulation that either matched or did not match the type of automatic reinforcement hypothesized to be produced by the participants' topography of challenging behavior. For example, one participant engaged in problematic hand mouthing that was automatically maintained. The researchers hypothesized that the automatic reinforcement was the result of either stimulation to the hand or oral stimulation. Therefore, his matched stimuli included items that produced similar stimulation (e.g., hand massager for hand stimulation or a mouth guard for oral stimulation). They found that access to matched items was more likely to result in low rates of challenging behavior when compared to items that were not matched to the hypothesized automatic reinforcement produced by the behavior. This finding was replicated across three different participants with automatically maintained challenging behavior, supporting the authors' hypotheses about the specific mechanism of automatic reinforcement maintaining behavior for those specific clients.

Another method of testing for the specific type of automatic reinforcement produced by SIB involves the use of protective equipment or other procedures that block a specific sensory consequence of the behavior (McKerchar, Kahng, Casioppo, & Wilson, 2001). For example, head banging may be maintained by several sensory inputs including the visual stimulation produced by movement of the head, the sound the head makes when it hits a surface, or the impact on the head. One can test the hypothesis that impacts to the head are producing automatic reinforcement by having the individual wear a helmet. Under this condition, any potential visual or auditory stimulation remains, but impacts to the head are attenuated. If SIB decreases when the helmet is worn, one can infer the behavior was maintained by the impact and not auditory or visual input.

Further complicating the issue of automatically maintained SIB is the potential for it to be multiply maintained by both automatic and social reinforcement (McKerchar et al., 2001; Scheithauer, Lomas Mevers, Call, & Shrewsbury, 2017; Smith, Iwata, Vollmer, & Zarcone, 1993). Assessing for multiply maintained SIB is challenging but is often important in guiding treatment. For example, consider a child who engages in SIB that is maintained by both automatic reinforcement and escape from demands. First-line treatments for the automatically maintained SIB are likely to include access to other preferred items that may compete with the automatic reinforcement or delivery of alternative reinforcers for the absence of SIB. Neither of these treatments is likely to resolve escape-maintained SIB. Thus, it is important for clinicians to determine whether automatically maintained SIB is also maintained by social consequences.

There are a handful of examples of this type of assessment in the literature (Contrucci Kuhn & Triggs, 2009; McKerchar et al., 2001; Scheithauer et al., 2017; Smith et al., 1993). The basic concept behind these assessments is that they control for the automatic reinforcement that is produced by the challenging behavior by using protective equipment (e.g., Scheithauer et al.) or providing preferred items that are known to compete with the automatic reinforcement (e.g., Smith et al.). Social test conditions are then repeated with these procedures in place. If problem behavior persists in test conditions designed to evaluate the role of social reinforcement after SIB maintained by automatic reinforcement has been reduced, then the behavior is likely also maintained by the associated social reinforcers.

Another issue further complicating the assessment and treatment of automatically maintained SIB is the possibility of self-restraint (Fisher & Iwata, 1996). According to a study of individuals with IDD admitted to a treatment facility for severe SIB, 21% of participants also engaged in self-restraint during assessment (Hagopian, Rooker, & Zarcone, 2015). Self-restraint occurs when an individual restricts his or her own movement through topographies such as sitting on the hands, wrapping hands in clothing, wrapping limbs around another person, or actively seeking out protective equipment such as arm restraints. When individuals present with severe SIB and self-restraint, it is important that the relationship between the two behaviors is thoroughly assessed, as self-restraint can often create difficulties in interpreting assessment and treatment results that are targeting SIB. There are several potential relationships between these behaviors (Fisher & Iwata, 1996). First, self-restraint may serve the same function as SIB, whether by producing automatic (Derby, Fisher, & Piazza, 1996) or social reinforcement (Rapp & Miltenberger, 2000). Second, SIB may be maintained by access to self-restraint (Rooker & Roscoe, 2005). For example, a teacher may prevent a child from engaging in self-restraint that interferes with classwork. However, if the child begins to engage in SIB, the teacher may allow self-restraint because it is the less dangerous of the two behaviors. Finally, self-restraint may be maintained by negative reinforcement in the form of discontinuation of SIB (Scheithauer et al., 2015; Smith, Iwata, Vollmer, & Pace, 1992). In this scenario, it is assumed that SIB actually produces aversive consequences. Self-restraint then occurs as it has historically resulted in avoiding or stopping SIB and the related aversive consequences. This can be an especially complicated explanation because if avoiding SIB is preferred, then one would expect the individual to simply refrain from engaging in SIB. However, in this scenario, it is possible that the SIB is not under the control of operant contingencies. Instead, the behavior may be the result of some physiological process that is not under the immediate control of the individual (e.g., tic disorders). It is also possible that the SIB produces some form of immediate positive reinforcement (e.g., sensory stimulation) followed by a later aversive consequence (e.g., pain). Thus, the self-restraint may serve as a self-control response to prevent the individual from engaging in an impulsive behavior that will result in immediate reinforcement but long-term aversive consequences.

In summary, SIB maintained by automatic reinforcement is difficult to assess and treat for numerous reasons. Most of the research in this field has relied on single-subject methods conducted in highly specialized settings, likely due to the intensity of assessment and intervention that is often necessary and the complexity of individual cases. Additional work is needed to identify best-practice methodologies that can be more widely disseminated across clinicians and be used in less specialized clinics.

Treatment Models for Self-Injurious Behavior

Pharmacological Treatments

Few drug studies have specifically targeted SIB in individuals with IDD (Gormez, Rana, & Varghese, 2014; Minshawi, Hurwitz, Morriss, & McDougle, 2015), and those that do directly assess SIB have generally relied on small-N designs. However, there is a larger body of research that targets behaviors related to SIB in this population, such as repetitive behaviors, irritability, and disruptive behavior. A summary of this literature is beyond the scope of this chapter: see Doyle and McDougle (2012) and Fung et al. (2016) for reviews of these topics. Instead, we focus on the most commonly studied drugs that target SIB specifically.

Antipsychotic drugs, which act as dopamine antagonists, are one of the oldest classes of drugs used to target maladaptive behaviors in individuals with IDD. Haloperidol, a typical antipsychotic, was one of the earliest drugs used to target SIB and related behaviors (e.g., Burk & Menolascino, 1968). Whereas reductions in maladaptive behaviors, including SIB, were noted, these drugs are not widely used as a first-line treatment today due to serious side effects, including dyskinesias (McDougle, Stigler, Erickson, & Posey, 2006; McPheeters et al., 2011). Instead, atypical antipsychotics are commonly used for individuals who engage in SIB. Two atypical antipsychotics (risperidone and aripiprazole) are approved drugs for use with ASD, based on randomized clinical trials demonstrating improvement in problem behavior broadly (Marcus et al., 2009; McCracken et al., 2002; Owen et al., 2009; Shea et al., 2004). Risperidone has also been shown to improve caregiver ratings of SIB specifically (Aman et al., 2002). A few studies have also directly targeted

SIB, with direct observation as the primary outcome measure with mixed results (Cox & Virues-Ortega, 2016).

The opiate blocker naltrexone is another drug studied in the treatment of SIB and is somewhat unique in that the primary target of those studies has been SIB specifically, as opposed to problem behavior more broadly. Sandman (1988) conducted a study analyzing the effects of naltrexone (an opiate blocker), on SIB with four individuals with IDD. Self-injury improved for three of the four, lending support for the neurobiological model describing the role of increased endorphin release in SIB. Additional support emerges from findings that individuals with higher levels of endorphins following SIB are more likely to respond to opiate blockers as treatment (Sandman, Hetrick, Taylor, & Chicz-DeMet, 1997; Scifo et al., 1996). Other theories suggest the mechanism of action for these drugs is lowering an individual's pain threshold, resulting in increased pain from the SIB and subsequent reductions. The positive effects of naltrexone have been replicated (e.g., Bernstein, Hughes, Mitchell, & Thompson, 1987; Bodfish et al., 1997; Gibson, Hetrick, Taylor, Sandman, & Touchette, 1995; Kars, Broekema, Glaudemans-van Gelderen, Verhoeven, & Van Ree, 1990); however, other studies have failed to replicate the positive results, finding that naltrexone does not result in significant improvements over placebos (Willemsen-Swinkels, Buitelaar, Nijhof, & van Engeland, 1995; Zingarelli et al., 1992). In a recent review of clinical trials evaluating opiate blockers, six out of ten published clinical trials identified statistically significant improvements in SIB (Roy, Roy, Deb, Unwin, & Roy, 2015). Unfortunately, there are currently no indications as to the characteristics that might predict a positive response. As a result, additional research is needed.

Medications targeting 5-HT have also been studied as a treatment for SIB. Open-label trials have suggested clomipramine, a tricyclic antidepressant (TCA), may be effective at reducing SIB (e.g., Garber, McGonigle, Slomka, & Monteverde, 1992). Selective serotonin reuptake inhibitors (SSRIs) are also now commonly prescribed. Fluoxetine is the most common SSRI studied as a treatment for SIB, with open-label trials indicating effectiveness in decreasing SIB in individuals with IDD (Bass & Beltis, 1991; Markowitz, 1992; Ricketts et al., 1993). Fluoxetine has also been shown to improve skin picking in a small randomized clinical trial (Simeon et al., 1997). Paroxetine, sertraline, and fluvoxamine have also been studied using retrospective reviews and open-labeled studies targeting SIB. Reviews of this literature have been mixed, with some suggesting the drugs may be effective (Janowsky, Shetty, Barnhill, Elamir, & Davis, 2005) and others suggesting there is insufficient evidence (Sohanpal et al., 2007). In summary, although medications targeting 5-HT have been studied, additional large-N clinical trials are needed to definitively determine efficacy.

N-Acetylcysteine (NAC), an over-the-counter dietary supplement, has also been shown to be effective with SIB when given at higher doses than is suggested as a supplement. NAC acts through enhancing antioxidant activity and reducing release of glutamate. NAC has shown promising results in open-label case studies (Marler, Sanders, & Veenstra-VanderWeele, 2014; Percinel & Yazici, 2014) and a small clinical trial targeting skin picking (Grant et al., 2016).

Another class of medications that has received attention in research is antiepileptic or anticonvulsant drugs, operating through stimulating GABA receptors. One of these medications, divalproex sodium (i.e., Depakote), was evaluated in an open-label trial with adults with ID and SIB with promising results (Ruedrich, Swales, Fossaceca, Toliver, & Rutkowski, 1999). However, a randomized clinical trial with a youth population studying valproate found no significant effect on indirect measures that are often related to SIB (such as irritability; Hellings et al., 2005). Therefore, additional research is needed to determine whether this class of medications should be indicated for SIB. There are also several other drugs not discussed here that are commonly prescribed for off-label use with this population, including anxiolytics, stimulants, and mood stabilizers (Tsiouris, Kim, Brown, Pettinger, & Cohen, 2013). Similar to anticonvulsant drugs, these need additional research before becoming clinically indicated for the treatment of SIB.

Sensory Treatments

There is some variability in the literature related to defining sensory approaches to the treatment of SIB (Ottenbacher, 1991). Here we discuss some of the documented cases of effective interventions in the literature that describe their approach in terms that relate to sensory input (Bright, Bittick, & Fleeman, 1981; Dura, Mulick, & Hammer, 1988; Favell, McGimsey, & Schell, 1982; Wells & Smith, 1983). Sensory integration is a commonly used approach among occupational therapists. Sensory integration is based on the concept that there is a breakdown in how sensory input is organized and analyzed, leading to self-stimulatory behavior such as SIB (Ayres, 1972). Sensory integration therapy for the treatment of SIB hypothesizes that including stimuli that satisfy the sensory dysfunction (i.e., the breakdown in organization and analysis of sensory input) will organize sensory processes and therefore decrease SIB. For example, Bright and colleagues included 1 h sessions once per day in which the individual was exposed to noncontingent vestibular, tactile, and auditory stimulation. This type of stimulation was included after observation that SIB occurred less frequently when tactile and vestibular stimulations were provided. The frequency of SIB decreased across the 5 months of data collection.

Other researchers have found mixed results for the use of sensory integration in the treatment of SIB (Arendt, MacLean, & Baumeister, 1988; Kinnealey, 2005; Mason & Iwata, 1990). Kinnealey found that SIB improved for the 1 h immediately following sensory integration, but the improvement did not maintain outside of this hour. Similarly, Mason and Iwata found that sensory integration therapy with attention (i.e., the therapist provided high-quality attention to the client during therapy) successfully reduced the frequency of SIB during therapy sessions. However, when the attention component was removed, the sensory integration sessions alone did not affect the frequency of SIB. These disparate findings may be related to the various factors commonly included in therapy. Specifically, during sensory integration sessions, there are typically no demands present, and attention is provided at least intermittently. These factors alone may contribute to decreases in SIB observed during the therapy sessions (Mason & Iwata) but may not create generalized improvement outside of treatment sessions. As such, discrepancies in the literature on the success of sensory integration therapy may stem from when observations are conducted (e.g., during appointments compared to a period of time after completion of the appointment) and the types of components included in the therapy.

Behavioral Treatments

The literature is rife with examples of behavioral treatments for SIB (Kahng, Iwata, & Lewin, 2002). Extinction, or the disruption of the relationship between SIB and the functional reinforcer, is a common treatment component. In cases of SIB, extinction consists of first identifying the functional reinforcer through behavioral assessments (see above and Chap. 25). After identification, the simplest way to disrupt the relationship between the functional reinforcer and SIB is to no longer provide the reinforcer when SIB occurs. For example, if SIB is maintained by escape from demands, then extinction could consist of following through with demands despite the occurrence of SIB. In the example discussed above with brushing teeth, this could involve hand-over-hand prompting to ensure the child completes teeth brushing. Conversely, if SIB is found to be maintained by access to attention, then extinction could involve ignoring all instances of SIB.

Extinction alone can effectively reduce SIB (Goh & Iwata, 1994), but greater and more rapid reductions occur when used in conjunction with a reinforcement component (Mazaleski, Iwata, Vollmer, Zarcone, & Smith, 1993; McCord, Thomson, & Iwata, 2001). Thus, it is common to incorporate reinforcement-based procedures when treating SIB. Kahng et al. (2002) found that more than 40% of 700 cases reporting treatments of SIB in the literature included a reinforcement component. There are several different methods by which reinforcement can be included in the treatment. First, differential reinforcement of alternative behavior (DRA) is commonly used and is implemented by identifying an alternative and appropriate behavior that results in access to the functional reinforcer (Vollmer & Iwata, 1992). For SIB maintained by access to escape, DRA could include providing a break contingent on compliance with demands (Roberts, Mace, & Daggett, 1995). Similarly, for SIB maintained by access to attention or tangible items, DRA treatments have been successful that target a functionally equivalent communication response, such as a request for the relevant reinforcer (Fisher, Kuhn, & Thompson, 1998; Hanley, Iwata, & Thompson, 2001). For problem behavior maintained by access to attention, this treatment could involve teaching the child to ask for attention and providing reinforcement when he or she does so.

Another form of reinforcement, differential reinforcement of other behavior (DRO), consists of providing reinforcement following a period of time without SIB. When SIB is maintained by positive or negative social reinforcement, the functional reinforcer is generally delivered when the required time interval without SIB has occurred. For example, if SIB is maintained by access to attention, a therapist may provide high-quality attention when the child has gone a specific amount of time without engaging in SIB (Mazaleski et al., 1993; Mazaleski, Iwata, Rodgers, Vollmer, & Iwata, 1992; Vollmer, Iwata, Zarcone, Smith, & Mazaleski, 1993). Other times, an arbitrary reinforcer (e.g., a preferred item that is not the functional reinforcer) may be provided, especially if the behavior is automatically maintained, because the functional reinforcer is difficult to deliver under such circumstances (Cowdery, Iwata, & Pace, 1990).

Noncontingent access to preferred items or functional reinforcers is another common strategy incorporated into treatments for SIB. This treatment technique can operate through decreasing an individual's motivation to access a functional reinforcer. In addition, when the functional reinforcer is being delivered noncontingently, it is no longer being provided contingent on SIB, which disrupts this relationship through extinction. For example, if SIB is maintained by access to escape, providing noncontingent breaks on a specific time schedule might decrease the individual's motivation to engage in SIB to get that same break while also implementing extinction as the break is no longer being provided contingent on SIB (Vollmer, Marcus, & Ringhdahl et al., 1995).

For automatically maintained SIB, providing noncontingent access to preferred items or attention may consist of creating an enriched environment that competes with the automatic reinforcement produced by SIB, subsequently decreasing motivation to engage in SIB (Shore, Iwata, DeLeon, Kahng, & Smith, 1997). To identify items that may sufficiently compete with automatic reinforcement and decrease SIB, a structured assessment can be conducted prior to starting treatment. This assessment, called a competing items assessment, consists of exposing the individual to various conditions in which a variety of potentially preferred items are freely available. Clinicians can then record item interaction and SIB to identify items likely to compete with SIB (Piazza et al., 1999; Piazza et al., 2000).

The strategies of differential reinforcement and noncontingent access to preferred items described above are often first-line treatments as they are minimally intrusive to the individual undergoing treatment. However, in some situations, reinforcement strategies alone are not sufficient to reduce SIB. For example, in the case of automatically maintained SIB, it may not always be possible to identify reinforcers that sufficiently compete with the functional reinforcer. In this situation, there are other strategies that can be considered.

Response interruption and redirection is one of these strategies that has resulted in decreases in SIB (Turner, Realon, Irvin, & Robinson, 1996). In this treatment, a clinician interrupts SIB and redirects the individual to emit an appropriate response, such as completion of demands or engaging in appropriate play. The effectiveness of this intervention may be the result of punishing the SIB by redirecting to a less-preferred task or interrupting a pattern of behavior that has historically resulted in automatic reinforcement (creating sensory extinction for additional instances of the behavior).

Response blocking of SIB is also a common component included in the treatment. Response blocking involves physically preventing the behavior from occurring. For example, for SIB that involves hitting, the therapist would place a physical barrier (e.g., therapist's hand or a soft surface) between the client's hand and the targeted area of the body to prevent the hitting from occurring. While effective, similar to response interruption redirection, there is debate on whether the mechanism responsible for reductions in SIB from response blocking is attributed to sensory extinction (Reid, Parsons, Phillips, & Green, 1993; Smith, Russo, & Le, 1999) or punishment (assuming that the blocking procedure is aversive; Lerman & Iwata, 1996).

Sensory extinction can also be implemented through the use of protective equipment used to eliminate or decrease the reinforcement or stimulation produced by engaging in SIB (Rincover & Devany, 1982; Roscoe, Iwata, & Goh, 1998). These techniques are only likely to be successful if the automatic reinforcement is sufficiently reduced by application of the protective equipment. Some protective equipment used in the literature may also operate through increasing the effort that it takes to engage in the behavior. For example, mechanical arm restraints that make it more effortful for the client to bend his or her arm may make it more difficult to engage in hand-to-head SIB (Deshais, Fisher, Hausman, & Kahng, 2015). As a result the response effort may be too high for the amount of automatic reinforcement produced by the behavior, subsequently reducing the frequency of the behavior (Irvin, Thompson, Turner, & Williams, 1998). It is important to note that when using protective equipment or mechanical restraint, it is important to consult with individuals with expertise in the use of these procedures for behavioral treatments and with medical professionals to ensure safety and proper care. For example, in using arm restraints, it is important to follow strict protocols for application, removal, and frequency of breaks to avoid muscle atrophy. In addition, the ultimate goal of treatment is generally to reduce and fade out the use of protective equipment so as to not use them as a long-term solution. Misuse of these technologies can result in physical damage if proper case is not taken.

Finally, when the methods described have been exhausted and are unsuccessful, and SIB is severe enough to cause significant harm, then it may be appropriate to consider the use of punishment procedures. These are events that are delivered following SIB that are nonpreferred and therefore likely to decrease future instances of SIB. Systematic assessments can be conducted prior to treatment to identify potential punishers (Fisher et al., 1994), with common procedures being blocking the individual's line of site, restraint, and time-out (Kahng et al., 2002). It should be noted that punishment procedures should only be implemented with highly trained clinicians in situations where other treatments have proven ineffective and the behavior is serious enough to warrant the invasiveness of the procedure. In addition, it is crucial that the effectiveness of the procedure is systematically analyzed to ensure that any punishment procedures used are effective and necessary.

Generalization and caregiver training are crucial components of all behavioral treatments for SIB. Although the topic of generalization is outside the scope of this chapter (see Stokes & Baer, 1977), clinicians should take care to ensure that treatments for SIB generalize to naturally occurring situations, environments, and caregivers. Caregiver training is an equally important component of treatments for SIB. Clinicians should consider robust training practices, including behavioral skills training followed by frequent checks for integrity. Research has shown that decreased fidelity negatively impacts treatment effectiveness (Fryling, Wallace, & Yassine, 2012).

Nonevidence-Based Treatments

Families of children with IDD such as ASD are increasingly seeking out complimentary, alternative treatments that have not been supported in the literature (Brondino et al., 2015). Some of these interventions are touted as treatments for challenging behavior including SIB. For example, individuals have made claims that the use of cannabis is effective in the treatment of SIB. However, these claims have been limited to anecdotes and small case studies (Kruger & Christophersen, 2006). Other studies have focused on the use of music (Lundqvist, Andersson, & Viding, 2009) or yoga (Rosenblatt et al., 2011) to reduce SIB. However, the scarcity of data limits the validity of findings pertaining to the use of these interventions.

Clinicians should focus on the use of strategies that meet evidence-based standards and should discuss with caregivers the possible limitations or harmful effects of nonevidence-based treatments. Broadly, evidence-based strategies involve interventions that have been systematically evaluated using randomized group trials or well-controlled single-subject designs that suggest the procedures are effective. Specific criteria have been proposed to identify an intervention as well established and probably efficacious within the field of psychology (Tolin, McKay, Forman, Klonsky, & Thombs, 2015). Due to the low prevalence of extremely severe cases of SIB and the intensity of assessment and treatments necessary for these cases, it is possible that some commonly used treatments described above may fall short of these proposed criteria. Nonetheless, the criteria serve as a guide for clinicians for identifying the type of studies that should be used to support the use of specific interventions.

Multidisciplinary Approach

In studying and treating SIB, it is important to consider a multidisciplinary approach whenever possible. In the development of SIB, it is likely that a variety of environmental, sensory, genetic, and physiological factors contribute to the emergence of SIB. For example, a child may engage in SIB that is escape maintained when presented with schoolwork (explained by the behavioral model). However, the child may engage in higher rates of SIB when presented with schoolwork during times that the child is also not feeling well, which is best explained by the sensory or neurobiological models. There are several examples in the literature of these situations where environmental and physiological factors act in conjunction to impact rates of SIB (e.g., Kennedy & Meyer, 1996). In this situation, one theory alone is not sufficient to adequately explain the child's SIB, and a combination of theories is necessary. Similarly, in treatment, it is common that children with IDD who engage in SIB will be undergoing multiple modalities of treatment. In these scenarios, it is important to assess the impact of each component of the child's treatment and to collaborate across providers. As such, techniques that consider multiple potential variables that may impact SIB will allow for the most comprehensive assessment and treatments. Especially in the case of severe self-injury, it is recommended that professionals work across disciplines to evaluate medical, sensory, and behavioral causes and potential treatment components (Minshawi et al., 2015).

References

Adamsen, D., Ramaekers, V., Ho, H. T. B., Britschgi, C., Rufenacht, V., Meili, D., … Thony, B. (2014). Autism spectrum disorder associated with low serotonin in CSF and mutations in the SLC29A4 plasma membrane monoamine transporter (PMAT) gene. *Molecular Autism, 5*. https://doi.org/10.1186/2040-2392-5-43

Allen, D. G., Lowe, K., Moore, K., & Brophy, S. (2007). Predictors, costs and characteristics of out of area placement for people with intellectual disability and challenging behaviour. *Journal of Intellectual Disability Research, 51*(6), 409–416.

Aman, M. G., De Smedt, G., Derivan, A., Lyons, B., Findling, R. L., & Risperidone Disruptive Behavior Study Group. (2002). Double-blind, placebo-controlled study of risperidone for the treatment of disruptive behaviors in children with subaverage intelligence. *American Journal of Psychiatry, 159*(8), 1337–1346.

Anderson, L. T., & Ernst, M. (1994). Self-injury in Lesch-Nyhan disease. *Journal of Autism and Developmental Disorders, 24*(1), 67–81.

Arendt, R. E., MacLean, W. E., & Baumeister, A. A. (1988). Critique of sensory integration therapy and its application in mental retardation. *American Journal on Mental Retardation, 92*, 401–411.

Arron, K., Oliver, C., Berg, K., Moss, J., & Burbidge, C. (2011). Prevalence and phenomenology of self-injurious and aggressive behavior in genetic syndromes. *Journal of Intellectual Disability Research, 55*, 109–120. https://doi.org/10.1111/j.1365-2788.2010.01337.x

Ayres, A. J. (1972). *Sensory integration and learning disorders*. Western Psychological Services: Los Angeles.

Baghdadli, A., Pascal, C., Grisi, S., & Aussilloux, C. (2003). Risk factors for self-injurious behaviours among 222 young children with autistic disorders. *Journal of Intellectual Disability Research, 47*(8), 622–627.

Baghdadli, A., Picot, M. C., Pry, R., Michelon, C., Burzstejn, C., Lazartigues, A., & Aussilloux, C. (2008). What factors are related to a negative outcome of self-injurious behaviour during childhood in pervasive developmental disorders? *Journal of Applied Research in Intellectual Disabilities, 21*(2), 142–149.

Barrera, F. J., Violo, R. A., & Graver, E. E. (2007). On the form and function of severe self-injurious behavior. *Behavioral Interventions, 22*(1), 5–33.

Bass, J. N., & Beltis, J. (1991). Therapeutic effect of fluoxetine on naltrexone-resistant self-injurious behavior in an adolescent with mental retardation. *Journal of Child and Adolescent Psychopharmacology, 1*(5), 331–340. https://doi.org/10.1089/cap.1991.1.331

Beavers, G. A., Iwata, B. A., & Lerman, D. C. (2013). Thirty years of research on the functional analysis of problem behavior. *Journal of Applied Behavior Analysis, 46*(1), 1–21. https://doi.org/10.1002/jaba.30

Bernstein, G. A., Hughes, J. R., Mitchell, J. E., & Thompson, T. (1987). Effects of narcotic antagonist on self-injurious behavior: A single case study. *American Academy of Child and Adolescent Psychiatry, 26*, 886–889.

Bodfish, J. W., McCuller, W. R., Madison, J. M., Register, M., Mailman, R. B., & Lewis, M. H. (1997). Placebo, double-blind evaluation of long-term naltrexone treatment effects for adults with mental retardation and self-injury. *Journal of Developmental and Physical Disabilities, 9*(2), 135–152.

Borrero, J. C., Vollmer, T. R., Wright, C. S., Lerman, D. C., & Kelley, M. E. (2002). Further evaluation of the role of protective equipment in the functional analysis of self-injurious behavior. *Journal of Applied Behavior Analysis, 35*(1), 69–72. https://doi.org/10.1901/jaba.2002.35-69

Borrero, S. W., & Borrero, J. C. (2008). Descriptive and experimental analyses of potential precursors to problem behavior. *Journal of Applied Behavior Analysis, 41*, 83–96.

Borthwick, S. A., Meyers, C. E., & Eyman, R. K. (1981). Comparative adaptive and maladaptive behavior of mentally retarded clients of five residential settings in three western states. In R. H. Bruininks, C. E. Meyers, B. B. Sigford, & K. C. Larkin (Eds.), *Deinstitutionalization and community adjustment of mentally retarded people* (pp. 351–359). Washington, DC: American Association on Mental Deficiency.

Bosch, J., Van Dyke, C., Smith, S. M., & Poulton, S. (1997). Role of medical conditions in the exacerbation of self-injurious behavior: An exploratory study. *Mental Retardation, 35*(2), 124–130. https://doi.org/10.1352/0047-6765(1997)035<0124:ROMCIT>2.0.CO;2

Bright, T., Bittick, K., & Fleeman, B. (1981). Reduction of self-injurious behavior using sensory integrative techniques. *The American Journal of Occupational Therapy, 35*(3), 167–172.

Brondino, N., Fusar-Poli, L., Rocchetti, M., Provenzani, U., Barale, F., & Politi, P. (2015). Complementary and alternative therapies for autism spectrum disorder. *Evidence Based Complementary and Alternative Medicine, 2015*, 1. https://doi.org/10.1155/2015/258589

Burk, H. W., & Menolascino, F. J. (1968). Haloperidol in emotionally disturbed mentally retarded individuals. *American Journal of Psychiatry, 124*(11), 1589–1591.

Carr, E. G. (1977). The motivation of self-injurious behavior: A review of some hypotheses. *Psychological Bulletin, 84*(4), 800–816.

Carr, E. G., & Smith, C. E. (1995). Biological setting events for self-injury. *Mental Retardation and Developmental Disabilities Research Reviews, 1*, 94–98. https://doi.org/10.1002/mrdd.1410010204

Contrucci Kuhn, S. A., & Triggs, M. (2009). Analysis of social variables when an initial functional analysis indicates automatic reinforcement as the maintaining variable for self-injurious behavior. *Journal of Applied Behavior Analysis, 42*, 679–683. https://doi.org/10.1901/jaba.2009.42-679

Cooper, S. A., Smiley, E., Allan, L. M., Jackson, A., Finlayson, J., Mantry, D., & Morrison, J. (2009). Adults with intellectual disabilities: Prevalence, incidence and remission of self-injurious behaviour, and related factors. *Journal of Intellectual Disability Research, 53*(3), 200–216. https://doi.org/10.1111/j.1365-2788.2008.01060.x

Cowdery, G. E., Iwata, B. A., & Pace, G. M. (1990). Effects and side effects of DRO as treatment for self-injurious behavior. *Journal of Applied Behavior Analysis, 23*(4), 497–506.

Cox, A. D., & Virues-Ortega, J. (2016). Interactions between behavior function and psychotropic medication. *Journal of Applied Behavior Analysis, 49*, 85–104. https://doi.org/10.1002/jaba.247

Crossland, S., Burns, M., Leach, C., & Quinn, P. (2005). Needs assessment in forensic learning disability. *Medicine, Science and the Law, 45*(2), 147–153.

Davis, K., & Gavidia-Payne, S. (2009). The impact of child, family, and professional support characteristics on the quality of life in families of young children with disabilities. *Journal of Intellectual and Developmental Disability, 34*(2), 153–162.

Derby, K. M., Fisher, W. W., & Piazza, C. C. (1996). The effects of contingent and noncontingent attention on self-injury and self-restraint. *Journal of Applied Behavior Analysis, 29*, 107–110. https://doi.org/10.1901/jaba.1996.29-107

Deshais, M. W., Fisher, A. B., Hausman, N. L., & Kahng, S. (2015). Further investigation of a rapid restraint analysis. *Journal of Applied Behavior Analysis, 48*(4), 845–859.

Devine, D. P. (2012). Animal models of self-injurious behaviour: An overview. In *Psychiatric disorders* (Methods and Protocols) (pp. 65–84). New York: Springer.

Dorsey, M. F., Iwata, B. A., Reid, D. H., & Davis, P. A. (1982). Protective equipment: Continuous and contingent application in the treatment of self-injurious behavior. *Journal of Applied Behavior Analysis, 15*(2), 217–230.

Doyle, C. A., & McDougle, C. J. (2012). Pharmacologic treatments for the behavioral symptoms associated with autism spectrum disorders across the lifespan. *Dialogues in Clinical Neuroscience, 14*(3), 263–279.

Drasgow, E., & Yell, M. L. (2001). Functional behavioral assessments: Legal requirements and challenges. *School Psychology Review, 30*(2), 239–251.

Duerden, E. G., Oatley, H. K., Mak-Fan, K. M., McGrath, P. A., Taylor, M. J., Szatmari, P., Roberts, S. W. (2012). Risk Factors Associated with Self-Injurious Behaviors in Children and Adolescents with Autism Spectrum Disorders. *Journal of Autism and Developmental Disorders, 42*(11):2460–2470.

Dura, J. R., Mulick, J. A., & Hammer, D. (1988). Rapid clinical evaluation of sensory integrative therapy for self-injurious behavior. *Mental Retardation, 26*, 83–87.

Emerson, E. (1992). Self-injurious behaviour: An overview of recent trends in epidemiological and behavioural research. *Mental Handicap Research (Journal of Applied Research in Intellectual Disabilities), 5*, 49–81. https://doi.org/10.1111/j.1468-3148.1992.tb00170.x

Emerson, E., Kiernan, C., Alborz, A., Reeves, D., Mason, H., Swarbrick, R., … Hatton, C. (2001a). The prevalence of challenging behaviors: A total population study. *Research in Developmental Disabilities, 22*(1), 77–93.

Emerson, E., Kiernan, C., Alborz, A., Reeves, D., Mason, H., Swarbrick, R., … Hatton, C. (2001b). Predicting the persistence of severe self-injurious behaviour. *Research in Developmental Disabilities, 22*, 67–75.

Favell, J. E., McGimsey, J. F., & Schell, R. M. (1982). Treatment of self-injury by providing alternate sensory activities. *Analysis and Intervention in Developmental Disabilities, 2*, 83–104.

Finucane, B., Haines Dirrigl, K., & Simon, E. W. (2001). Characterization of self-injurious behaviors in children and adults with Smith-Magenis syndrome. *American Journal on Mental Retardation, 106*(1), 52–58.

Fisher, W. W., & Iwata, B. A. (1996). On the function of self-restraint and its relationship to self-injury. *Journal of Applied Behavior Analysis, 29*, 93–98. https://doi.org/10.1901/jaba.1996.29-93

Fisher, W. W., Kuhn, D. E., & Thompson, R. H. (1998). Establishing discriminative control of responding using functional and alternative reinforcers during functional communication training. *Journal of Applied Behavior Analysis, 31*(4), 543–560.

Fisher, W. W., Piazza, C. C., Bowman, L. G., Kurtz, P. F., Sherer, M. R., & Lackman, S. R. (1994). A preliminary evaluation of empirically derived consequences for the treatment of pica. *Journal of Applied Behavior Analysis, 27*, 447–457.

Fritz, J. N., Iwata, B. A., Hammond, J. L., & Bloom, S. E. (2013). Experimental analysis of precursors to severe problem behavior. *Journal of Applied Behavior Analysis, 46*(1), 101–129.

Fryling, M. J., Wallace, M. S., & Yassine, J. N. (2012). Impact of treatment integrity on intervention effectiveness. *Journal of Applied Behavior Analysis, 45*(2), 499–453.

Fung, L. K., Mahajan, R., Nozzolillo, A., Bernal, P., Krasner, A., Jo, B., … Hardan, A. (2016). Pharmacologic treatment of severe irritability and problem behavior in autism: A systematic review and meta-analysis. *Pediatrics, 137*, S124.

Garber, H. J., McGonigle, J. J., Slomka, G. T., & Monteverde, E. (1992). Clomipramine treatment of stereotypic behaviors and self-injury in patients with developmental disabilities. *Journal of the American Academy of Child & Adolescent Psychiatry, 31*(6), 1157–1160. https://doi.org/10.1097/00004583-199211000-00027

Gibson, A. K., Hetrick, W. P., Taylor, D. V., Sandman, C. A., & Touchette, P. (1995). Relating the efficacy of naltrexone in treating self injurious behaviour to motivation assessment scale. *Journal of Developmental and Physical Disabilities, 7*, 215–220.

Goh, H., & Iwata, B. A. (1994). Behavioral persistence and variability during extinction of self-injury maintained by escape. *Journal of Applied Behavior Analysis, 27*(1), 173–174.

Gormez, A., Rana, F., & Varghese, S. (2014). Pharmacological interventions for self-injurious behaviour in adults with intellectual disabilities: Abridged republication of a Cochrane systematic reviews. *Journal of Psychopharmacology, 28*, 264–632. https://doi.org/10.1177/0269881114531665

Grant, J. E., Chamberlain, S. R., Redden, S. A., Leppink, E. W., Odlaug, B. L., & Kim, S. W. (2016). N-acetylcysteine in the treatment of excoriation disorder: A randomized clinical trial. *JAMA Psychiatry, 73*(5), 490–496.

Gresham, F. M., Watson, T. S., & Skinner, C. H. (2001). Functional behavioral assessment: Principles, procedures, and future direction. *School Psychology Review, 30*(2), 156.

Griffin, J. C., Williams, D. E., Stark, M. T., Altmeyer, B. K., & Mason, M. (1986). Self-injurious behavior: A state-wide prevalence survey of the extent and circumstances. *Applied Research in Mental Retardation, 7*(1), 105–116.

Hagopian, L. P., Rooker, G. W., & Zarcone, J. R. (2015). Delineating subtypes of self-injurious behavior maintained by automatic reinforcement. *Journal of Applied Behavior Analysis, 48*(3), 523–543.

Hall, A. G., Bouldin, E. D., Andresen, E. M., & Ali, A. K. (2012). Maintaining employment among caregivers of individuals on a medicaid waitlist for services. *Journal of Disability Policy Studies, 23*(2), 121–128.

Hanley, G. P., Iwata, B. A., & McCord, B. E. (2003). Functional analysis of problem behavior: A review. *Journal of Applied Behavior Analysis, 36*(2), 147–185. https://doi.org/10.1901/jaba.2003.36-147

Hanley, G. P., Iwata, B. A., & Thompson, R. H. (2001). Reinforcement schedule thinning following treatment with functional communication training. *Journal of Applied Behavior Analysis, 31*(1), 17–38. https://doi.org/10.1002/jaba.236

Hartley, S. L., Barker, E. T., Baker, J. K., Seltzer, M. M., & Greenberg, J. S. (2012). Marital satisfaction and life circumstances of grown children with autism across 7 years. *Journal of Family Psychology, 26*(5), 688.

Hellings, J. A., Weckbaugh, M., Nickel, E. J., Cain, S. E., Zarcone, J. R., Reese, R. M., … Cook, E. H. (2005). A double-blind, placebo-controlled study of valproate for aggression in youth with pervasive developmental disorders. *Journal of Child and Adolescent Psychopharmacology, 15*(4), 682–692.

Irvin, D. S., Thompson, T. J., Turner, W. D., & Williams, D. E. (1998). Utilizing increased response effort to reduce chronic hand mouthing. *Journal of Applied Behavior Analysis, 31*(3), 375–385.

Iwata, B. A., Dorsey, M. F., Slifer, K. J., Bauman, K. E., & Richman, G. S. (1982/1994). Toward a functional analysis of self-injury. *Journal of Applied Behavior Analysis, 27*(2), 197–209.

Jacobson, C. M., & Gould, M. (2007). The epidemiology and phenomenology of non-suicidal self-injurious behavior among adolescents: A critical review of the literature. *Archives of Suicide Research, 11*, 129–147. https://doi.org/10.1080/13811110701247602

Janowsky, D. S., Shetty, M., Barnhill, J., Elamir, B., & Davis, J. M. (2005). Serotonergic antidepressant effects on aggressive, self-injurious and destructive/disruptive behaviours in intellectually disabled adults: A retrospective, open-label, naturalistic trial. *International Journal of Neuropsychopharmacology, 8*(1), 37–48. https://doi.org/10.1017/S146114570400481X

Jones, E., Allen, D., Moore, K., Phillips, B., & Lowe, K. (2007). Restraint and self-injury in people with intellectual disabilities: A review. *Journal of Intellectual Disabilities, 11*(1), 105–118.

Kahng, S., Hausman, N. L., Fisher, A. B., Donaldson, J. M., Cox, J. R., Lugo, M., & Wiskow, K. M. (2015). The safety of functional analyses of self-injurious behavior. *Journal of Applied Behavior Analysis, 48*(1), 107–114.

Kahng, S., Iwata, B. A., & Lewin, A. B. (2002). Behavioral treatment of self-injury, 1964 to 2000. *American Journal on Mental Retardation, 107*(3), 212–221.

Kars, H., Broekema, W., Glaudemans-van Gelderen, I., Verhoeven, W. M. A., & Van Ree, J. M. (1990). Naltrexone attenuates self-injurious behavior in mentally retarded subjects. *Biological Psychiatry, 27*(7), 741–746.

Kennedy, C. H., & Meyer, K. A. (1996). Sleep deprivation, allergy symptoms, and negatively reinforced problem behavior. *Journal of Applied Behavior Analysis, 29*, 133–135. https://doi.org/10.1901/jaba.1996.29-133

Kinnealey, M. (2005). Effects of sensory integration intervention on self-stimulating and self-injurious behaviors. *The American Journal of Occupational Therapy, 59*(4), 418–425.

Klonsky, E. D. (2007). The functions of deliberate self-injury: A review of the evidence. *Clinical Psychology Review, 27*(2), 226–239.

Kruger, T., & Christophersen, E. (2006). An open label study of the use of dronabinol (marinol) in the management of treatment-resistant self-injurious behavior in 10 retarded adolescent patients. *Journal of Developmental and Behavior Pediatrics, 27*(5), 433.

Kurtz, P. F., Chin, M. D., Huete, J. M., & Cataldo, M. F. (2012). Identification of emerging self-injurious behavior in young children: A preliminary study. *Journal of Mental Health Research in Intellectual Disabilities, 5*, 260–285. https://doi.org/10.1080/19315864.2011.600809

Kurtz, P. F., Chin, M. D., Huete, J. M., Tarbox, R. S., O'Connor, J. T., Paclawskyj, T. R., & Rush, K. S. (2003). Functional analysis and treatment of self-injurious behavior in young children: A summary of 30 cases. *Journal of Applied Behavior Analysis, 36*(2), 205–219.

Lakin, K. C., Hill, B. K., Hauber, F. A., Bruininks, R. H., & Heal, L. W. (1983). New admissions and readmissions to a national sample of public residential facilities. *American Journal of Mental Deficiency, 88*, 13–20.

Lance, E. I., York, J. M., Lee, L. C., & Zimmerman, A. W. (2014). Association between regression and self injury among children with autism. *Research in Developmental Disabilities, 35*(2), 408–413.

Lerman, D. C., & Iwata, B. A. (1996). A methodology for distinguishing between extinction and punishment effects of response blocking. *Journal of Applied Behavior Analysis, 29*, 231–233.

Lundqvist, L., Andersson, G., & Viding, J. (2009). Effects of vibroacoustic music on challenging behaviors in individuals with autism and developmental disabilities. *Research in Autism Spectrum Disorders, 3*, 390–400.

Lundqvist, L. O. (2013). Prevalence and risk markers of behavior problems among adults with intellectual disabilities: A total population study in Örebro County, Sweden. *Research in Developmental Disabilities, 34*(4), 1346–1356.

Mandell, D. S. (2008). Psychiatric hospitalization among children with autism spectrum disorders. *Journal of Autism and Developmental Disorders, 38*(6), 1059–1065.

Marcus, R. N., Owen, R., Kamen, L., Manos, G., McQuade, R. D., Carson, W. H., & Aman, M. G. (2009). A placebo-controlled, fixed-dose study of aripiprazole in children and adolescents with irritability associated with autistic disorder. *Journal of the American Academy of Child and Adolescent Psychiatry, 48*(11), 1110–1119.

Markowitz, P. I. (1992). Effect of fluoxetine on self-injurious behavior in the developmentally disabled: A preliminary study. *Journal of Clinical Psychopharmacology, 12*(1), 27–31. https://doi.org/10.1097/00004714-199202000-00005

Marler, S., Sanders, K. B., & Veenstra-VanderWeele, J. (2014). N-acetylcysteine as treatment for self-injurious behavior in a child with autism. *Journal of Child and Adolescent Psychopharmacology, 24*(4), 231–234. https://doi.org/10.1089/cap.2013.0137

Mason, S. A., & Iwata, B. A. (1990). Artifactual effects of sensory-integrative therapy on self-injurious behavior. *Journal of Applied Behavior Analysis, 23*, 361–370.

Mazaleski, J. L., Iwata, B. A., Rodgers, T. A., Vollmer, T. R., & Iwata, B. A. (1992). Differential reinforcement as treatment for behavior disorders: Procedural and functional variations. *Research in Developmental Disabilities, 13*(4), 393–417.

Mazaleski, J. L., Iwata, B. A., Vollmer, T. R., Zarcone, J. R., & Smith, R. G. (1993). Analysis of the reinforcement and extinction components in DRO contingencies with self-injury. *Journal of Applied Behavior Analysis, 26*(2), 143–256.

McClintock, K., Hall, S., & Oliver, C. (2003). Risk markers associated with challenging behaviours in people with intellectual disabilities: A meta-analytic study. *Journal of Intellectual Disability Research, 47*(6), 405–416.

McCord, B. E., Thomson, R. J., & Iwata, B. A. (2001). Functional analysis and treatment of self-injury associated with transitions. *Journal of Applied Behavior Analysis, 34*(2), 195–210.

McCracken, J. T., McGough, J., Shah, B., Cronin, P., Hong, D., Aman, M. G., … Research Units on Pediatric Psychopharmacology Autism Network. (2002). Risperidone in children with autism and serious behavioral problems. *New England Journal of Medicine, 347*(5), 314–321.

McDougle, C., Stigler, K., Erickson, C., & Posey, D. (2006). Pharmacology of autism. *Clinical Neuroscience Research, 6*(3–4), 179–188. https://doi.org/10.1016/j.cnr.2006.06.012

McKerchar, T. L., Kahng, S., Casioppo, E., & Wilson, D. (2001). Functional analysis of self-injury maintained by exposing masked social functions. *Behavioral Interventions, 16*, 59–63. https://doi.org/10.1002/bin.78

McPheeters, M. L., Warren, A., Sathe, N., Bruzek, J. L., Krishnaswami, S., Jerome, R. N., & Veenstra-VanderWeele, J. (2011). A systematic review of medical treatments for children with autism spectrum disorders. *Pediatrics, 127*(5), e1312. https://doi.org/10.1542/peds.2011-0427

Minshawi, N. F., Hurwitz, S., Morriss, D., & McDougle, C. J. (2015). Multidisciplinary assessment and treatment of self-injurious behavior in autism spectrum disorder and intellectual disability: Integration of psychological and biological theory and approach. *Journal of Autism and Developmental Disorders, 45*(6), 1541–1568. https://doi.org/10.1007/s10803-014-2307-3

Moore, J. W., Fisher, W. W., & Pennington, A. (2004). Systematic application and removal of protective equipment in the assessment of multiple topographies of self-injury. *Journal of Applied Behavior Analysis, 37*(1), 73–77. https://doi.org/10.1901/jaba.2004.37-73

Muller, C. L., Anacker, A. M., & Veenstra-VanderWeele, J. (2016). The serotonin system in autism spectrum disorder: From biomarker to animal models. *Neuroscience, 321*, 24–41.

Neidert, P. L. (2007). Prevalence and functions of self-injurious behavior in the Prader-Willi syndrome (Doctoral dissertation, University of Florida).

Oliver, C., Petty, J., Ruddick, L., & Bacarese-Hamilton, M. (2012). The association between repetitive, self-injurious and aggressive behavior in children with severe intellectual disability. *Journal of Autism and Developmental Disorders, 42*(6), 910–919.

Ottenbacher, K. (1991). Research in sensory integration: Empirical perceptions and progress. In A. G. Fisher, E. A. Murray, & A. C. Bundy (Eds.), *Sensory integration: Theory and practice*. Philadelphia: F. A. Davis.

Owen, R., Sikich, L., Marcus, R. N., Corey-Lisle, P., Manos, G., McQuade, R. D., … Findling, R. L. (2009). Aripiprazole in the treatment of irritability in children and adolescents with autistic disorder. *Pediatrics, 124*(6), 1533–1540.

Peebles, K. A., & Price, T. J. (2012). Self-injurious behaviour in intellectual disability syndromes: Evidence for aberrant pain signaling as a contributing factor. *Journal of Intellectual Disability Research, 56*(5), 441–452. https://doi.org/10.1111/j.1365-2788.2011.01484.x

Percinel, I., & Yazici, K. U. (2014). Glutamatergic dysfunction in skin-picking disorder: Treatment of a pediatric patient with N-acetylcysteine. *Journal of Clinical Psychopharmacology, 34*(6), 772–774.

Piazza, C. C., Adelinis, J. D., Hanley, G. P., Goh, H. L., & Delia, M. D. (2000). An evaluation of the effects of matched stimuli on behaviors maintained by automatic reinforcement. *Journal of Applied Behavior Analysis, 33*(1), 13–27.

Piazza, C. C., Fisher, W. W., Hanley, G. P., LeBlanc, L. A., Worsdell, A. S., Lindauer, S. E., & Keeney, K. M. (1999). Treatment of pica through multiple analyses of its reinforcing functions. *Journal of Applied Behavior Analysis, 31*, 165–189.

Rapp, J. T., & Miltenberger, R. G. (2000). Self-restraint and self-injury: A demonstration of separate functions and response classes. *Behavioral Interventions, 15*, 37–51. https://doi.org/10.1002/(SICI)1099-078X(200001/03)15:1<37::AID-BIN40>3.0.CO;2-5

Rattaz, C., Michelon, C., & Baghdadli, A. (2015). Symptom severity as a risk factor for self-injurious behaviours in adolescents with autism spectrum disorders. *Journal of Intellectual Disability Research, 59*, 730–740. https://doi.org/10.1111/jir.12177

Reid, D. J., Parsons, M. B., Phillips, J. E., & Green, C. W. (1993). Reduction of self-injurious hand mouthing using response blocking. *Journal of Applied Behavior Analysis, 26*, 139–140.

Richards, C., Oliver, C., Moss, J., & Nelson, L. (2012). Self-injurious behaviour in individuals with autism spectrum disorder and intellectual disability. *Journal of Intellectual Disability Research, 56*, 476–489. https://doi.org/10.1111/j.1365-2788.2012.01537.x

Richardson, J. S., & Zaleski, W. A. (1983). Naloxone and self-mutilation. *Biological Psychiatry, 18*(1), 99–101.

Richman, D. M., Barnard-Brak, L., Bosch, A., Thompson, S., Grubb, L., & Abby, L. (2013). Predictors of self-injurious behaviour exhibited by individuals with autism spectrum disorder. *Journal of Intellectual Disability Research, 47*, 429–439. https://doi.org/10.1111/j.1365-2788.2012.01628.x

Richman, D. M., & Lindauer, S. (2005). Longitudinal assessment of stereotypic, proto-injurious, and self-injurious behavior exhibited by young children with developmental delays. *American Journal of Mental Retardation, 110*, 439–450. https://doi.org/10.1352/0895-8017(2005)110[439:LAOSPA]2.0.CO;2

Ricketts, R. W., Goza, A. B., Ellis, C. R., Singh, Y. N., Singh, N. N., & Cooke, J. C. (1993). Fluoxetine treatment of severe self-injury in young adults with mental retardation. *Journal of the American Academy of Child & Adolescent Psychiatry, 32*(4), 865–869. https://doi.org/10.1097/00004583-199307000-00024

Rincover, A., & Devany, J. (1982). The application of sensory extinction procedures to self-injury. *Analysis and Intervention in Developmental Disabilities, 2*(1), 67–81.

Roberts, M. A., Mace, F. C., & Daggett, J. A. (1995). Preliminary comparison of two negative reinforcement schedules to reduce self-injury. *Journal of Applied Behavior Analysis, 28*(4), 579–580.

Rojahn, J., Schroeder, S. R., & Hoch, T. A. (2007). *Self-injurious behavior in intellectual disabilities* (Vol. 2). Amsterdam: Elsevier.

Rojahn, J., Wilkins, J., Matson, J. L., & Boisjoli, J. (2010). A comparison of adults with intellectual disabilities with and without ASD on parallel measures of challenging behaviour: The behavior problems inventory-01 (BPI-01) and autism spectrum disorders-behavior problems for intellectually disabled adults (ASD-BPA). *Journal of Applied Research in Intellectual Disabilities, 23*(2), 179–185.

Rojas, D. C. (2014). The role of glutamate and its receptors in autism and the use of glutamate receptor antagonists in treatment. *Journal of Neural Transmission, 121*(8), 891–905. https://doi.org/10.1007/s00702-014-1216-0

Romanczyk, R. G., Kistner, J. A., & Plienis, A. (1982). Self-stimulatory and self-injurious behavior: Etiology and treatment. *Advances in Child Behavioral Analysis and Therapy, 2*, 189–254.

Rooker, G. W., & Roscoe, E. M. (2005). Functional analysis of self-injurious behavior and its relation to self-restraint. *Journal of Applied Behavior Analysis, 35*, 537–542. https://doi.org/10.1901/jaba.2005.12-05

Roscoe, E. M., Iwata, B. A., & Goh, H. (1998). A comparison of noncontingent reinforcement and sensory extinction as treatments for self-injurious behavior. *Journal of Applied Behavior Analysis, 31*(4), 635–646.

Rosenblatt, L. E., Gorantla, S., Torres, J. A., Yarmush, R. S., Rao, S., Park, E. R., … Levine, J. B. (2011). Relaxation response-based yoga improves functioning in young children with autism: A pilot study. *Journal of Alternative and Complimentary Medicine, 17*(11), 1029–1035.

Roy, A., Roy, M., Deb, S., Unwin, G., & Roy, A. (2015). Are opioid antagonists effective in reducing self-injury in adults with intellectual disability? A systematic review. *Journal of Intellectual Disability Research, 59*(1), 55–67. https://doi.org/10.1111/jir.12111

Ruedrich, S., Swales, T. P., Fossaceca, C., Toliver, J., & Rutkowski, A. (1999). Effect of divalproex sodium on aggression and self-injurious behaviour in adults with intellectual disability: A retrospective review. *Journal of Intellectual Disability Research, 43*(2), 105–111.

Sandman, C. A. (1988). Β-endorphin dysregulation in autistic and self-injurious behavior: A neurodevelopmental hypothesis. *Synapse, 2*(3), 193–199. https://doi.org/10.1002/syn.890020304

Sandman, C. A., Hetrick, W. P., Taylor, D. V., & Chicz-DeMet, A. (1997). Dissociation of POMC peptides after self-injury predicts response to centrally acting opiate blockers. *American Journal of Mental Retardation, 102*, 182–199. https://doi.org/10.1352/0895-8017(1997)102<0182:DOPPAS>2.0.CO;2

Sandman, C. A., & Kemp, A. S. (2011). Opioid antagonists may reverse endogenous opiate "dependence" in the treatment of self-injurious behavior. *Pharmaceuticals, 4*(2), 366–381.

Scheithauer, M., Connor, J. O'., Toby, L. M. (2015). Assessment of self-restraint using a functional analysis of self-injury. *Journal of Applied Behavior Analysis, 48*(4):907–911.

Scheithauer, M., Lomas Mevers, J., Call, N., & Shrewsbury, A. N. (2017). Using a test for multiply-maintained self injury to develop function-based treatments. *Journal of Developmental and Physical Disabilities, 29*, 443–460. https://doi.org/10.1007/s10882-017-9535-3

Schneider, M. J., Bijam-Schulte, A. M., Janssen, C. G. C., & Stolk, J. (1996). The origin of self-injurious behaviour of children with mental retardation. *The British Journal of Development Disabilities, 42*(83), 136–148.

Scifo, R., Cioni, M., Nicolosi, A., Batticane, N., Tirolo, C., Testa, N., … Marchetti, B. (1996). Opioid-immune interactions in autism: Behavioural and immunological assessment during a double-blind treatment with naltrexone. *Annali Dell'Istituto Superiore di Sanita, 32*(3), 351–359.

Shattuck, P. T., Seltzer, M. M., Greenberg, J. S., Orsmond, G. I., Bolt, D., Kring, S., … Lord, C. (2007). Change in autism symptoms and maladaptive behaviors in adolescents and adults with an autism spectrum disorder. *Journal of Autism and Developmental Disorders, 37*(9), 1735–1747. https://doi.org/10.1007/s10803-006-0307-7

Shea, S., Turgay, A., Carroll, A., Schulz, M., Orlik, H., Smith, I., & Dunbar, F. (2004). Risperidone in the treatment of disruptive behavioral symptoms in children with autistic and other pervasive developmental disorders. *Pediatrics, 114*(5), 634–641.

Shinohe, A., Hashimoto, K., Nakamura, K., Tsujii, M., Iwata, Y., Tsuchiya, K. J., et al. (2006). Increased serum levels of glutamate in adult patients with autism. *Progress in Neuro- Psychopharmacology and Biological Psychiatry, 30*(8), 1472–1477.

Shore, B. A., Iwata, B. A., DeLeon, I. G., Kahng, S., & Smith, R. G. (1997). An analysis of reinforce substitutability using object manipulation and self-injury as competing responses. *Journal of Applied Behavior Analysis, 30*(1), 21–41.

Simeon, D., Stein, D. J., Gross, S., Islam, N., Schmeidler, J., & Hollander, E. (1997). A double-blind trial of fluoxetine in pathologic skin picking. *The Journal of Clinical Psychiatry, 58*(8), 341–347. https://doi.org/10.4088/JCP.v58n0802

Smith, A. C., Dykens, E., & Greenberg, F. (1998). Behavioral phenotype of Smith-Magenis syndrome (del 17p11. 2). *American Journal of Medical Genetics, 81*(2), 179–185.

Smith, R. G., Iwata, B. A., Vollmer, T. R., & Pace, G. M. (1992). On the relationship between self-injurious behavior and self-restraint. *Journal of Applied Behavior Analysis, 25*, 433–445. https://doi.org/10.1901/jaba.1992.25-433

Smith, R. G., Iwata, B. A., Vollmer, T. R., & Zarcone, J. R. (1993). Experimental analysis and treatment of multiply controlled self-injury. *Journal of Applied Behavior Analysis, 26*, 183–196. https://doi.org/10.1901/jaba.1993.26-183

Smith, R. G., Russo, L., & Le, D. D. (1999). Distinguishing between extinction and punishment effects of response blocking: A replication. *Journal of Applied Behavior Analysis, 32*(3), 367–370.

Sohanpal, S. K., Deb, S., Thomas, C., Soni, R., Lenotre, L., & Unwin, G. (2007). The effectiveness of antidepressant medication in the management of behaviour problems in adults with intellectual disabilities: A systematic review. *Journal of Intellectual Disability Research, 51*, 750–765. https://doi.org/10.1111/j.1365-2788.2006.00935.x

Stokes, T. E., & Baer, D. M. (1977). An implicit technology of generalization. *Journal of Applied Behavior Analysis, 10*(2), 349–367.

Symons, F. J. (2011). Self-injurious behavior in neurodevelopmental disorders: Relevance of nociceptive and immune mechanisms. *Neuroscience and Biobehavioral Reviews, 35*(5), 1266–1274. https://doi.org/10.1016/j.neubiorev.2011.01.002

Symons, F. J., Harper, V. N., McGrath, P. J., Breau, L. M., & Bodfish, J. W. (2009). Evidence of increased nonverbal behavioral signs of pain in adults with neurodevelopmental disorders and chronic self-injury. *Research in Developmental Disabilities, 30*(3), 521–528.

Symons, F. J., Shinde, S., Clary, J., Harper, V., & Bodfish, J. W. (2010). A sham-controlled sensory testing protocol for nonverbal individuals with intellectual and developmental disabilities: Self-injury and gender effects. *Journal of Pain, 11*, 773–781.

Taylor, D. V., Rush, D., Hetrick, W. P., & Sandman, C. A. (1993). Self-injurious behavior within the menstrual cycle of women with mental retardation. *American Journal on Mental Retardation*, *97*, 659–664.

Taylor, L., Oliver, C., & Murphy, G. (2011). The chronicity of self-injurious behaviour: A long-term follow-up of a total population study. *Journal of Applied Research in Intellectual Disabilities, 24*(2), 105–117.

Thomason-Sassi, J. L., Iwata, B. A., Neidert, P. L., & Roscoe, E. M. (2011). Response latency as an index of response strength during functional analyses of problem behavior. *Journal of Applied Behavior Analysis, 44*(1), 51–67. https://doi.org/10.1901/jaba.2011.44-51

Thompson, R. H., & Iwata, B. A. (2007). A comparison of outcomes from descriptive and functional analyses of problem behavior. *Journal of Applied Behavior Analysis, 40*(2), 333–338. https://doi.org/10.1901/jaba.2007.56-06

Tiger, J. H., Hanley, G. P., & Bruzek, J. (2008). Functional communication training: A review and practical guide. *Behavior Analysis in Practice, 1*(1), 16–23. https://doi.org/10.1007/BF03391716

Tolin, D. F., McKay, D., Forman, E. M., Klonsky, E. D., & Thombs, B. D. (2015). Empirically supported treatment: Recommendations for a new model. *Clinical Psychology: Science and Practice, 22*(4), 317–338. https://doi.org/10.1111/cpsp.12122

Tsiouris, J., Kim, S., Brown, W., Pettinger, J., & Cohen, I. (2013). Prevalence of psychotropic drug use in adults with intellectual disability: Positive and negative findings from a large scale study. *Journal of Autism and Developmental Disorders, 43*(3), 719–731. https://doi.org/10.1007/s10803-012-1617-6

Turner, W. D., Realon, R. E., Irvin, D., & Robinson, E. (1996). The effects of implementing program consequences with a group of individuals who engaged in sensory maintained hand mouthing. *Research in Developmental Disabilities, 17*(4), 311–330.

Vollmer, T. R. (1994). The concept of automatic reinforcement: Implications for behavioral research in developmental disabilities. *Research in Developmental Disabilities, 15*(3), 187–207.

Vollmer, T. R., & Iwata, B. A. (1992). Differential reinforcement as treatment for behavior disorders: Procedural and functional variations. *Research in Developmental Disabilities, 13*(4), 393–417.

Vollmer, T. R., Iwata, B. A., Zarcone, J. R., Smith, R. G., & Mazaleski, J. L. (1993). The role of attention in the treatment of attention-maintained self-injurious behavior: Noncontingent reinforcement and differential reinforcement of other behavior. *Journal of Applied Behavior Analysis, 26*(1), 9–21.

Vollmer, T. R., Marcus, B. A., & Ringdahl, J. E. (1995). Noncontingent escape as treatment for self-injurious behavior maintained by negative reinforcement. *Journal of Applied Behavior Analysis, 28*(1), 15–26.

Vollmer, T. R., Marcus, B. A., Ringdahl, J. E., & Roane, H. S. (1995). Progressing from brief assessments to extended experimental analyses in the evaluation of aberrant behavior. *Journal of Applied Behavior Analysis, 28*(4), 561–576. https://doi.org/10.1901/jaba.1995.28-561

Wells, M. E., & Smith, D. W. (1983). Reduction of self-injurious behavior of mentally retarded persons using sensory integrative techniques. *American Journal of Mental Deficiency, 87*, 664–666.

Whitlock, J., Eckenrode, J., & Silverman, D. (2006). Self-injurious behaviors in a college population. *Pediatrics, 117*(6), 1939–1948.

Willemsen-Swinkels, S. H., Buitelaar, J. K., Nijhof, G. J., & van Engeland, H. (1995). Failure of naltrexone hydrochloride to reduce self-injurious and autistic behavior in mentally retarded adults: Double-blind placebo-controlled studies. *Archives of General Psychiatry, 52*(9), 766–773. https://doi.org/10.1001/archpsyc.1995.03950210060011

Wolff, J. J., Hazlett, H. C., Lightbody, A. A., Reiss, A. L., & Piven, J. (2013). Repetitive and self-injurious behaviors: Associations with caudate volume in autism and fragile X syndrome. *Journal of Neurodevelopmental Disorders, 5*(1), 12.

Wong, D. F., Harris, J. C., Naidu, S., Yokoi, F., Marenco, S., Dannals, R. F., … Bryan, R. N. (1996). Dopamine transporters are markedly reduced in Lesch-Nyhan disease in vivo. *Proceedings of the National Academy of Sciences, 93*(11), 5539–5543.

Zingarelli, G., Ellman, G., Hom, A., Wymore, M., Heidorn, S., & Chicz-DeMet, A. (1992). Clinical effects of naltrexone on autistic behavior. *American Journal of Mental Retardation, 97*(1), 57–63.

Pica

33

Matthew O'Brien

Pica is an eating disorder that is characterized by the consumption of nonfood substances. It is believed to affect individuals with intellectual and developmental disabilities (ID/DD) at a disproportionally higher rate than typically developing individuals (Ali, 2001). Pica is often referred to as a self-injurious behavior (SIB) based upon the potential for numerous harmful outcomes, including poisoning, infection, intestinal blockages and perforations, and even death (Sturmey & Williams, 2016). Given such outcomes, it should not be a surprise that much research has been dedicated to understanding the etiology of pica and searching for effective treatments. In fact, several books have been written exclusively on the subject of pica (Sturmey & Williams, 2016; Young, Sherman, Lucks, & Pelto, 2011). Yet, there continues to be a debate about the cause(s) and the most effective treatments for this disorder. Additionally, there is still much to be learned about its effects on different populations, prevalence rates, and connections to other disorders. This chapter presents an overview of pica in the ID/DD population, including what is known about diagnosis, prevalence, theories of etiology, and prevention and treatment research.

Diagnosis

Pica comes from the Latin word for magpie (T.E.C, 1969), a bird that is notorious for eating nearly anything, including nonfood items. Like the magpie, individuals diagnosed with pica consume nonfood items. This behavior is the essential feature of pica diagnosis. The *Diagnostic and Statistical Manual of Mental Disorders, Fifth Edition* (DSM-5; American Psychiatric Association, 2013), which characterizes pica as an eating disorder alongside anorexia and bulimia, defines pica by four criteria:

1. Persistent eating of nonnutritive, nonfood substances for a period of at least 1 month.
2. The eating of nonnutritive, nonfood substances is inappropriate to the developmental level of the individual.
3. The eating behavior is not part of a culturally supported or socially normative practice.
4. If occurring with another mental disorder (e.g., autism or intellectual disability), or during a medical condition (e.g., pregnancy), it is severe enough to warrant independent clinical attention.

Based on these criteria, individuals exempt from a diagnosis of pica include those from cultures where consumption of nonfood items is accepted and may be perceived to offer health benefits

---

M. O'Brien (✉)
University of Iowa Center for Disabilities and Development, University of Iowa Stead Family Children's Hospital, Iowa City, IA, USA
e-mail: matthew-j-obrien@uiowa.edu

J. L. Matson (ed.), *Handbook of Intellectual Disabilities*, Autism and Child Psychopathology Series, https://doi.org/10.1007/978-3-030-20843-1_33

(e.g., consumption of clay by pregnant women in Kenya; Geissler et al., 1999) and very young children (i.e., below the age of 2 years) for whom exploration of objects by mouth is deemed a typical behavior. It could still be argued that for both of these exempt groups, the absence of a diagnosis does not preclude intervention to prevent harmful outcomes, including toxicity to a developing fetus (Gundacker et al., 2017). Moreover, mouthing of hands and objects without consumption, which is distinguishable from pica, may pose health risks warranting treatment as well.

The DSM criteria do not offer further classification of pica behavior based upon important dimensions, such as severity, frequency, and form or type of item consumed. Although there are no validated tools available to evaluate pica severity, the *Severity Index for Pica* was created by Don Williams to delineate pica from mild to life-threatening forms (Sturmey & Williams, 2016). This scale categorizes five levels of severity (mild, moderate, severe, dangerous, and life-threatening), discriminating based upon the size and potential danger associated with the items consumed, the frequency of pica, and the negative outcomes experienced by the individual (e.g., surgery to remove an item).

Case reports of pica have documented the consumption of many different nonfood items, resulting in dozens of forms of pica, each with a unique name. Items targeted for consumption range from those more commonly documented, such as dirt, sand, clay (geophagia), and ice (pagophagia), to those rarely described in the literature, including glass (hyalophagia) and drinking urine (urophagia). McAdam, Breidbord, Levine, and Williams (2012) classified the various forms of pica found in case reports and research literature into six categories: biological secretions (e.g., blood, vomit, feces), biological solids (e.g., skin, fingernails, hair), chemicals (e.g., copper, lead paint chips), food items (e.g., raw potatoes, eggs), organic materials (e.g., dust, laundry starch, book pages), and materials causing physical damage (e.g., sharp items, stones, cigarette butts). Whether these classifications provide insight into the etiology of pica or are helpful in treatment selection has not been studied.

Epidemiology

It is difficult to provide an accurate estimate of the prevalence of pica in the general population in cultures where pica is not an acceptable behavior (APA, 2013). Nevertheless, rates of pica within the general population are believed to be much lower than those for individuals with developmental disabilities (Matson, Belva, Hattier, & Matson, 2011), with the possible exceptions of pregnant women (Fawcett, Fawcett, & Mazmanian, 2016) and very young children (Barltop, 1966). Among individuals with a developmental disability, prevalence estimates vary broadly, which is likely due to the combination of differences in how researchers have defined pica, the data collection methods used, and differences across the sampled groups (e.g., community setting versus institution). At the low end of the range, Hove (2004) found that 2.9% of adults with ID living in the general community displayed pica according to a survey completed by healthcare workers on their clientele. On the high end, Danford and Huber (1982) reported pica in 25.8% of children and adults with ID living in an institution. Studies of ID/DD individuals living in institutional settings have generally found higher rates of pica than studies of ID/DD individuals living within the general community (Matson et al., 2011). This may be attributable to the increased severity of disability and challenging behavior often found in institutionalized individuals. In fact, several studies have noted that the risk for pica in individuals with ID increases as the classification of intellectual disability moves from mild to moderate to severe/profound (Ashworth, Martin, & Hirdes, 2008; Smith, Branford, Collacott, Cooper, & McGrother, 1996; Tewari, Krishnan, Valsalan, & Roy, 1995).

Within the ID/DD population, a few subgroups are likely at an increased risk of pica. Individuals with Prader-Willi syndrome, a rare genetic condition, often exhibit insatiable appetites (hyperphagia) and obsessive foraging for food, even including food that has been discarded (Dykens, Leckman, & Cassidy, 1996). Additionally, individuals with Prader-Willi syndrome may consume nonfood items, such as paint chips or paper (Dimitropoulos et al., 2000) and contaminated food (Dykens, 2000). Despite strong evidence that pica is a dangerous behavior more likely to occur in individuals with Prader-Willi syndrome, to date no known studies have sought to estimate the prevalence of pica in this group, and Prader-Willi syndrome is conspicuously absent in most writings about pica in ID/DD populations.

Numerous case studies, predominately from the field of applied behavior analysis (ABA), report on assessment and/or treatment approaches for pica with individuals diagnosed with an autism spectrum disorder (Falcomata, Roane, & Pabico, 2007; Hagopian et al., 2011; Ing, Roane, & Veenstra, 2011; Mitteer, Romani, Greer, & Fisher, 2015; Piazza et al., 1998; Rapp, Dozier, & Carr, 2001; Slocum, Mehrkam, Peters, & Vollmer, 2017). However, only a couple of studies have examined the prevalence of pica in individuals with autism. The highest estimate of pica in autism came from a study by Kinnell (1985) who reviewed case records of 70 individuals of all ages with autism. Sixty percent had pica at some stage in life, leading Kinnell to suggest that all individuals with autism should be screened for pica. Clark, Vandermeer, Simonetti, and Buka (2010) identified a slightly lower prevalence rate (48%) of pica in a sample of 48 children with autism using a community sample. Finally, among adults in an institution with both autism and ID, Lovullo and Matson (2009) found 36% displayed pica. Most of the individuals included in these studies had also been diagnosed with ID, which raises the possibility that the connection between autism and pica is more likely a function of cognition deficits than the diagnosis of autism.

Cri du chat syndrome is a genetic condition often resulting in moderate to severe intellectual disability. In addition to a number of self-injurious behaviors, pica is regularly observed in this population. Based upon reports by 66 families of children and adults with Cri du Chat, 27% of the participants displayed pica (Ross, Collins & Cornish, 2002). However, like autism, it is unclear whether pica is particular to this syndrome or individuals with more significant cognitive impairments.

Etiology of Pica

Many theories have been posited on the cause of pica. Given the many forms of pica, it seems highly unlikely that a singular account could explain all pica cases. In fact, Sturmey and Williams (2016) devote an entire chapter in their book on pica in the ID/DD population strictly to etiology, with descriptions of ten different accounts for the existence of pica. As these authors note, each theory may make more sense "…in relationship to different populations and all with very limited quantities of evidence to support or test them…" (pg. 37). As such, for individuals with ID/DD, a behavioral explanation may be most appropriate, given the deeper history of assessment and effective treatments coming from that line of work.

Despite some overlap among the theories, theoretical accounts with supporting research may generally be categorized into one of three broad perspectives: biomedical, psychiatric, or behavioral. Each of these areas is described below with both supporting evidence and challenges to the suppositions. Note that the descriptions below are related to pica that is deemed aberrant within the culture that one lives, which excludes some culturally supported examples of pica (e.g., geophagia in African communities). Additionally, a number of theories that have been proposed are not described below as they have insufficient or no evidence in the research literature to support their account.

Biomedical Perspective

Biomedical accounts are based upon the premise that pica is the result of nutritional deficiencies, neurological impairment, and/or faulty neurochemical transmission. Nutritional deficiencies in individuals with pica have been the focus of much research and serves as one of the earliest hypotheses on the cause of pica (Young et al., 2011). Theories based upon nutritional deficits suggest that individuals who exhibit a nutritional shortfall are more likely to seek out nonfood items to resolve the insufficiency (Cavdar, Arcasoy, Cin, Babacan, & Gozdasoglu, 1983) or the insufficiency leads to a neurological disturbance, which produces pica behavior (Chisholm & Martin, 1981). The prime foci of nutritional deficit arguments have been inadequate zinc and iron (Miao, Young, & Golden, 2015), with the latter receiving the majority of interest and the most research evidence. Indeed, numerous studies have shown a link between iron-deficiency anemia and pica (Young, 2010). It should be noted that many studies linking iron deficiency and pica have focused largely on pregnant women and young children, which are populations most likely to suffer from iron deficiencies (McLean, Cogswell, Egli, Wojdyla, & de Benoist, 2009). The most convincing study is a meta-analysis conducted by Miao et al. (2015), in which the data for 43 previous studies with a diverse range in population demographics was examined to determine whether a link between iron and zinc deficiencies and pica exists. The authors found a strong correlation between pica and low iron level, as well as pica and low zinc levels. Unfortunately, this link does not provide a clear indicator of the direction of the relationship (i.e., Do deficiencies cause pica or does pica lead to deficiencies?). A challenge to the iron and zinc deficiency hypothesis is that iron and zinc supplementation rarely has been shown to reduce pica behavior (Gutelius, Millican, Layman, Cohen, & Dublin, 1962; Nchito, Geissler, Mubila, Friis, & Olsen, 2004), outside of a few case reports (e.g., Arbiter & Black, 1991; see study by Lofts, Schroeder, & Maier, 1990 for exception).

A neurological impairment hypothesis suggests that pica is the result of brain damage, dementia, or faulty neurochemical transmission. Evidence for pica as a result of brain injury is scant and largely confined to nonhuman animal studies (Aggleton & Passingham, 1981) and case reports (Lilly, Cummings, Benson, & Frankel, 1983). However, dementia patients may provide some clues as to the cause of pica. At least three studies have found higher rates of pica, particularly coprophagia (consumption of fecal matter), in individuals with dementia (Ghaziuddin & McDonald, 1985; Morris, Hope, & Fairburn, 1989; Josephs, Whitwell, Parisi, & Lapid, 2016). These studies have offered a number of possible explanations for the connection between pica and dementia, but the most common suggestion is that pica results from atrophy of the medial temporal lobe, a structure mostly associated with semantic memory.

Singh, Ellis, Crews, and Singh (1994) posited that pica may be caused by faulty dopaminergic neurotransmission. This hypothesis is supported by a study they conducted involving three adolescents with ID and a history of pica. All three participants were exposed to three conditions: a no-medication baseline, a thioridazine condition (a medication with anti-dopaminergic effects), and a methylphenidate condition (a known dopamine agonist). If pica is the result of deficient dopamine transmission, then differences in the rate of pica should be demonstrated across these conditions. Indeed, Singh et al. found that the thioridazine condition was associated with the highest rates of pica, and the methylphenidate condition resulted in the lowest rates of pica, supporting their hypothesis. Interestingly, no further studies have been conducted to test this hypothesis.

Psychiatric Perspective

Theories from a psychiatric perspective have argued that pica is a manifestation of another mental illness or a form of obsessive-compulsive disorder (OCD). Pica has been tied to several psychiatric diagnoses, including schizophrenia (Sinha & Mallick, 2010) and depression (Jawed, Krishnan, Prasher, & Corbett, 1993), but any theories explaining the relationship between these diagnoses and pica have not been fully elucidated.

Pica has been conceptualized as a form of OCD by some who see pica behavior as a compulsive act (Gundogar, Demir, & Eren, 2003; Luiselli, 1996; Stein, Bouwer, & van Heerden, 1996; Zeitlin & Polivy, 1995). In this model of pica, individuals have obsessive thoughts, often brought about by life stressors, about nonfood items. The obsessive thoughts are then relieved by the act of eating the items they have obsessed about. Support for this account comes from a few studies demonstrating remission or reduction of pica behavior when a common pharmacologic treatment (e.g., selective serotonin reuptake inhibitors) or behavior treatment (e.g., exposure and response prevention) for OCD has been employed (Gundogar et al., 2003; Stein et al., 1996; Zeitlin & Polivy, 1995).

Behavioral/Learning Perspective

Within a behavioral framework, pica is explained as a result of some rewarding outcome (i.e., reinforcement) resulting from pica behavior or due to an individual's failed discrimination of appropriate consumables. Thus, pica behavior may be learned or the result of poor stimulus control. Identifying the rewarding outcome for pica behavior has been the focus of numerous studies. Pica has rarely been found to be maintained by socially mediated reinforcers, but at least one study (Piazza, Hanley, & Fisher, 1996) identified social attention as the function- or reward-maintaining pica. More commonly, pica is thought to be reinforced by the sensory stimulation associated with consuming the nonfood items (e.g., the texture, taste) or, in the case of tobaccophagia (eating cigarette butts), the physiological effects derived from the tobacco. In support of a sensory stimulation hypothesis, studies by Piazza et al. (1998) and Favell, McGimsey, and Schell (1982) have shown that when items that match the oral sensory stimulation produced by pica are available, pica is reduced; however, when items that do not match the same sensory experience are available, there is no reduction in pica behavior. The idea that tobaccophagia arises from the effects of tobacco and not some other component of a cigarette was evaluated by Piazza et al. (1996). They recorded pica when various aspects of the cigarette butt (i.e., tobacco, filter, paper) were available and again when either cigarette butts with tobacco or cigarette butts with herbal ingredients were available. Tobacco was identified as the maintaining component based upon higher rates of pica when it was available than when other components were made available.

The argument that pica is a result of failed stimulus control suggests that individuals who engage in pica do not appropriately discriminate between food and nonfood items. More microscopically, properties of food (e.g., its appearance or smell), stimuli often associated with the delivery of food, times when food is likely to be available, and locations associated with eating food have failed to exert control over the eating behavior of some individuals. This argument has found support in research on discrimination training (Bogart, Piersel, & Gross, 1995; Finney, Russo, & Cataldo, 1982; Fisher et al., 1994; Johnson, Hunt, & Siebert, 1994). In these studies, individuals were often successfully taught to only eat food that was associated with specific stimuli, such as a placemat (Fisher et al., 1994). An argument in opposition to this account is that many individuals with pica have been described as having an obsessive pursuit of their respective pica items, but not for food items. Thus, if this theory were true, such individuals should display similarly obsessive pursuits for food items.

Treatment of Pica

Pica can be a difficult disorder to treat and may require thorough assessment to identify the most effective treatment approach. Research reveals preventive strategies, treatment strategies, and combination approaches to effectively reduce pica. Although much of the research has been focused on the use of behavior analytic tactics, biomedical treatments may also be considered, particularly for individuals who exhibit nutrient deficiencies. Although pica is occasionally observed in non-ID/DD populations, the following review reports on research exclusively using ID/DD individuals.

Behavioral Assessment

It is generally a good idea to conduct functional behavioral assessment of pica to determine what environmental variables may influence pica.

Functional behavioral assessments focus on identification of the environmental state preceding the behavior (i.e., antecedents), as well as the consequences that follow such behavior, with the goal of identifying a functional relationship between the behavior and the environment. Assessment methods vary in rigor, from those with limited validity (e.g., indirect measures) to those providing only correlational data (e.g., direct measurement) to methods that reveal causal relationships (i.e., experimental analyses). The more rigorous the assessment method, the more likely the results will lead to effective treatment outcomes (Hall, 2005).

A functional analysis (FA), which involves the manipulation of antecedent and consequence variables to determine function, requires baiting the environment with safe-to-consume items, such as uncooked beans, Play-Doh®, and candles (Piazza, Roane, Keeney, Boney, & Abt, 2002). Most research studies involving FAs of pica have indicated an automatic function, suggesting that this behavior does not tend to be sensitive to social contingencies (e.g., Hagopian, Rooker, & Rolider, 2011); however, this finding is not guaranteed. For example, Piazza et al. (1998) conducted functional analyses on three children with ID and found that pica was maintained by attention for one participant and may have been maintained, in part, by access to tangibles (i.e., cola beverage) for another participant.

Even when pica is likely to be automatically maintained, functional analyses can be beneficial in determining environmental variables that may increase or decrease the likelihood of pica. For example, in the first functional analysis of pica to be published, Mace and Knight (1986) varied the level of staff interaction and protective equipment (i.e., the use of a helmet, with and without a face shield) with a young adult with ID and pica. The analysis showed that frequent interaction and limited interaction consistently resulted in less pica than a no-interaction condition. In addition, a condition involving no protective equipment resulted in lower levels of pica than conditions where the participant was required to wear protective equipment. The assessment results were used to develop a treatment involving limited interaction and no protective equipment that ultimately led to substantial reductions in pica.

Behavioral Strategies to Reduce Pica

Much research has been conducted on strategies grounded in behavior analytic principles to reduce pica; however, for several reasons, behavior analytic studies can make it difficult to draw firm conclusions about the effectiveness of pica treatments and their generalizability. Most behavior analytic studies utilize single-case design and generally few participants. Additionally, many strategies developed using behavior analytic principles have included packaged treatments that consist of multiple treatment components, sometimes without isolating which treatment components were necessary for adequate behavior reduction.

Successful behavioral strategies can be broadly classified as prevention or treatment efforts, although a combination of prevention and treatment strategies may be optimal. Prevention strategies generally involve manipulating the environment in some fashion to reduce easy access to pica items, while treatment strategies focus on using behavioral procedures to reduce the likelihood that pica will recur, often through reinforcing the absence of pica, extinguishing attempts at pica, or punishing pica behavior. The use of restraint and response blocking may be considered preventive in that these strategies reduce the likelihood that an individual will access pica items; however, both of these strategies will be regarded as treatment strategies within the sections below.

Prevention

There are many justifications for the use of prevention strategies to reduce pica. Such strategies may be essential for individuals whose pica involves consumption of potentially lethal substances and for individuals with a long history of pica behavior. Preventive efforts may also be needed for individuals who do not have adequate supervision and in cases where treatments have not led to satisfactory reductions of pica behavior. For example, Goh, Iwata, and Kahng (1999) tried multiple treatment procedures in an attempt to reduce cigarette pica in an adult male with ID, but without sufficient reduction. As a result, they chose to forego treatment and settle for preventive efforts, including regularly cleaning areas surrounding the participant and increased supervision.

Although preventive strategies may stand alone, based upon a broad review of the research literature, Williams and McAdam (2012) suggest that prevention methods may be most beneficial when used in combination with treatment procedures. Additionally, they recommend that preventive efforts continue to be utilized until treatment strategies have been shown to be effective and generalized across people, settings, and time.

Prevention of pica is typically done in three steps: First, a comprehensive inventory is taken of the nonnutritive, nonfood substances that an individual may consume. This should include a fairly exhaustive survey, in some cases leading to a substantial list of items. Second, efforts are taken to eliminate the nonnutritive items (and other items with similar properties) from the immediate environment and/or to increase the response effort required to access such items. Finally, the third step involves regular monitoring of the environment for any items that may be considered a pica target and identification and removal of new pica items. A comprehensive management program for adults in group homes exhibiting pica was originally described by Williams, Kirkpatrick-Sanchez, Enzinna, Dunn, and Borden-Karasack (2009) and included several prevention components. This program, which effectively reduced pica for a large number of clients, included a *Pica-Safe Checklist* to be used in monitoring of pica items and pica behavior in the group home (see Sturmey & Williams, 2016, for a full description of the prevention efforts).

Pica-proofing strategies may be effective in private settings where it is easier to eliminate potential pica items, but in public places such strategies are difficult to implement and are unlikely to be effective in reducing access to pica items. Additionally, when employed as the sole strategy to address pica, prevention strategies are unlikely to produce successful long-term outcomes.

Treatment

Treatment Selection

Because pica can be a life-threatening disorder, the goal of treatment is generally to reduce pica behavior to zero (Williams & McAdam, 2012), which makes treatment selection critical. In much of the research on pica treatments, treatments deemed effective are often presented in "packaged" form with multiple treatment components required to attain sufficient reductions in pica. For example, Call, Simmons, Mevers, and Alvarez (2015) reviewed data on 11 patients who engaged in pica and were treated in an intensive day treatment program from 2001 to 2013. On average, clinicians tried more than 4 different treatment components for the 11 patients, and the final treatment for each patient, on average, consisted of nearly 2.5 different treatment components.

Deciding which treatment components to include in a treatment package may be challenging. When pica is maintained by social contingencies (e.g., caregiver attention), effective treatment will likely incorporate manipulation of the motivating operations for pica. For example, in a study by Piazza et al. (1998), a functional analysis indicated that pica was maintained by attention for a young boy with autism who exhibited pica with various items. This finding led Piazza and colleagues to develop a treatment using noncontingent attention and stimuli that matched the sensory consequences of pica, resulting in a substantial reduction of pica.

Given that pica behavior is most often automatically maintained, treatments are unlikely to be function-based, increasing the challenging of treatment selection. Such decisions require consideration of several important factors. For example, more severe or harmful pica, such as ingesting toxic substances or items that lead to intestinal blockages, may require treatments that reduce pica to zero, regardless of the intrusiveness of the intervention, whereas chronically consuming less toxic items (e.g., ice or small amounts of paper) may not necessitate the same absolute goal of reducing pica to zero and also may allow for less restrictive interventions to be tried first. Williams and McAdam (2012) raised the concern that treatments deemed "successful" because they substantially reduce rates of pica may not be sufficient, especially if pica continues to occur. They argue that evaluation of treatment effectiveness should consider both reduction in pica behavior over baseline and suppression of pica behavior to zero.

Considering suppression data is especially important for choosing treatments for life-threatening pica. McAdam et al. (2012) conducted a meta-analysis of nine behavioral treatments for pica, but considered both the effect size for pica reduction and the effect size for pica suppression (i.e., pica behavior at zero) for each treatment. Interestingly, several behavioral treatments, including negative practice, physical restraint, habit reversal, and the use of aversive stimuli, were considered moderately to highly effective at reducing pica, but showed questionable effectiveness for pica suppression. By contrast, response blocking, which only outperformed three of the other eight treatments evaluated in terms of pica reduction, was the most effective in terms of pica suppression. Thus, depending on the risk for safety, treatment selection will require consideration of the effects on pica reduction and pica suppression.

For pica that has been deemed low-risk, treatment selection should consider a hierarchical model, with the least restrictive interventions serving as the first-line option and more intrusive treatment strategies utilized when those fail (McAdam et al., 2012). When considering punishment-based approaches, clinicians must also weigh the ethics, permissibility, and acceptability of using such procedures. In addition, all treatment options must be evaluated for the practicality of treatment implementation, especially for treatments considered intensive (e.g., overcorrection).

As noted previously, nearly all published studies on treatments attaining successful reduction of pica incorporate some combination of reinforcement, punishment, and/or extinction components. However, in the following review of treatment strategies, treatments have been divided into classifications of those primarily involving strengthening of non-pica behaviors (i.e., reinforcement-based approaches) and those focused primarily on pica reduction procedures (i.e., punishment and/or extinction).

Reinforcement-Based Strategies

Differential Reinforcement

Differential reinforcement is the most popular reinforcement-based strategy used in pica treatments and in most cases is combined with other treatment components. Differential reinforcement procedures involve a combination of reinforcement for a response class or properties of a response class (e.g., an alternative behavior to pica), while withholding reinforcement for another response class (e.g., pica behavior). For pica, like many behaviors that are maintained by automatic reinforcement, a great challenge exists in withholding reinforcement for pica behavior as it may be inherently reinforcing immediately upon engaging in the act (Vollmer, 1994). Additionally, treatments for pica cannot simply differentially reinforce more acceptable behaviors while using the same maintaining putative reinforcer for pica. Thus, without adding treatment components that make it more difficult to engage in pica (e.g., response blocking) or components involving punishment procedures (e.g., contingent presentation of aversives), selection of the optimal alternative reinforcer requires that it compete with the reinforcement that results from pica.

Differential reinforcement of alternative behavior (DRA) requires that an individual engages in specific alternative behaviors to obtain reinforcement, while differential reinforcement of incompatible behavior (DRI) involves reinforcement of behaviors that are incompatible with pica behavior. Despite this difference, most studies using DRA procedures identify an alternative behavior to target for reinforcement that is incompatible with pica.

Pica treatments employing DRA and DRI procedures have targeted a variety of behaviors for reinforcement, including gum chewing (Donnelly & Olczak, 1990), toy playing (Mitteer et al., 2015), and vacuuming pica items (Schmidt et al., 2017). Smith (1987) chose to target a variety of incompatible behaviors for reinforcement for a young adult with ID who ingested dangerous inedible items. Using edibles and preferred activities as reinforcers, the behaviors programmed for reinforcement included keeping his hands on his work materials, staying in his work area, actively working, and keeping his mouth clear.

The alternative behaviors targeted for reinforcement in most studies have involved handing the pica items over to a caregiver (Donnelly & Olczak, 1990; Goh et al., 1999; Hagopian et al., 2011; Kern, Starosta, & Adelman, 2006) or discarding the items in the trash (Bogart et al., 1995; Ricciardi, Luiselli, Terrill, & Reardon, 2003; Slocum et al., 2017). Handing over a pica item to a caregiver for a tangible reinforcer has been dubbed “pica exchange.” One of the earliest demonstrations of the pica exchange procedure was by Goh et al. (1999), for four middle-aged adults with severe to profound ID who engaged in cigarette pica (tobaccophagia). All four participants were taught to hand cigarettes to a therapist in exchange for a preferred edible. The combination of pica exchange and response blocking for attempts at pica was successful in reducing pica for three of the four participants.

Treatment protocols using differential reinforcement of other behavior (DRO) also provide reinforcement for nontarget behavior; however, in contrast to DRA or DRI procedures, reinforcement is delivered for the absence of a behavior targeted for reduction over a period of time. Thus, in DRO procedures, if an individual engages in any non-pica behaviors over a specified period of time, reinforcement would be delivered. Clinically, DRO procedures are common as a treatment component for pica; however, few studies have chosen DRO over other differential reinforcement procedures in the treatment of pica. Exceptions include studies by Finney et al. (1982) and Paniagua, Braverman, and Capriotti (1986), both of which used DRO procedures, but required additional punishment components for successful reduction of pica.

Noncontingent Reinforcement and Environmental Enrichment

Noncontingent reinforcement (NCR) and environmental enrichment (EE) both involve the provision of known reinforcers, such as toys, activities, and snacks, on a noncontingent basis; however, they differ in that NCR, or time-based delivery of reinforcers, provides known reinforcers at fixed or variable intervals, while in EE these things are made available in the individual’s environment at all times. In treatments for pica, NCR and EE provide alternative sources of stimulation that, when successful, compete with the stimulation garnered from pica. These treatments, and particularly EE, may be relatively simple to implement, but there is one important caveat: In using strategies involving competing stimuli, frequent use of alternative sources of stimulation or providing access for long durations increases the possibility of satiation and likely a reduction in treatment effectiveness. This is especially true when foods serve to compete with pica and may be one reason that NCR and EE are often merely one component in effective treatment packages.

Effective NCR and EE components call for a mindful selection of stimuli to compete with pica. Stimulus preference assessments are commonly used to identify alternative sources of stimulation. It stands to reason that the more preferred an item or activity is to an individual, the more likely it will compete with the reinforcement that individual obtains through pica. For example, prior to implementing treatment for a young girl with autism who exhibited coprophagia, Ing et al. (2011) conducted preference assessments for food and nonfood items. Each item or snack was paired with a pica item (artificial feces), and the results suggested that only food items would effectively compete. Treatment, consisting of NCR with food, effectively eliminated pica when treatment was in place.

Preference assessments may be enhanced by the inclusion of items that are thought to match the stimulation accessed through pica. Piazza et al. (1998) developed an assessment to do just that. They hypothesized that pica was at least partially maintained by the oral stimulation it produced for three child participants. Stimulus preference assessments were used to evaluate preference and pica behavior for various items that would provide oral stimulation and various items that would not. For all three participants, items thought to match the oral stimulation of pica were more preferred (as measured by item interaction) and were associated with lower levels of pica. Treatments utilizing matched stimuli were shown to produce greater and more stable reductions in pica for all three participants.

Although EE and NCR are often paired with other treatment components, both may be used as a stand-alone strategy. In fact, several studies have been published showing substantial reductions in pica when employing EE as the sole treatment component for pica (Baker, Valenzuela, & Wieseler, 2005; Favell et al., 1982) or using NCR exclusively (Ing et al., 2011). Piazza et al. (2002) added an interesting twist to this line of research by varying the response effort required to access pica items and competing items in an EE context. For three teenage females with severe ID and pica, effort manipulations indicated the lowest rates of pica when (a) competing items required low effort to access, and (b) pica items required high effort. Thus, Piazza et al.'s study suggests that the most effective use of EE may require that competing stimuli are easily accessible and potential pica items require greater effort to obtain.

Discrimination Training

McAdam et al. (2012) conceptualized that pica may be due, in part, to a lack of stimulus control in which individuals poorly discriminate between proper food items and nonfood items. If this is the case, then training individuals who engage in pica to discriminate between those things they should not eat and those items that are appropriate for consumption would lead to reductions in pica. Discrimination training procedures for pica typically involve presentation of a discriminative stimulus (physical or verbal) associated with food items, followed by reinforcement of either selection or consumption of food under the presence of that stimulus, or by accurately identifying whether the item is food or nonfood. Examples of the discriminative stimuli used in published studies include placemats (Fisher et al., 1994; Johnson et al., 1994) and red dishware (Bogart et al., 1995) and questions, such as "Should you put this in your mouth?" (Finney et al., 1982). Although reinforcement of appropriate responding is essential in discrimination training, most studies also incorporate a punishment component, whereby selection of the nonfood item results in the presentation of an aversive. For example, in the Johnson et al. (1994) study, if the participant selected a food from a specific placemat, he was allowed to consume that item; however, if he chose a nonfood item, a therapist firmly said "no," "stand up," and "spit it out," followed by washing the individual's face with a cold washcloth.

Punishment-Based Approaches

The number of studies employing punishment-based treatment approaches for problem behavior reduced substantially following the introduction of a comprehensive functional analysis technology by Iwata, Dorsey, Slifer, Bauman, and Richman (1982/1994; Lydon, Healy, Moran, & Foody, 2015). The ability to identify functional relationships between environmental events and behavior has meant that treatments may target that relationship by identification and reinforcement of functionally equivalent alternative behaviors. Yet, for pica, which has consistently been found to be automatically maintained, this approach has reduced utility. Moreover, reinforcement-only approaches have rarely provided the necessary rapid reductions of pica that are desired. Thus, punishment-based approaches have long been an essential common component in behavioral treatments of pica. Successful treatment using punishment often owes to the fact that aversive environmental events (i.e., the punisher) can overpower the reinforcement garnered from the behavior being targeted for reduction (Azrin & Holz, 1966).

Successful reduction of pica through punishment procedures may be accompanied by a number of unwanted side effects. Increased aggression and emotional responding by the individual targeted for treatment have been suggested as possible side effects of punishment procedures; however, this finding is more ubiquitous in basic (nonhuman) studies than applied studies (Lerman & Vorndran, 2002). Punishment procedures may also produce escape/avoidance behavior in the presence of stimuli associated with punishment, including the individual administering the punishment (Cooper, Heron, & Heward). Lerman and Vorndran (2002) suggest that unwanted side effects of punishment procedures may be mitigated by using brief exposure to punishing stimuli and by utilizing potent reinforcers contingent upon alternative behaviors or the absence of the problem behavior.

The following is a brief description of punishment procedures that have been used to treat pica, albeit often in packages that include reinforcement-based approaches.

Response Blocking

Given the potential safety risks of pica, it should not be a surprise that many behavioral treatment packages for pica incorporate a response-blocking component. Response blocking rarely serves as a stand-alone treatment for pica (see LeBlanc, Piazza, & Krug, 1997, and McCord, Grosser, Iwata, & Powers, 2005 for exceptions). Some procedural variations exist, but response blocking typically involves placing a hand or arm between the pica item and the individual's mouth, with the greatest effectiveness when blocking is earliest in the pica chain (e.g., touching a pica item; McCord et al., 2005). It is possible that the reductive effects of this procedure are due to extinction (i.e., attempts at pica do not result in reinforcement), but more likely it serves to positively punish pica behavior (see Lerman & Iwata, 1996 for more information on distinguishing between extinction and punishment effects).

A major challenge in conducting effective response blocking is that it requires hypervigilance on the part of the person implementing it and it may require that these procedures be in place continuously for it to be effective. McCord et al. (2005) demonstrated the lack of generality of response blocking in a study involving three adult males with severe ID who all engaged in pica. After showing substantial reductions in pica using response blocking (contingent upon touching a pica item) for all three participants, McCord and colleagues had two participants return to an alone condition that previously resulted in high rates of pica for each. For both participants, rates of pica returned to levels observed in the alone condition prior to treatment.

Another problem common to response blocking, but also found with many punishment procedures, is the side effect of increased aggression (Lerman & Vorndran, 2002). Given that response blocking includes physically intervening, the implementer of treatment stands to be an easy target for aggressive behavior. Indeed, Hagopian and Adelinis (2001) conducted a treatment evaluation with a 26-year-old male with ID who exhibited high rates of aggression when a stand-alone response-blocking component was used to treat pica. Using a reversal design, Hagopian and Adelinis alternated between response blocking only and response blocking with redirection to a preferred food item. With the added redirection component, both pica and aggression were substantially reduced over the condition involving response blocking without redirection.

Time-out, Facial Screening, and Restraint

Unlike many of the punishment methods used to treatment pica, time-out, facial screening, and restraint have often been conceptualized as negative punishment procedures in that they employ removal of, or restriction from, reinforcing stimuli contingent upon pica behavior. The latter two could arguably be considered as positive punishers as well, but this distinction is an unnecessary argument for this discussion. These three procedures vary greatly in restrictiveness and acceptability.

Time-out from reinforcement is one of the most widely used and accepted parenting strategies to treat problem behavior (Barkin, Scheindlin, Ip, Richardson, & Finch, 2007), yet it has rarely been documented as a treatment strategy for pica. Kalfus, Fisher-Gross, Marvullo, and Nau (1987) conducted a unique study on the treatment of pica by using time-out without implementing additional restrictive procedures (i.e., facial screening or restraint). Using a combination of DRO with praise and tickles and a 15-s time-out from toys and attention contingent upon pica, they were able to obtain substantial reductions in pica for a 4-year-old boy with ID.

Facial screening, which involves creating a visual barrier for an individual to reduce visual stimulation, has been used extensively in treatment packages for pica (Bogart et al., 1995; Falcomata et al., 2007; Fisher et al., 1994; Mitteer et al., 2015; Singh & Winton, 1984; Williams et al., 2009). A variety of implements have been used to block visual stimulation, including blindfold (Singh & Winton, 1984), bib (Bogart et al., 1995), goggles (Williams et al., 2009), and hands (Fisher et al., 1994; Mitteer et al., 2015), most with a duration of less than 1 min. While facial screening has rarely been implemented without other treatment components (see Singh & Winton, 1984, for exception), Fisher et al. (1994) found facial screening contingent upon pica to be the most effective punishment component trialed for all three participants in their study.

In an effort to abide by the principal of least restrictiveness, physical and mechanical restraints should only be considered within a treatment package that includes reinforcement-based strategies and monitoring by a licensed professional or Board Certified Behavior Analyst trained in the treatment of problem behavior (Association for Behavior Analysis International, 2010). While such restrictive procedures are less common now than in the past, they may still be considered necessary in cases of pica that are life-threatening. Restraint may be mechanical or physical restraint, but the latter (sometimes referred to as protective equipment) has often been utilized on an antecedent basis to reduce the possibility of pica, while the latter has been used contingent upon pica behavior. Mechanical restraints range from a camisole (Rojahn, Schroeder, & Mulick, 1980) to canvas arm sleeves and a helmet with a facemask (LeBlanc et al., 1997). When mechanical restraints prevent or inhibit pica, the down side is that they may also inhibit more desirable behaviors, such as social interaction and toy play (LeBlanc et al., 1997).

Like mechanical restraint, brief physical restraint procedures have many variations. Physical restraint, in the form hand holding (Bogart et al., 1995), restriction of arms to the side or behind the back (Bucher, Reykdal, & Albin, 1976), and basket hold procedures (Fisher et al., 1994; Northup, Fisher, Kahang, & Harrell, 1997), has been an effective treatment component for pica. These studies have typically used a very brief duration for restraint (i.e., 30 s or less), which follows a finding by Winton and Singh (1983) that restraint duration as short as 10 s may be more effective than longer restraint durations (i.e., 30 s and 3 min). Although the physical demands of restraint protocols may make them impractical, Northup et al. (1997) demonstrated that even lean schedules of physical restraint (i.e., VR 2 and VR 4 schedules) may be sufficient to reduce pica to low levels.

Overcorrection

Few recent studies have incorporated overcorrection as a treatment component for pica despite strong evidence of its effectiveness. Williams and McAdam (2012) surmised that the decline in usage may be due, in part, to the physical demands and necessary oversight required on the part of the treatment implementer. A positive punishment procedure, overcorrection for pica, generally involves a contingent demand for pica behavior. The contingent demand typically is related to pica and requires the individual to either practice a more appropriate behavior or improve the environment where it happened. The most frequent corrective behavior documented has been oral hygiene tasks (Matson, Stephens, & Smith, 1978; Mulick, Barbour, Schroeder, & Rojahn, 1980; Singh & Bakker, 1984; Singh & Winton, 1985). Foxx and Martin (1975) published the first example of overcorrection for pica with four adults with ID who engaged in coprophagia or cigarette pica. Participants were required to complete an extensive overcorrection procedure that lasted up to 30 min to complete fully contingent upon pica behavior. Beginning with the discharge of any pica items from the mouth, the participant was expected to clean his or her mouth and gums with a toothbrush soaked in antiseptic cleaner, wash his or her hands in a bathroom sink, and clean the area where he or she engaged in pica (e.g., empty trashcans, flush toilets). This procedure has been noted for the large and rapid reduction in pica.

Aversive Presentations

Another positive punishment strategy regularly cited in pica treatment literature is the use of contingent aversive stimuli. It could be argued that many of the other punishment procedures described above involve contingent presentation of aversive stimuli; however, in these studies, a known aversive sensory experience, often in the form of a physical substance, is presented subject to pica behavior and is often accompanied by a reprimand (e.g., "No!"). Most studies using aversives have treated pica using a substance with an aversive taste, such as lemon juice (Paisey & Whitney, 1989), tapioca pudding (Ferreri, Tamm, & Wier, 2006), and hot sauce (Goh et al., 1999). Water mist sprayed in the face has been evaluated multiple times, but with mixed results (Fisher et al., 1994; Goh et al., 1999; Paisey & Whitney, 1989; Rojahn et al., 1987). Rapp et al. (2001) failed to obtain adequate reductions in pica for a young girl with autism using a variety of reinforcement- and punishment-based approaches, but were able to obtain suppression using contingent loud auditory tones presented for 2–3 s each time.

Many studies employing aversives have failed to report how they selected the aversive stimuli used for treatment. By not conducting an empirical evaluation of possible aversives, prolongation of treatment may occur when something believed to be aversive is not experienced as such by the individual engaging in pica. In some cases, what is believed to be aversive may turn out to be perceived as pleasurable (and thus potentially reinforcing). Fisher et al. (1994) used a two-step process to identify potential punishers for treatment of pica: First, a stimulus avoidance assessment was conducted to identify procedures that were highly, moderately, or minimally aversive as measured by negative vocalizations and escape/avoidance behaviors by the participant. This was followed by a punisher assessment using a multielement design to test stimuli from each level of aversiveness as a punisher for pica behavior. Through this assessment, the authors were able to identify effective punishers and avoid spending time using less effective or ineffective stimuli.

Biomedical Treatments for Pica

Behavioral treatments dominate the research on pica and especially for pica in ID/DD populations. Given the large number of studies demonstrating effective behavioral treatment of pica, it has been suggested that non-behavioral treatments may not be a worthy interest to clinicians based upon a paucity of research, poorly designed experimentation, and equivocal outcomes (Matson, Hattier, Belva, & Matson, 2013). Nonetheless, research on nutritional and pharmacological interventions exists and warrants further review and additional research, particularly for difficult-to-treat cases of pica. In the following sections, existing research on nutritional treatments is briefly reviewed, followed by pharmacological treatments of pica. It should be noted that in this review of biomedical treatments, no attempt has been made to distinguish between studies exclusively involving ID/DD populations and those that do not as most research on biomedical treatments does not clearly delineated these groups.

Nutritional Treatments

A number of studies have indicated that pica is more likely to occur in individuals with nutritional deficiencies, particularly involving the minerals iron and zinc (Lofts et al., 1990; Miao et al., 2015; Swift, Paquette, Davidson, & Saeed, 1999). As a result, research studies have evaluated nutritional supplementation as a treatment for pica. Unfortunately, most studies demonstrating reductions of pica using nutritional supplementation have had inadequate experimental design, and they rarely fully suppressed pica. Lofts et al. (1990) added zinc supplementation to the diets of adults living in an institution who exhibited pica and low zinc plasma levels. The added zinc resulted in substantial decreases in average pica over a 2-week period. Two controlled double-blind studies of iron supplementation on geophagy (Gutelius et al., 1962; Nchito et al., 2004) failed to show a significant effect on pica behavior.

The effects of supplementation with multivitamins on pica without respect to any specifically identified nutritional deficiency have been evaluated in two studies. Using a single-case design (i.e., BAB reversal), Pace and Toyer (2000) demonstrated that a liquid multivitamin resulted in decreased pica for a young girl with ID. Bugle and Rubin (1993) used a nutritional supplement with amino acids and other essential nutrients for three individuals with coprophagia. The results showed modest improvement in pica over baseline.

Pharmacological Treatments

Unlike treatments involving nutritional supplementation, pharmacological treatments require a prescription by a licensed physician and are more likely to be associated with unwanted side effects. Any medications prescribed for pica are done so "off label" as there is currently no FDA-approved drug for the treatment of pica. Many published accounts of successful treatment of pica with medication have been case reports with limited accompanying data for review and with participants exhibiting accompanying comorbid psychiatric diagnoses. Such reports have indicated some success with a range of medications, including central nervous stimulants (Hergüner & Hergüner, 2010), atypical antipsychotics (Harada, Yamamoto, & Saito, 2006), selective serotonin reuptake inhibitors (Bhatia & Gupta, 2009), and alpha-adrenergic agonists (Saddichha, Vibha, & Vishnuvardhan, 2012).

Interesting and sometimes conflicting results have come from case reports. For example, in one case report, olanzapine, an atypical antipsychotic, effectively treated pica in an adult woman with ID (Lerner, 2008), but the same drug was singled out as a possible cause of pica in an adult male with schizophrenia (Huang, Shiah, Lee, Tzang, & Chang, 2010) and an adult female with schizophrenia (Chawla, Charan, Kumar, & Pattanayak, 2016). In another case report, Brahm, Farmer, and Brown (2006) described successful treatment of cigarette pica in a 50-year-old male with profound ID with bupropion. Interestingly, bupropion, a norepinephrine-dopamine reuptake inhibitor, has been shown to be a highly effective component of smoking cessation programs (Jorenby, 2002).

One of the few rigorous studies on pharmacological interventions for pica was conducted by Singh et al. (1994). Using a double-blind, placebo-controlled single-case design, an evaluation was carried out on the separate effects of thioridazine (a dopamine antagonist) and methylphenidate (a stimulant that increases dopamine) on pica for three adolescents with profound ID. All three participants had previously failed to respond to "well-designed" behavioral treatments. The use of thioridazine led to the highest levels of pica for all participants, while methylphenidate usage was associated with the lowest levels of pica for all three. The authors hypothesized that decreased dopaminergic transmission leads to increases in pica in some individuals, which their data support.

Summary and Conclusions

Pica is a difficult-to-treat and potentially life-threatening disorder that is commonly observed in the ID/DD population. Thus, it warrants a great deal of attention from caregivers, treatment providers, and researchers alike. Caregivers of individuals with ID/DD must be aware of the relatively high prevalence rate of pica in this population and be vigilant of the signs and symptoms. For particularly dangerous pica, even momentary disregard may lead to tragic outcomes. The treatment provider should acknowledge possible biomedical underpinnings (e.g., nutrient deficiencies), but be inclined to develop treatments based upon behavior analytic principles. They are faced with the daunting task of choosing among a broad range of treatment components found in the research literature. Moreover, it is essential for treatment providers to consider the practicality of treatment and whether a potential treatment would be acceptable if it leads only to reduced pica behavior, but not full suppression. There are many knowledge gaps that researchers must fill. Of chief importance is ascertaining the cause(s) of pica. A number of theories have been proffered, but most give an incomplete account or fail to hold adequate supporting evidence. A definitive account for the basis of pica would likely contribute to preventive approaches and more effective treatments. Treatment must also be a focus of continued research. Despite a large number of published treatment studies, especially in the field of applied behavior analysis, the majority have been of the small-n variety and rarely with replication. Furthermore, many research studies have utilized treatment packages incorporating multiple treatment components and rely on punishment-based approaches, which are traditionally associated with weaker social validity.

References

Aggleton, J. P., & Passingham, R. E. (1981). Syndrome produced by lesions of the amygdala in monkeys (Macaca mulatta). *Journal of Comparative and Physiological Psychology, 95*(6), 961–977.

Ali, Z. (2001). Pica in people with intellectual disability: A literature review of aetiology, epidemiology and complications. *Journal of Intellectual & Developmental Disability, 26*, 205–215.

American Psychiatric Association. (2013). *The diagnostic and statistical manual* (5th ed.). Washington: Author.

Arbiter, E. A., & Black, D. (1991). Pica and iron-deficiency anaemia. *Child Care Health and Development, 17*(4), 231–234.

Ashworth, M., Martin, L., & Hirdes, J. P. (2008). Prevalence and correlates of pica among adults with intellectual disability in institutions. *Journal of Mental Health Research in Intellectual Disabilities, 1*, 176–190.

Association for Behavior Analysis International. (2010). Statement on Restraint and Seclusion, 2010. https://www.abainternational.org/about-us/policies-and-positions/restraint-and-seclusion,2010.aspx.

Azrin, N. H., & Holz, W. C. (1966). Punishment. In W. K. Honig (Ed.), *Operant behavior: Areas of research and application* (pp. 380–447). New York: Appleton-Century-Crofts.

Baker, D. J., Valenzuela, S., & Wieseler, N. A. (2005). Naturalistic inquiry and treatment of coprophagia in one individual. *Journal of Developmental and Physical Disabilities, 17*, 361–367.

Barkin, S., Scheindlin, B., Ip, E. H., Richardson, I., & Finch, S. (2007). Determinants of parental discipline practices: A national sample from primary care practices. *Clinical Pediatrics, 46*(1), 64–69. https://doi.org/10.1177/0009922806292644

Barltop, D. (1966). The prevalence of pica. *American Journal of Diseases in Children, 112*, 116–123.

Bhatia, M. S., & Gupta, R. (2009). Pica responding to SSRI: An OCD spectrum disorder? *World Journal of Biological Psychiatry, 10*(4 Pt 3), 936–938. https://doi.org/10.1080/15622970701308389

Bogart, L. C., Piersel, W. C., & Gross, E. J. (1995). The long-term treatment of life-threatening pica: A case study of a woman with profound mental retardation living in an applied setting. *Journal of Developmental and Physical Disabilities, 7*(1), 39–50. https://doi.org/10.1007/BF02578713

Brahm, N. C., Farmer, K. C., & Brown, R. C. (2006). Pica episode reduction following initiation of bupropion in a developmentally disabled adult. *Annals of Pharmacotherapy, 40*(11), 2075–2076. https://doi.org/10.1345/aph.1H232

Bucher, B., Reykdal, B., & Albin, J. (1976). Brief physical restraint to control Pica in retarded children. *Journal of Behavior Therapy and Experimental Psychiatry, 7*(2), 137–140. https://doi.org/10.1016/0005-7916(76)90071-9

Bugle, C., & Rubin, H. B. (1993). Effects of a nutritional supplement on coprophagia: A study of three cases. *Research in Developmental Disabilities, 14*, 445–456.

Call, N. A., Simmons, C. A., Mevers, J. E., & Alvarez, J. P. (2015). Clinical outcomes of behavioral treatments for Pica in children with developmental disabilities. *Journal of Autism and Developmental Disorders, 45*(7), 2105–2114. https://doi.org/10.1007/s10803-015-2375-z

Cavdar, A. O., Arcasoy, A., Cin, S., Babacan, E., & Gozdasoglu, S. (1983). Geophagia in Turkey: Iron and zinc deficiency, iron and zinc absorption studies and response to treatment with zinc in geophagia cases. *Progress in Clinical and Biological Research, 129*, 71–97.

Chawla, N., Charan, D., Kumar, S., & Pattanayak, R. D. (2016). Pica associated with initiation of atypical antipsychotic drugs: Report of two cases. *Psychiatry and Clinical Neurosciences, 70*(8), 363–364. https://doi.org/10.1111/pcn.12408

Chisholm, J. C., & Martin, H. I. (1981). Hypozincemia, ageusia, dysosmia, and toilet tissue pica. *Journal of the National Medical Association, 73*(2), 163–164.

Clark, B., Vandermeer, B., Simonetti, A., & Buka, I. (2010). Is lead a concern in Canadian autistic children? *Paediatrics & Child Health, 15*, 17–22.

Collins, M. S., & Cornish, K. (2002). A survey of the prevalence of stereotypy, self-injury and aggression in children and young adults with Cri du Chat syndrome. *Journal of Intellectual Disability Research, 46*, 133–140.

Danford, D. E., & Huber, A. E. (1982). Pica among mentally retarded adults. *American Journal of Mental Deficiency, 87*, 141–146.

Dimitropoulos, A., Feurer, I. D., Roof, E., Stone, W., Butler, M. G., Sutcliffe, J., & Thompson, T. (2000). Appetitive behavior, compulsivity, and neurochemistry in Prader-Willi syndrome. *Mental Retardation and Developmental Disabilities Research Reviews, 6*(2), 125–130. https://doi.org/10.1002/1098-2779(2000)6:2<125::aid-mrdd6>3.0.co;2-t

Donnelly, D. R., & Olczak, P. V. (1990). The effect of differential reinforcement of incompatible behaviors (DRI) on pica for cigarettes in persons with intellectual disability. *Behavior Modification, 14*(1), 81–96. https://doi.org/10.1177/01454455900141006

Dykens, E. M., Leckman, J. F., & Cassidy, S. B. (1996). Obsessions and compulsions in Prader-Willi syndrome. *Journal of Child Psychology and Psychiatry, 37*(8), 995–1002.

Dykens, E. M. (2000). Contaminated and unusual food combinations: What do people with Prader-Willi syndrome choose? *Mental Retardation, 38*, 163–171. doi:10.1352/0047-6765(2000)038<0163:CAUFCW>2.0.CO;2

Falcomata, T. S., Roane, H. S., & Pabico, R. R. (2007). Unintentional stimulus control during the treatment of Pica displayed by a young man with autism. *Research in Autism Spectrum Disorders, 1*(4), 350–359.

Favell, J. E., McGimsey, J. F., & Schell, R. M. (1982). Treatment of self-injury by providing alternate sensory activities. *Analysis & Intervention in Developmental Disabilities, 2*(1), 83–104. https://doi.org/10.1016/0270-4684(82)90007-6

Fawcett, E. J., Fawcett, J. M., & Mazmanian, D. (2016). A meta-analysis of the worldwide prevalence of pica during pregnancy and the postpartum period. *International Journal of Gynaecology and Obstetrics, 133*(3), 277–283. https://doi.org/10.1016/j.ijgo.2015.10.012

Finney, J. W., Russo, D. C., & Cataldo, M. F. (1982). Reduction of pica in young children with lead poisoning. *Journal of Pediatric Psychology, 7*(2), 197–207.

Fisher, W. W., Piazza, C. C., Bowman, L. G., Kurtz, P. F., Sherer, M. R., & Lachman, S. R. (1994). A preliminary evaluation of empirically derived consequences for the treatment of pica. *Journal of Applied Behavior Analysis, 27*(3), 447–457. https://doi.org/10.1901/jaba.1994.27-447

Foxx, R. M., & Martin, E. D. (1975). Treatment of scavenging behavior (coprophagy and pica) by overcorrection. *Behavior Research and Therapy, 13*(2–3), 153–162.

Ferreri, S. J., Tamm, L., & Wier, K. G. (2006). Using food aversion to decrease severe pica by a child with autism. *Behavior Modification, 30*, 456–471.

Geissler, P. W., Prince, R. J., Levene, M., Poda, C., Beckerleg, S. E., Mutemi, W., & Shulman, C. E. (1999). Perceptions of soil-eating and anaemia among pregnant women on the Kenyan coast. *Social Science & Medicine, 48*, 1069–1079.

Ghaziuddin, N., & McDonald, C. (1985). A clinical study of adult coprophagics. *British Journal of Psychiatry, 147*, 312–313.

Goh, H. L., Iwata, B. A., & Kahng, S. W. (1999). Multicomponent assessment and treatment of cigarette pica. *Journal of Applied Behavior Analysis, 32*(3), 297–316. https://doi.org/10.1901/jaba.1999.32-297

Gundacker, C., Kutalek, R., Glaunach, R., Deweis, C., Hengstschlager, M., & Prinz, A. (2017). Geophagy during pregnancy: Is there a health risk for infants? *Environmental Research, 156*, 145–147. https://doi.org/10.1016/j.envres.2017.03.028

Gundogar, D., Demir, S. B., & Eren, I. (2003). Is pica in the spectrum of obsessive-compulsive disorders? *General Hospital Psychiatry, 25*(4), 293–294.

Gutelius, M. F., Millican, F. K., Layman, E. M., Cohen, G. J., & Dublin, C. C. (1962). Nutritional studies of children with pica. I controlled study evaluating nutritional status. *Pediatrics, 29*, 1012–1023.

Hagopian, L. P., & Adelinis, J. D. (2001). Response blocking with and without redirection for the treatment of pica. *Journal of Applied Behavior Analysis, 34*(4), 527–530. https://doi.org/10.1901/jaba.2001.34-527

Hagopian, L. P., González, M. L., Rivet, T. T., Triggs, M., & Clark, S. B. (2011). Response interruption and differential reinforcement of alternative behavior for the treatment of pica. *Behavioral Interventions, 26*(4), 309–325. https://doi.org/10.1002/bin.339

Hagopian, L. P., Rooker, G. W., & Rolider, N. U. (2011). Identifying empirically supported treatments for pica in individuals with intellectual disabilities. *Research in Developmental Disabilities: A Multidisciplinary Journal, 32*, 2114–2120.

Hall, S. S. (2005). Comparing descriptive, experimental, and informant-base assessments of problem behaviors. *Research in Developmental Disabilities, 26*, 1514–1526.

Harada, K. I., Yamamoto, K., & Saito, T. (2006). Effective treatment of coprophagia in a patient with schizophrenia with the novel atypical antipsychotic drug perospirone. *Pharmacopsychiatry, 39*(3), 113. https://doi.org/10.1055/s-2006-941487

Hergüner, S., & Hergüner, A. S. (2010). Pica in a child with attention deficit hyperactivity disorder and successful treatment with methylphenidate. *Progress in Neuropsychopharmacology and Biological Psychiatry, 34*(6), 1155–1156. https://doi.org/10.1016/j.pnpbp.2010.06.005

Hove, O. (2004). Prevalence of eating disorders in adults with mental retardation living in the community. *American Journal of Mental Retardation, 109*(6), 501–506. https://doi.

org/10.1352/0895-8017(2004)109<501:poedia>2.0.co;2

Huang, J. H., Shiah, I. S., Lee, W. K., Tzang, R. F., & Chang, K. J. (2010). Olanzapine-associated pica in a schizophrenia patient. *Psychiatry and Clinical Neurosciences, 64*(4), 444. https://doi.org/10.1111/j.1440-1819.2010.02099.x

Ing, A. D., Roane, H. S., & Veenstra, R. A. (2011). Functional analysis and treatment of coprophagia. *Journal of Applied Behavior Analysis, 44*(1), 151–155. https://doi.org/10.1901/jaba.2011.44-151

Iwata, B. A., Dorsey, M. F., Slifer, K. E., Bauman, K. E., & Richman, G. S. (1982/1994). Toward a functional analysis of self-injury. *Journal of Applied Behavior Analysis, 26*, 197–206.

Jawed, S. H., Krishnan, V. H., Prasher, V. P., & Corbett, J. A. (1993). Worsening of pica as a symptom of depressive illness in a person with severe mental handicap. *The British Journal of Psychiatry, 162*, 835–837. https://doi.org/10.1192/bjp.162.6.835

Johnson, C. R., Hunt, F. M., & Siebert, M. J. (1994). Discrimination training in the treatment of pica and food scavenging. *Behavior Modification, 18*(2), 214–229. https://doi.org/10.1177/01454455940182005

Jorenby, D. (2002). Clinical efficacy of bupropion in the management of smoking cessation. *Drugs, 62*(Suppl 2), 25–35.

Josephs, K. A., Whitwell, J. L., Parisi, J. E., & Lapid, M. I. (2016). Coprophagia in neurologic disorders. *Journal of Neurology, 263*(5), 1008–1014. https://doi.org/10.1007/s00415-016-8096-1

Kalfus, G. R., Fisher-Gross, S., Marvullo, M. A., & Nau, P. A. (1987). Outpatient treatment of pica in a developmentally delayed child. *Child & Family Behavior Therapy, 9*(3–4), 49–63. https://doi.org/10.1300/J019v09n03_04

Kern, L., Starosta, K., & Adelman, B. E. (2006). Reducing pica by teaching children to exchange inedible items for edibles. *Behavior Modification, 30*(2), 135–158. https://doi.org/10.1177/0145445505283414

Kinnell, H. G. (1985). Pica as a feature of autism. *British Journal of Psychiatry, 147*, 80–82.

LeBlanc, L. A., Piazza, C. C., & Krug, M. A. (1997). Comparing methods for maintaining the safety of a child with pica. *Research in Developmental Disabilities, 18*, 215–220.

Lerman, D. C., & Vorndran, C. M. (2002). On the status of knowledge for using punishment: Implications for treating behavior disorders. *Journal of Applied Behavior Analysis, 35*(4), 431–464. https://doi.org/10.1901/jaba.2002.35-431

Lerner, A. J. (2008). Treatment of pica behavior with olanzapine. *CNS Spectrums, 13*(1), 19.

Lilly, R., Cummings, J. L., Benson, D. F., & Frankel, M. (1983). The human Kluver-Bucy syndrome. *Neurology, 33*(9), 1141–1145.

Lofts, R. H., Schroeder, S. R., & Maier, R. H. (1990). Effects of serum zinc supplementation on pica behavior of persons with mental retardation. *American Journal of Mental Retardation, 95*(1), 103–109.

LoVullo, S. V., & Matson, J. L. (2009). Comorbid psychopathology in adults with autism spectrum disorders and intellectual disabilities. *Research in Developmental Disabilities, 30*, 1288–1296.

Luiselli, J. K. (1996). Pica as obsessive-compulsive disorder. *Journal of Behavior Therapy and Experimental Psychiatry, 27*(2), 195–196.

Lerman, D.C., & Iwata, B. (1996). A methodology for distinguishing between extinction and punishment effects associated with response blocking. *Journal of Applied Behavior Analysis, 29*(2):231–233

Lydon, S., Healy, O., Moran, L., & Foody, C. (2015) A quantitative examination of punishment research. *Research in Developmental Disabilities 36*:470–484

Mace, F. C., & Knight, D. (1986). Functional analysis and treatment of severe pica. *Journal of Applied Behavior Analysis, 19*(4), 411–416. https://doi.org/10.1901/jaba.1986.19-411

Matson, J. L., Belva, B., Hattier, M. A., & Matson, M. L. (2011). Pica in persons with developmental disabilities: Characteristics, diagnosis, and assessment. *Research in Autism Spectrum Disorders, 5*, 1459–1464.

Matson, J. L., Hattier, M. A., Belva, B., & Matson, M. L. (2013). Pica in persons with developmental disabilities: Approaches to treatment. *Research in Developmental Disabilities, 34*(9), 2564–2571. https://doi.org/10.1016/j.ridd.2013.05.018

Matson, J. L., Stephens, R. M., & Smith, C. (1978). Treatment of self-injurious behaviour with overcorrection. *Journal of Mental Deficiency Research, 22*(3), 175–178.

McAdam, D. B., Breidbord, J., Levine, M., & Williams, D. E. (2012). *Pica Handbook of evidence-based practice in clinical psychology, Vol 1: Child and adolescent disorders* (pp. 303–321). Hoboken, NJ: John Wiley & Sons.

McCord, B. E., Grosser, J. W., Iwata, B. A., & Powers, L. J. (2005). An analysis of response-blocking parameters in the prevention of pica. *Journal of Applied Behavior Analysis, 38*(3), 391–394. https://doi.org/10.1901/jaba.2005.92-04

McLean, E., Cogswell, M., Egli, I., Wojdyla, D., & de Benoist, B. (2009). Worldwide prevalence of anaemia, WHO vitamin and mineral nutrition information system, 1993-2005. *Public Health Nutrition, 12*(4), 444–454. https://doi.org/10.1017/s1368980008002401

Miao, D., Young, S. L., & Golden, C. D. (2015). A meta-analysis of pica and micronutrient status. *American Journal of Human Biology, 27*, 84–93.

Mitteer, D. R., Romani, P. W., Greer, B. D., & Fisher, W. W. (2015). Assessment and treatment of pica and destruction of holiday decorations. *Journal of Applied Behavior Analysis, 48*(4), 912–917. https://doi.org/10.1002/jaba.255

Morris, C., Hope, R., & Fairburn, C. (1989). Eating Habits in Dementia: A Descriptive Study. *British Journal of Psychiatry, 154*(6), 801–806. https://doi.org/10.1192/bjp.154.6.801

Mulick, J. A., Barbour, R., Schroeder, S. R., & Rojahn, J. (1980). Overcorrection of pica in two profoundly retarded adults: Analysis of setting effects, stimulus, and response generalization. *Applied Research in Mental Retardation, 1*(3–4), 241–252.

Nchito, M., Geissler, P. W., Mubila, L., Friis, H., & Olsen, A. (2004). Effects of iron and multimicronutrient supplementation on geophagy: A two-by-two factorial study among Zambian schoolchildren in Lusaka. *Transactions of the Royal Society of Tropical Medicine and Hygiene, 98*, 218–227.

Northup, J., Fisher, W., Kahang, S. W., & Harrell, R. (1997). An assessment of the necessary strength of behavioral treatments for severe behavior problems. *Journal of Developmental and Physical Disabilities, 9*(1), 1–16. https://doi.org/10.1023/A:1024984526008

Pace, G. M., & Toyer, E. A. (2000). The effect of a vitamin supplement on the pica of a child with severe mental retardation. *Journal of Applied Behavior Analysis, 33*, 619–622.

Paisey, T. J., & Whitney, R. B. (1989). A long-term case study of analysis, response suppression, and treatment maintenance involving life-threatening pica. *Behavioral Residential Treatment, 4*(3), 191–211. https://doi.org/10.1002/bin.2360040304

Paniagua, F. A., Braverman, C., & Capriotti, R. M. (1986). Use of a treatment package in the management of a profoundly mentally retarded girl's pica and self-stimulation. *American Journal of Mental Deficiency, 90*, 550–557.

Piazza, C. C., Fisher, W. W., Hanley, G. P., LeBlanc, L. A., Worsdell, A. S., Lindauer, S. E., & Keeney, K. M. (1998). Treatment of pica through multiple analyses of its reinforcing functions. *Journal of Applied Behavior Analysis, 31*(2), 165–189. https://doi.org/10.1901/jaba.1998.31-165

Piazza, C. C., Hanley, G. P., & Fisher, W. W. (1996). Functional analysis and treatment of cigarette pica. *Journal of Applied Behavior Analysis, 29*(4), 437–449.; quiz 449-450. https://doi.org/10.1901/jaba.1996.29-437

Piazza, C. C., Roane, H. S., Keeney, K. M., Boney, B. R., & Abt, K. A. (2002). Varying response effort in the treatment of pica maintained by automatic reinforcement. *Journal of Applied Behavior Analysis, 35*(3), 233–246. https://doi.org/10.1901/jaba.2002.35-233

Rapp, J. T., Dozier, C. L., & Carr, J. E. (2001). Functional assessment and treatment of pica: A single-case experiment. *Behavioral Interventions, 16*, 111–125.

Ricciardi, J. N., Luiselli, J. K., Terrill, S., & Reardon, K. (2003). Alternative response training with contingent practice as intervention for pica in a school setting. *Behavioral Interventions, 18*, 219–226.

Rojahn, J., Schroeder, S., & Mulick, J. A. (1980). Ecological assessment of self-protective devices in three profoundly retarded adults. *Journal of Autism and Developmental Disorders, 10*, 59–66.

Rojahn, J., McGonigle, J. J., Curcio, C., & Dixon, M. J. (1987). Suppression of pica by water mist and aromatic ammonia: A comparative analysis. *Behavior Modification, 11*, 65–74.

Saddichha, S., Vibha, P., & Vishnuvardhan, G. (2012). Sapophagia (compulsive soap eating) and attention-deficit/hyperactivity disorder in a child responsive to clonidine. *Journal of Clinical Psychopharmacology, 32*(2), 291–291. https://doi.org/10.1097/JCP.0b013e3182499ad2

Schmidt, J., Long, A., Goetzel, A., Tung, C., Pizarro, E., Phillips, C., & Hausman, N. (2017). Decreasing Pica attempts by manipulating the environment to support prosocial behavior. *Journal of Developmental & Physical Disabilities, 29*(5), 683–697. https://doi.org/10.1007/s10882-017-9548-y

Singh, N. N., & Bakker, L. W. (1984). Suppression of pica by overcorrection and physical restraint: A comparative analysis. *Journal of Autism and Developmental Disorders, 14*(3), 331–341.

Singh, N. N., Ellis, C. R., Crews, W. D., & Singh, Y. N. (1994). Does diminished dopaminergic neurotransmission increase pica? *Journal of Child and Adolescent Psychopharmacology, 4*, 93–99.

Singh, N. N., & Winton, A. S. (1984). Effects of a screening procedure on pica and collateral behaviors. *Journal of Behavior Therapy and Experimental Psychiatry, 15*(1), 59–65.

Singh, N. N., & Winton, A. S. (1985). Controlling pica by components of an overcorrection procedure. *American Journal of Mental Deficiency, 90*(1), 40–45.

Sinha, M., & Mallick, A. K. (2010). Pica in the backdrop of psychiatric disorders: A complex etiology. *International Medical Journal, 17*(1), 21–23.

Slocum, S. K., Mehrkam, L. R., Peters, K. P., & Vollmer, T. R. (2017). Using differential reinforcement of a discard response to treat pica. *Behavioral Interventions, 32*(3), 234–241. https://doi.org/10.1002/bin.1483

Smith, M. D. (1987). Treatment of pica in an adult disabled by autism by differential reinforcement of incompatible behavior. *Journal of Behavior Therapy and Experimental Psychiatry, 18*(3), 285–288. https://doi.org/10.1016/0005-7916(87)90012-7

Smith, S., Branford, D., Collacott, R. A., Cooper, S. A., & McGrother, C. (1996). Prevalence and cluster typology of maladaptive behaviors in a geographically defined population of adults with learning disabilities. *The British Journal of Psychiatry, 169*, 219–227.

Stein, D. J., Bouwer, C., & van Heerden, B. (1996). Pica and the obsessive-compulsive spectrum disorders. *South African Medical Journal, 86*(12 Suppl), 1586–1588. 1591-1582.

Sturmey, P., & Williams, D. E. (2016). *Pica in individuals with developmental disabilities*. Cham, Switzerland: Springer International Publishing.

Swift, I., Paquette, D., Davidson, K., & Saeed, H. (1999). Pica and trace metal deficiencies in adults with developmental disabilities. *British Journal of Developmental Disabilities, 45*, 111–117.

T. E. C. (1969). The origin of the word *pica*. *Pediatrics, 44*(4), 548–548.

Tewari, S., Krishnan, V. H. R., Valsalan, V. C., & Roy, A. (1995). Pica in a learning disability hospital: A clinical survey. *British Journal of Developmental Disabilities, 41*, 13–22.

Vollmer, T. R. (1994). The concept of automatic reinforcement: Implications for behavioral research in developmental disabilities. *Research in Developmental Disabilities, 15*, 187–207.

Williams, D. E., Kirkpatrick-Sanchez, S., Enzinna, C., Dunn, J., & Borden-Karasack, D. (2009). The clinical management and prevention of pica: A retrospective follow-up of 41 individuals with intellectual disabilities and pica. *Journal of Applied Research in Intellectual Disabilities, 22*(2), 210–215.

Williams, D. E., & McAdam, D. (2012). Assessment, behavioral treatment, and prevention of pica: Clinical guidelines and recommendations for practitioners. *Research in Developmental Disabilities, 33*(6), 2050–2057. https://doi.org/10.1016/j.ridd.2012.04.001

Winton, A. S., & Singh, N. N. (1983). Suppression of pica using brief-duration physical restraint. *Journal of Mental Deficiency Research, 27*(Pt 2), 93–103.

Young, S. L. (2010). Pica in pregnancy: New ideas about an old condition. *Annual Review of Nutrition, 30*, 403–422. https://doi.org/10.1146/annurev.nutr.012809.104713

Young, S. L., Sherman, P. W., Lucks, J. B., & Pelto, G. H. (2011). Why on earth?: Evaluating hypotheses about the physiological functions of human geophagy. *Quarterly Review of Biology, 86*(2), 97–120.

Zeitlin, S. B., & Polivy, J. (1995). Coprophagia as a manifestation of obsessive-compulsive disorder: A case report. *Journal of Behavior Therapy and Experimental Psychiatry, 26*(1), 57–63.

Feeding Disorders

34

Mitch Fryling, Kimberley L. M. Zonneveld,
Aline Zaragoza, and Catherine L. McHugh

Introduction

Feeding disorders encompass a variety of behavioral topographies and affect a wide range of people. Prevalence rates for feeding problems vary across definitions and populations. While feeding problems occur among typically developing populations (e.g., Benjasuwantep, Chaithirayanon, & Eiamudomkan, 2013), feeding disorders are much more common with individuals with disabilities (see Kerwin, 1999, Munk & Repp, 1994; Volkert & Piazza, 2012 for overviews). Several authors have also noted an increased prevalence of feeding disorders with individuals on the autism spectrum specifically, with some estimates being as high as 90% (e.g., Kodak & Piazza, 2008; Schreck, Williams, & Smith, 2004; Twachtman-Reilly, Amaral, & Zebrowski, 2008). Importantly, feeding disorders are not limited to young children and may occur in various forms throughout the life span.

Feeding disorders are also complex and may be related to various medical conditions (Field, Garland, & Williams, 2003). For example, a child may begin to avoid eating as a result of painful experiences associated with eating caused by gastroesophageal reflux disease. Alternatively, an individual may have difficulty swallowing and avoid eating for reasons related to this. As these examples highlight, it is important that an interdisciplinary assessment be conducted to identify possible medical factors that may need to be considered prior to implementing a behavioral intervention. In addition to the complex range of factors that can be involved in feeding disorders, feeding disorders are also diagnosed in various ways, which can make it difficult to identify exactly when a problem exists (see Piazza, 2008). This chapter provides an overview of the behavior analytic literature on feeding problems. We begin by providing a brief overview of the behavioral position in general, followed by a consideration of common intervention components, and conclude with more detailed overviews of the literature on two relatively under-researched areas, packing and rapid eating.

Taking a Behavior Analytic Approach

A variety of disciplines and theoretical approaches have addressed feeding disorders. Several behavior analytic strategies have been demonstrated to be effective, with others showing promise (see Bachmeyer, 2009; Kerwin, 1999; Volkert & Piazza, 2012). In addition, a

---

M. Fryling (✉) · A. Zaragoza
California State University, Los Angeles, CA, USA
e-mail: Mitchell.Fryling2@calstatela.edu

K. L. M. Zonneveld · C. L. McHugh
Brock University, St. Catharines, ON, Canada

J. L. Matson (ed.), *Handbook of Intellectual Disabilities*, Autism and Child Psychopathology Series, https://doi.org/10.1007/978-3-030-20843-1_34

small number of researchers have begun to compare behavior analytic strategies to those advocated for in other disciplines. For example, Addison et al. (2012) compared a behavioral intervention to sensory integration intervention with two children with feeding problems and found that the behavioral intervention was more effective toward improving food consumption and decreasing inappropriate mealtime behavior (IMB). Similarly, Peterson, Piazza, and Volkert (2016) compared a behavioral intervention to a sensory approach with six children with feeding problems and found that food consumption only increased when the behavioral intervention was implemented. While more research comparing different types of interventions is certainly needed, there is now considerable support for assessment and intervention strategies based on the principles of behavior analysis.

Consistent with behavioral approaches more generally, behavior analytic approaches to feeding problems focus on understanding how factors in the environmental context are functionally related to behaviors considered to be socially important. This sort of assessment, often termed *Functional Behavioral Assessment* in behavior analytic research and practice, is foundational to all intervention work in behavior analysis (more on this below). Behavioral approaches are also committed to objective measures of socially relevant target behaviors over time (Baer, Wolf, & Risley, 1968). For this reason, behavioral work in the area of feeding disorders involves the objective measurement of important dimensions of feeding behavior. Common targets within the feeding literature often include food acceptance (e.g., the number of times food passes the plane of an individual's lips), food consumption (e.g., the number of times the individual swallows a bite of food), and various topographies of IMB (e.g., throwing food items, yelling, hitting the spoon). A variety of other behaviors involved in eating have also been targeted, especially as prerequisite feeding skills are increasingly considered in the research literature. For example, recent research has targeted chewing as a dependent variable as this is indeed a critical skill involved in eating (Volkert, Peterson, Zeleny, & Piazza, 2014; Volkert, Piazza, Vaz, & Frese, 2013). Self-feeding has also been studied, which is an important step toward developing independence more generally (e.g., Rivas et al., 2014. Several behavior analysts have also conducted research on rumination (i.e., bringing up previously swallowed food; see Lang et al., 2011, Luiselli, 2015). As we will elaborate on below, behavior analysts have also focused on the time between taking bites when bites are taken too quickly (i.e., to reduce rapid eating) and packing or storing food in one's mouth. As these examples highlight, behavior analysts approach feeding problems with a focus on a range of objective measures of socially significant target behaviors. Again, such measurement is fundamental to the identification of meaningful target behaviors and the ongoing assessment of intervention effectiveness. In the following section we provide an overview of behavior analytic assessment and intervention research on feeding disorders. We begin by reviewing research on the functional assessment of IMB.

Functional Assessment

Indirect Assessments. As with the functional assessment of other topographies of problem behavior, the assessment of IMB takes three general forms: indirect assessments, descriptive assessments, and functional analysis. Indirect assessment of IMB typically involves questionnaires and rating scales in which respondents are asked to respond to questions related to numerous variables that may influence feeding problems. Several indirect feeding assessments have been developed to assess feeding problems in individuals with and without developmental and intellectual disabilities across the life span (see Seiverling, Williams, & Sturmey, 2010).

For example, the Behavioral Pediatric Feeding Assessment Scale (Crist & Napier-Phillips, 2001) is a 35-item questionnaire designed to assess the mealtime behavior of children. Caregivers rate 25 items on the child's mealtime behaviors and rate 10 items on their feelings or strategies to manage feeding problems using a 5-point Likert scale from never to always. The Brief Autism Mealtime Behavior Inventory (Lukens & Linscheid, 2008) is an 18-item questionnaire designed to assess the mealtime problem behavior among children with ASD. Parents rate each item using a 5-point Likert scale from never to always to measure the frequency of mealtime problem behavior, rote behavior characteristic of ASD (e.g., "My child is flexible around mealtime routines," "My child is willing to try new foods"), and preferences. The Screening Tool of Feeding Problems (Matson & Kuhn, 2001) assesses feeding problems in individuals, regardless of age, with developmental disabilities. It consists of 23 questions arranged into 5 categories: aspiration risk, food selectivity, feeding skills, food refusal behavior problems, and nutrition-related behavior problems. Respondents rate each question according to frequency and severity of the feeding problem. Finally, the Parent Mealtime Action Scale (Hendy, Williams, Camise, Eckman, & Hedemann, 2009) is a 31-item questionnaire designed to assess caregiver strategies to manage mealtime problem behavior (e.g., "You insisted the child 'try one bite,'" "You offered the child a toy or a favorite activity as a reward for eating"). Caregivers rate the frequency of their behavior on a 3-point Likert scale from never to always.

Descriptive Assessments. Although indirect assessments are relatively simple to conduct, they do not offer specific information regarding the nature, extent, or frequency of IMB. Conversely, descriptive feeding assessments involve the direct observation of typical mealtime routines and as such allow the observer to obtain more specific information on the topography, extent, frequency, and variables that may influence IMB in the natural environment. For example, Borrero, Woods, Borrero, Masler, and Lesser (2010) conducted descriptive assessments with 25 child-parent dyads during typical mealtimes and found that parents delivered attention, access to tangible items, and escape following their child's IMB. The researchers then compared the conditional probabilities of these common parent-delivered consequences given IMB and acceptance to the unconditional probabilities of each event and found that parents delivered escape and attention most frequently for food refusal. The authors did not conduct subsequent functional analyses to verify the results of their descriptive assessments; therefore, the extent to which descriptive assessments may provide information regarding the variable(s) influencing IMB could not be delineated.

To examine the extent to which the results of descriptive assessments of IMB predict those of functional analyses of IMB, Borrero, England, Sarcia, and Woods (2016) compared descriptive assessment and functional analysis of IMB outcomes for 10 child-parent dyads. The researchers compared the conditional probabilities of the descriptive assessment data to the functional analysis outcomes and found that these assessment outcomes corresponded on 71% of cases. Because this is the first study to examine correspondence between descriptive assessments and functional analyses of IMB, more research is needed to replicate these effects and perhaps to refine the descriptive assessment procedures to produce higher correspondence rates. At the same time, these preliminary data seem promising and may suggest that descriptive assessments may be a viable alternative when a functional analysis of IMB is not feasible (Borrero et al., 2016).

Functional Analysis. Piazza, Fisher, et al. (2003) were the first researchers to demonstrate that the consequences commonly arranged in a functional analysis of IMB were observed in the natural environment. The researchers conducted a descriptive assessment of 15 child-parent dyads during mealtime and found that parents provided attention (coaxing, reprimands), escape, and access to preferred toys and food following IMB. The researchers then arranged these consequences using functional analysis procedures similar to those described by Iwata, Dorsey, Slifer, Bauman, and Richman (1994) by alternating control, attention, escape, and tangible conditions. To ensure the relevant establishing operation or discriminative stimulus was in place during each condition, the researchers instructed participants to take a bite of food every 30 s in all conditions. Piazza, Fisher, et al. found that escape was the most commonly identified function, attention was the second most commonly identified function, and access to tangible items was the least commonly identified function of IMB for their participants.

Najdowski et al. (2008) extended upon the study by Piazza et al. (2003) by using caregivers as therapists. Clear functions were obtained, and caregivers also conducted the functional analyses with a high degree of procedural fidelity. However, Najdowski et al. used alternative functional analysis procedures and only delivered prompts to take a bite of food during the escape condition. That is, during all other test conditions, the researchers placed a plate of non-preferred foods in front of the participant but did not instruct the participants to take a bite. These researchers found that all six participant's IMB was maintained by escape but noted that their procedural variation may have produced false-positive outcomes because the establishing operation or discriminative stimulus may not have been present during all other test conditions.

Bachmeyer et al. (2009) demonstrated that the identification and function-matched treatments of all possible functions of problem behavior are critical to successful treatment outcomes. The researchers first used the functional analysis procedure similar to that described by Piazza, Fisher, et al. (2003) to identify four children whose IMB was multiply controlled by escape and attention. The researchers then evaluated and compared the effectiveness of both function-based extinction procedures, individually and in combination. Results showed that (a) attention extinction alone did not increase acceptance or decrease IMB, (b) escape extinction alone produced an increase in acceptance and a decrease in IMB, and (c) attention and escape extinction combined produced the best treatment outcomes for all participants (i.e., 100% acceptance and near-zero levels of IMB). These results further emphasize the importance of assessment techniques that can accurately identify all possible functions of IMB.

While research on functional assessment has largely focused on IMB and certain factors that may influence its presence, functional assessment may be much broader than this. Researchers and clinicians alike are encouraged to consider functional analysis as a general framework for evaluating the extent to which different factors contribute to the presence of diverse types of feeding problems, skills deficits, and more. In the following section, we provide a brief overview of behavioral interventions for feeding problems. Our aim in doing so is to provide the reader with a broad look into the behavioral approach to intervention for feeding problems.[1] Also, while we categorize interventions to organize our discussion, we acknowledge that feeding interventions probably most often involve treatment packages comprised of several components. We begin by reviewing antecedent interventions.

Intervention

Antecedent Interventions. Antecedent interventions may be considered any intervention that aims to influence a target behavior prior to the behavior occurring. Process wise, these interventions are effective when they alter the aversive characteristics of the feeding context in some way, increase the value of reinforcement for food consumption, and/or reduce the response effort for engaging in the target behavior. For example, several researchers have studied an antecedent procedure known as simultaneous presentation (e.g., Ahearn, 2003; Piazza et al., 2002). This strategy involves providing non-preferred foods at the same time as preferred foods (e.g., by putting preferred condiments on the non-preferred foods). When this procedure works, it alters the aversive characteristics of the non-preferred food so as to increase consumption. A number of researchers have also studied a procedure known as the high-probability instructional sequence. In this procedure 2–3 responses which the individual has a high probability of complying with are presented just before the non-preferred food (e.g., Dawson et al., 2003; Meier, Frylíng, & Wallace, 2012; Patel et al., 2007). Others have manipulated the texture of the target food as a means to improve food consumption and have found that this may also be an important variable to consider (e.g., Patel, Piazza, Layer, Coleman,

---

[1] More exhaustive reviews of feeding intervention are available (e.g., Bachmeyer, 2009; Fernand, Saksena, Penrod, & Fryling, in press; Sharp, Jaquess, Morton, & Herzinger, 2010; Silbaugh et al., 2016).

& Swartzwelder, 2005). Additional research has examined different ways of presenting bites of food, such as using a flipped spoon or Nuk brush (Sharp, Harker, & Jaquess, 2010). Demand fading has been examined as well, where the amount or number of bites of food is initially decreased and later increased systematically contingent upon food consumption (e.g., Najdowski et al., 2010). While a large number of antecedent interventions have been studied, less information is known about exactly when such interventions are more or less likely to be effective, both in isolation and in combination with other interventions (Seubert, Fryling, Wallace, Jimenez, & Meier, 2014). It is possible that antecedent interventions are more likely to be effective in isolation when they are used with individuals who engage in little or no IMB. In addition, in circumstances where antecedent interventions are not effective in isolation, they may enhance the effects of other interventions (e.g., further reducing IMB; Patel et al., 2006). Additional research is needed to explore these possibilities.

Reinforcement Procedures. Behavior analysts have also explored the use of reinforcement procedures in the treatment of feeding disorders. For example, differential reinforcement of alternative behavior (DRA) procedures have been evaluated in the treatment of feeding problems. In a typical DRA procedure, reinforcers such as toys, attention, and/or preferred foods are provided contingent upon a particular target behavior (in this case food consumption), and not at any other time. Feeding researchers have evaluated DRA for some time, with early studies on the topic being published nearly 40 years ago (e.g., Riordan, Iwata, Wohl, & Finney, 1980) and continuing to more recent times (e.g., Brown, Spencer, & Swift, 2002). Variations of DRA have also been evaluated, including differential negative reinforcement (i.e., providing a break from the meal contingent upon engaging in the target behavior; Kahng, Tarbox, & Wilke, 2001). DRA has also been examined in conjunction with other procedures (e.g., Najdowski et al., 2010), and the extent to which DRA contributes to treatment outcomes relative to extinction procedures has also been considered (e.g., Piazza, Patel, Gulotta, Sevin, & Layer, 2003). Noncontingent reinforcement (NCR) has also been examined in the treatment of feeding problems. Wilder, Normand, and Atwell (2005) examined the effects of an NCR procedure, which consisted of providing continuous access to a preferred video during mealtimes and found that this reduced self-injurious behavior and increased acceptance of bites. Like DRA, NCR has also been evaluated in combination with other interventions (e.g., Reed et al., 2004). Others have compared variations of reinforcement procedures (e.g., Allison et al., 2012; Patel, Piazza, Martinez, Volkert, & Santana, 2002). Similar to antecedent interventions, more research is needed to identify when reinforcement-based interventions are more or less likely to be useful in isolation and in combination with other interventions.

Extinction Procedures. There is considerable support for the use of extinction procedures, particularly escape extinction (EE), in the treatment of feeding disorders (see Piazza, 2008; Volkert & Piazza, 2012). Two of the most utilized extinction procedures include non-removal of the spoon (NRS) and physical guidance (PG) (Piazza, 2008). In NRS, the feeder holds the feeding utensil at the child's lips until the child accepts or consumes the presented food while IMB is ignored. During PG, the feeder gently presses the jaw (or mandibular joint) of the child to prompt the mouth to open and then deposits the food.[2] Importantly, however, extinction procedures may also be associated with extinction bursts, whereby challenging behavior may initially increase or worsen when they are initially implemented. These initial struggles may increase the probability of integrity failures, which could ultimately worsen an individual's feeding problem (see Bachmeyer, 2009). Given the nature of feeding problems, this could be dangerous, and therefore it is very important that extinction procedures be used with caution and only with professionals with experience using the procedure and in settings which are equipped and prepared for possible challenges. Related to

---

[2] See Fernand et al. (in press) for a more detailed overview of extinction procedures, variations, etc., in the treatment of feeding problems.

this, parents may find other interventions more acceptable than extinction procedures (e.g., Vazquez, Fryling, & Hernandez, in press), again suggesting that such procedures should only be used in certain situations and with therapists with very specialized training.

While a wide range of behavioral procedures have been evaluated in the feeding literature, we are yet to develop a systematic approach to matching particular interventions to particular individuals. For this reason, additional attention should be given to pre-treatment assessment by both clinicians and researchers alike. More specific participant information will help guide treatment selection, as there is a need for the field to further understand *when* different interventions are more or less likely to be successful with different populations. The majority of behavior analytic studies on the topic of feeding disorders have focused on food selectivity and food refusal, with relatively less attention being given to several other important behaviors. Given this the remainder of the chapter will focus on two important target behaviors that are common among individuals with disabilities and feeding problems, packing and rapid eating.

Packing

Packing involves storing foods or liquids in one's mouth (e.g., pocketing food on the side of one's mouth without swallowing). Packing may emerge for various reasons, including physical abnormalities, oral-motor skills deficits, and positive and negative contingencies of reinforcement. Packing is important for a variety of reasons; it may lead to choking, aspiration, decreased caloric intake, extended meal times, and parental stress. If left untreated, packing can result in dental problems, dehydration, malnutrition, and failure to thrive (see Silbaugh, Swinnea, & Penrod, 2018 for an overview). Despite the numerous health and safety risks associated with packing, researchers have not widely addressed this issue in the literature—a situation that may have arisen because researchers often consider packing a *collateral response* (e.g., Levin, Volkert, & Piazza, 2014; Sevin, Gulotta, Sierp, Rosica, & Miller, 2002; Vaz, Piazza, Stewart, Volkert, & Groff, 2012). In fact, several researchers reported that packing emerged only after they observed improvements in the target dependent variable (e.g., acceptance [Levin et al., 2014; Riordan et al., 1980; Sevin et al., 2002; Vaz et al., 2012] and expulsion [Sevin et al., 2002]). For example, Sevin et al. (2002) used NRS to increase acceptance in one child who presented with food refusal. As acceptance increased, the researchers observed a concomitant increase in expulsion, which they treated with representation of expelled bites. As expulsion decreased, the researchers observed a concomitant increase in packing, which they subsequently treated with redistribution. The authors noted that these findings indicate that packing may belong to the same response class as the other food refusal behaviors (Sevin et al., 2002). The following section provides an overview of some of the behavior analytic intervention research on packing.

Intervention

Researchers have evaluated the efficacy of several treatments of packing in isolation or in combination: differential reinforcement of acceptance (Buckley & Newchok, 2005; Riordan et al., 1980), response cost (Buckley & Newchok), simultaneous presentation (Buckley & Newchok), food redistribution (Gulotta, Piazza, Patel, & Layer, 2005; Levin et al., 2014; Sevin et al., 2002; Volkert, Vaz, Piazza, Frese, & Barnett, 2011), swallow facilitation (Volkert et al., 2011), a chaser (Vaz et al., 2012), and food texture manipulation (Patel et al., 2005). We first review seminal behavioral studies on each of these treatments for packing, followed by suggestions for future research and clinical practice.

Differential Reinforcement of Alternative Behavior. In one of the first reports of packing in the behavioral literature, Riordan et al. (1980) evaluated the effects of DRA on the acceptance of non-preferred foods in two children (ages 6 and 9) with developmental disabilities. Initially, the researchers arranged the contingency on acceptance of non-preferred food or drink (i.e., researchers provided a bite of preferred food contingent on the child's acceptance of a non-preferred food or drink). Researchers found that DRA not only produced an increase in acceptance, but they also observed in a collateral increase in packing. Therefore, they added swallowing to the existing contingency such that they required children to consume the non-preferred food or drink prior to delivering reinforcement. After the researchers revised the contingency, they observed an increase in consumption of all target foods for both children. It should be noted that the researchers did not collect data on packing; therefore, it is unclear if the revised DRA contingency was responsible for the observed decrease in packing. Regardless, these findings (a) provide a demonstration of the emergence of packing following successful treatment of acceptance of food and (b) seem to suggest that simply arranging a contingency on the consumption of food—at least for some individuals—can decrease packing and may actually circumvent the emergence of packing altogether.

Differential Reinforcement with Response Cost and Simultaneous Presentation. In another study involving differential reinforcement, Buckley and Newchok (2005) compared the effectiveness of differential reinforcement with response cost, differential reinforcement with response cost and simultaneous presentation, and simultaneous presentation alone on the packing of a 9-year-old girl with autism spectrum disorder. During differential reinforcement with response cost, researchers presented the child's preferred movie at the start of the session, terminated the movie clip contingent on packing, and provided praise for a clean mouth (consumption). When researchers evaluated simultaneous presentation, either within the context of differential reinforcement plus response cost or in isolation, they presented a spoon that contained a bite of cookie placed behind the target food. Researchers found that all three treatments were effective, but that simultaneous presentation alone was the most effective. However, because simultaneous presentation was not implemented alone prior to being combined with differential reinforcement and response cost, it is unclear if simultaneous presentation alone would have been effective if it had been implemented prior to it being combined with differential reinforcement and response cost.

Texture Manipulation. Patel et al. (2005) assessed the influence of food texture on the packing and caloric consumption in three children between the ages of 3 and 4 years diagnosed with a feeding disorder. The researchers developed individualized treatments for each child based on previous assessment outcomes and used textures recommended by an occupational therapist. All treatments involved several of the same components: (a) fixed time bite presentations (15 s or 30 s), (b) NRS, (c) representation of expelled bites, (d) prompts to swallow packed bites (for one participant, the researchers also used redistribution with bristle massaging toothbrush), and (e) brief praise or brief access to preferred toys contingent on a swallow response. The researchers observed lower levels of packing and greater calorie consumption with lower textures of food relative to higher textures of food. Because the researchers included NRS, it is unclear if manipulating texture alone would have produced the same decrease in packing. However, because NRS was present during baseline and treatment phases, it is unlikely that NRS was responsible for the differentiated levels of acceptance across textures for all participants. In addition, although Patel et al. (2005) did not identify the mechanisms responsible for the effectiveness of the lower food textures, they suggested several potential rationales for observed decreases in packing with lower food textures: (a) lower food textures may simply be easier to swallow, (b) higher food textures require more advanced oral-motor skills, and (c) higher food textures may be a conditioned aversive stimulus such that individuals avoid swallowing foods at higher textures as opposed to lower textures. Because these possibilities can have different implications for treatment (e.g., treatment may consist of teaching prerequisite oral-motor skills or reconditioning higher-textured food), researchers should identify the exact mechanism(s) responsible for the effectiveness of lower textures on packing.

Chaser. Vaz et al. (2012) used a chaser (a food or liquid each child consistently accepted and swallowed) to decrease the packing of three children between the ages of 24 months to 10 years. As in Patel et al. (2005), the researchers arranged NRS for food refusal, representation for expulsion, and brief praise for acceptance and consumption in all phases. Due to individual participant characteristics, the chaser treatment was procedurally different for each child. For one child, researchers presented the chaser (4 ml of liquid) immediately following acceptance of the target food by holding a nosey cup at the child's lips, instructing the child to "take a drink," and depositing the liquid in the child's mouth. For another child, researchers waited to present the chaser after initial acceptance and conducted a clean-mouth check 15 s after the child accepted the target food. If the child packed the food, they presented the chaser (a bite of fruit) on a spoon on a plate in front of the child. If the child did not feed himself the chaser within 5 s, the researchers used a Nuk brush to roll the chaser on his tongue. For the final participant, the researchers presented the liquid chaser in a cup with a straw and instructed the child to "take a drink" immediately after he accepted the target food. The authors noted that although the children in their study consistently accepted at least one liquid or food and accepted the chaser during the evaluation, this may not be the case for all individuals who pack food. As such, a chaser may not be a viable treatment alternative for those individuals who do not consistently accept the chaser inside or outside of the treatment protocol. The authors also noted that a chaser may not be a viable treatment alternative for those children whose medical evaluations indicate that they are at risk for aspiration—or if their risk of aspiration is unknown. Finally, although the authors provided the chaser immediately after acceptance for two of three participants, they stated that it may be important to provide participants an opportunity to chew the target food before presenting the chaser to avoid situations in which participants would have mixed textures in their mouths (e.g., solid food plus liquid chaser), which may increase the risk of aspiration for some individuals. The exact reason for the effectiveness of the chaser remains unclear; though, several researchers have suggested that it may facilitate the swallow response by (a) addressing oral-motor deficits (e.g., Levin et al., 2014; Vaz et al., 2012) or (b) decreasing response effort associated with swallowing. In addition, the chaser may function as escape extinction (Vaz et al., 2012).

Food Redistribution. Gulotta et al. (2005) used food redistribution to decrease the packing and latency to clean mouth in four children with feeding disorders. Throughout all phases, the researchers arranged NRS for food refusal, representation for expulsion, and reinforcement (a token, brief access to a toy, or brief praise) for acceptance and swallowing. During redistribution, researchers used a brush to scoop any remaining food in the mouth and turn the brush to place the food in the middle of the child's tongue. Researchers either implemented the redistribution procedure immediately (1 child), after 15 s (2 children) or after 30 s (1 child) of the children accepting the target food in the mouth. Although the researchers only observed clinically significant decreases in packing for two of four children, they observed a mean decrease in latency to clean mouth for the two children for whom redistribution only produced a small decrease in packing (these children packed food on 50% or more of trials). This posttreatment decrease in mean latency to clean mouth may be considered a clinically significant treatment outcome because these children were holding food in their mouths for less time after treatment than they were prior to treatment. In addition, all children consumed more grams of food following treatment. Given these findings, the authors noted that it may be important to collect data on multiple measures to accurately capture the effects redistribution has on packing.

Packages Involving Redistribution

Redistribution + Swallow Facilitation. Volkert et al. (2011) used redistribution and swallow facilitation to reduce the packing behavior in two children with feeding disorders. During all phases, researchers arranged NRS for food refusal, representation for expulsion, and brief praise for acceptance and mouth clean (consumption). During treatment, researchers implemented redistribution with a flipped spoon and swallow facilitation in which the researchers collected packed food on the spoon, rotated the spoon 180°, placed the flipped spoon behind the middle of the child's tongue, and applied slight pressure while pulling the spoon toward the lips. The swallow facilitation component involved the placement of the food toward the posterior of the tongue, as opposed to the middle of the tongue, along with the application of slight pressure to the tongue while pulling the food toward the lips. These components appear to be the critical components involved in swallow facilitation because the combination of placing the bolus toward the posterior of the tongue plus the application of slight pressure may act as an eliciting stimulus that elicit the swallow response.

Redistribution + Swallow Facilitation + Chaser. Levin et al. (2014) found that redistribution, swallow facilitation, and a chaser decreased the packing behavior in two children with autism spectrum disorder. During all phases, researchers arranged NRS for food refusal, representation for expulsion, and brief praise for acceptance and clean mouth (consumption). During treatment, the researchers presented the liquid chaser immediately after the child accepted the target food. If the child packed the bite, researchers collected and removed the packed food with a Nuk brush (1 child) or a spoon (1 child), re-inserted the food on the Nuk brush or spoon back into the child's mouth, placed the Nuk brush or spoon onto the posterior (1 child) or middle (1 child) of the child's tongue, rotated the Nuk brush or spoon 180°, and applied slight pressure while dragging the brush or spoon toward the child's lips. For one child, the swallow facilitation component involved both the posterior placement of the food on the tongue and the application of slight pressure while dragging the spoon toward the lips. For the other child, the swallow facilitation component only involved the application of slight pressure while dragging the spoon toward the lips (i.e., food was placed mid-tongue). Because this study involved numerous procedural variations within and across participants, treatment components, and combinations of treatments, the necessary and sufficient treatment components are unknown.

Given the limited number of studies on redistribution, researchers have ample opportunities to contribute to this literature. For example, it may be the case that redistribution is more or less effective when implemented at different latencies from initial acceptance. Gulotta et al. (2005) implemented redistribution either immediately, after 15 s, or after 30 s of initial food acceptance with their participants; however, they did not compare this procedural variation on the level of packing behavior; therefore, researchers should investigate the effects of the latency from initial acceptance with which redistribution is implemented on packing. Researchers may also consider evaluating the effects of other procedural differences within the redistribution procedure (e.g., type of instrument used to redistribute food [e.g., upright spoon, flipped spoon, Nuk brush], position of food on the tongue, application of pressure on the tongue). Researchers can compare the effectiveness of redistribution when implemented alone or in combination with other treatment components (e.g., swallow facilitation, texture manipulation, or the addition of a chaser).

Recommendations for Clinical Practice

Because packing behavior can be influenced by physical abnormalities, oral-motor skill deficits, operant processes, or some combination of these causes, it is imperative that an interdisciplinary assessment be conducted to obtain a comprehensive understanding of an individual's disordered feeding. This is a particularly important consideration in the treatment of packing given that unmatched treatments can increase safety risks such as choking or aspiration. For example, if an individual's tongue lateralization is deficient such that he or she cannot form or bolus, using regular-textured foods within a differential reinforcement of consumption contingency will likely prove ineffective and may present safety risks.

Several researchers observed that packing increased following the treatment of acceptance (e.g., Riordan et al., 1980; Sevin et al., 2002; Vaz et al., 2012) and expulsion (e.g., Sevin et al.).

Clinicians should be sensitive to this possibility and conduct clean-mouth checks during feeding intervention to assess concomitant increases in packing. Clean-mouth checks also allow the clinician to identify the amount of packed food in client's mouth such that he or she can determine if it is safe to provide another bite of food. Conversely, the interdisciplinary team may determine that any amount of food in the mouth poses a safety risk to the individual such that he or she must swallow all food in the mouth before the clinician presents additional food. Finally, after the physician, speech therapist, or occupational therapist identifies the client as a safe oral feeder (this should be established before a behavioral intervention is implemented), clinicians should consider arranging the reinforcement contingency on *consumption* of food rather than on the *acceptance* of food. This may circumvent the emergence of expulsion, packing, or both following the treatment of acceptance, which may subsequently lead to more rapid treatment of food refusal.

Rapid Eating

Rapid eatingis a relatively understudied behavior that also falls under the broad umbrella of feeding problems. Rapid eating is distinct from other areas of the feeding literature in that it does not easily fit into either the skills deficit (e.g., chewing skills) or behavioral excess (e.g., throwing food, yelling) categories in the traditional sense. Rather, it is the amount of time between the behavior, in this case taking bites of food, that presents concerns. The problem of rapid eating is not only concerning for social reasons (e.g., it prevents one from contacting the reinforcers associated with sitting down and eating with others over a period of time), but it also presents threats to one's safety and health, in both the short- and long-term. Immediate threats include the possibility of choking and/or throwing up food, whereas more long-term threats may include obesity and all of the challenges that are associated with that. Interestingly, rapid eating is a concern across populations of individuals, with several researchers noting concerns with rapid eating in typically developed adults (e.g., Azrin, Brooks, Kellen, Ehle, & Vinas, 2008; Britt & Singh, 1985). At the same time, individuals with developmental disabilities may be especially at risk for rapid eating, and for this reason, rapid eating warrants specific attention from clinicians working with this population. While rapid eating is clearly a socially important area for research and practice, it has been given relatively little attention by behavioral researchers, especially when compared to other feeding problems. Given this, the final section of this chapter will focus on the behavior analytic research on rapid eating.

One of the earliest behavior analytic studies on rapid eating was conducted by Favell, McGimsey, and Jones (1980). This study involved four individuals with developmental disabilities (three males and one female), ranging from 9 to 21.5 years of age. Each of the individuals had difficulties related to their rapid eating which prevented them from accessing less restrictive environments. Specifically, the participants ate an average of 10.5 bites of food every 30 s. Before the experiment, the researchers were curious as to whether or not access to additional food would slow down the participants eating. In other words, the researchers wondered if the individuals ate rapidly simply because they were hungry. However, permitting the individuals to eat additional servings of food did not impact rapid eating in any meaningful way; the individuals simply ate more food. Favell et al. then examined a behavioral treatment package comprised of reinforcing pauses between bites of food, shaping longer pauses, and preventing attempts to eat rapidly by prompting the participants to wait. The reinforcement schedule was thinned contingent upon reductions in rapid eating.

The results of this treatment package demonstrated that the number of bites per 30 s decreased for each of the four participants. Specifically, during baseline participants took an average of 10–12 bites per 30 s, whereas during intervention participants took an average of 3–4 bites per 30 s. Moreover, the staff working in the participant's residential units were easily taught how to implement the procedure and were able to continue using the procedure while they were carrying out their other job duties. As a result of this, the results of 3–4 bites per 30 s maintained for 3 months, leading to the participants being moved to less restrictive settings. There were also number of indirect effects of the intervention; one participant who was initially rather messy during mealtimes was no longer messy, and another participant, who vomited during 15% of the baseline meals, no longer vomited during intervention. Finally, although not measured formally, the researchers reported that none of the participants engaged in challenging behavior during the intervention, suggesting that the intervention was not unpleasant for the participants. This study represents an important initial effort for behavior analysts to help individuals who struggle with rapid eating.

Lennox, Miltenberger, and Donnelly (1987) evaluated three treatments to reduce rapid eating. Three adults with developmental disabilities participated in the evaluation: a 32-year-old female, a 44-year-old male, and a 28-year-old male. Researchers selected a target inter-response time (IRT) of 15 s by observing two individuals who were considered to be socially appropriate eaters. Three intervention conditions were evaluated: (1) fixed-interval response blocking, (2) differential reinforcement of low rates of behavior (DRL), and (3) DRL + prompting. In the fixed-interval response blocking condition, eating responses that occurred 15 s after the previous eating response were reinforced with access to food, whereas eating responses before the 15 s interval were blocked (i.e., the first response that occurred after the 15 s IRT was reinforced). During the DRL condition, attempts at eating before the 15 s IRT had passed continued to be blocked, and attempts to eat prior to the passing of 15 s also resulted in the resetting of the 15 s interval (i.e., participants had to wait an additional 15 s; they were not permitted to take a bite until 15 s without any attempts at eating had passed). During the DRL + prompting condition, a competing response was prompted immediately after participants took a bite of food. Specifically, participants were prompted to put their utensils on their plates and their hands on their laps, with less and less prompting being required as participants were more independent. For one of the participants, an additional time-out from food was added to the DRL + prompting condition due to challenging behavior. This meant that attempts to eat before the 15 s interval had passed resulted in the tray of food being removed for 15 s, followed by the participant waiting for an additional 15 s when the food tray was represented.

Results of the evaluation showed that for two of the participants, mean IRT's did not reach the 15 s target level until the DRL + prompting condition was implemented. For a third participant, the 15 s target IRT was not met in any of the conditions, and despite the addition of a time-out procedure to the DRL + prompting condition, this participant engaged in challenging behavior throughout the evaluation. At the same time, it should be noted that while the 15 s goal was not met with this third participant, a mean IRT of 11.5 s was achieved, representing a substantial increase over baseline levels. This study underscored the importance of considering challenging behavior when evaluating intervention for rapid eating.

Wright and Vollmer (2002) conducted a systematic replication of the study by Lennox et al. (1987) with a 17-year-old female with profound developmental disability and cerebral palsy. Remember, the DRL procedure evaluated by Lennox et al. was always set at 15 s, regardless of the participant's performance, and the 15 s interval was reset upon each attempt to take a bite prior to the 15 s interval. This meant that it was possible that participants would never contact the reinforcer and that the interval could be continuously reset. In an effort to be more sensitive to this possibility, Wright and Vollmer evaluated an adjusting DRL procedure, whereby the length of the DRL interval depended upon the participant's average IRT across the previous five sessions. After rapid eating remained unchanged during baseline conditions and an initial fixed DRL condition, rapid eating decreased during the adjusting DRL condition, suggesting that it is important to continue to adjust the DRL interval based upon participant's behavior. Moreover, while challenging behavior (specifically self-injurious behavior) occurred during the initial intervention conditions, challenging behavior decreased over time, suggesting that increases in challenging behavior during intervention for rapid eating may be temporary. From a practical standpoint, it is important for clinicians to recognize that challenging behaviors may be temporary. From a research standpoint, it is important that future research further consider strategies that might reduce the occurrence or severity of challenging behavior during behavioral treatment as this may compromise safety and caregiver fidelity.

Technology-Enhanced Interventions

While the aforementioned studies represent important steps toward understanding effective interventions for rapid eating, they all require the presence of a therapist during mealtimes, especially during the early stages of treatment. It is possible that prompts, typically provided by therapists, could also be provided through the use of technology. To evaluate this possibility, Anglesea, Hoch, and Taylor (2008) evaluated the use of a pager prompt in treatment of rapid eating with three teenagers with autism. Each of the participants had a history of rapid eating, and two of them also had a history of gastroesophageal reflux and rumination. Two types of pagers were used in the intervention, depending on their availability in the participant's classroom (i.e., they either used one or the other throughout the study). One of the devices could be programmed to vibrate at specific intervals and stopped automatically after 3 s, and the other vibrated at programmed intervals and continued until someone pressed any button on the device (each pager vibrated at set intervals regardless of this difference in how the vibration stopped). To set specific intervals for each individual and target food, the authors first timed a typically developed adult eating one of the target foods. As such, the length of vibration intervals varied across participants and their respective target food.

The effects of the pager on rapid eating were evaluated in three reversal designs, one for each participant. During baseline sessions participants were presented with the target food while wearing a pager that was not activated. Sessions were conducted during lunchtime, and participants were first presented with the target food, and after they consumed it, the rest of their lunch was presented. During training, pagers were set to vibrate at varying interval lengths, in order to avoid developing time-based responding and to increase the chances of the vibration itself developing stimulus control. The trainer (a teacher) first prompted the participants to touch the pager and blocked any attempts to eat before the pager vibrated. After this, the participants were no longer required to touch the pager, while attempts to eat before the pager vibrated continued to be blocked. Participants were only permitted to take a bite of their food when the pager vibrated and were prompted to do so, if needed. After eating a bite of food, participants were prompted to put their hand back on the pager and were praised for waiting until the pager vibrated. Training sessions consisted of 10 trials, and training continued until participants only took bites after the pager vibrated for 100% of the trials for two consecutive sessions. After training the participants entered a pager-activated phase, which was the same as baseline with the exception that the pager was turned on. Participants then completed an additional baseline phase and a final intervention phase to complete the ABAB reversal design.

The results demonstrated that the total number of seconds eating the target food item increased only during the pager-activated condition and not during either of the baseline conditions, suggesting that the pager was likely to be responsible for the change in behavior. Importantly, the authors of the study also measured the total number of bites taken during each session and found that the total number of bites did not vary across baseline and intervention conditions. This confirmed that it was indeed the time between bites that increased during the intervention condition (i.e., and not the amount of food consumed during each bite). Finally, researchers conducted an additional probe condition with one of the participants, with no adult present, and found that the length of time between bites continued at target levels.

Echeverria and Miltenberger (2013) further evaluated the use of pager prompts, alone and in combination with other interventions, to reduce the rapid eating of two adults with developmental disabilities. Two adults (51 and 32 years old), living in group homes, participated in the study; both participants had a history of rapid eating and choking prior to participating in the study. Sessions were conducted during lunch or dinner times, at the participant's regular mealtime setting. Staff members sat next to the participant in order to provide prompts as needed. Using a multiple-baseline and reversal designs, the effects of a pager, alone and in combination with verbal prompts, were evaluated.

After baseline conditions participants went through a training session aimed at establishing the pager vibrating as a discriminative stimulus for taking a bite of food. Training involved the trainer providing instructions to the participant to only take a bite after the pager vibrated and then setting the pager on the table. To start, the trainer modeled this by placing their hand on the pager, and, when the pager vibrated, the trainer pretended to take a bite of food (with no food or other materials present). Following this model trial, the participant was instructed to try. Praise was provided by the trainer when the participant also engaged in the behavior after the pager had gone off. Upon engaging in three correct responses, the participant was instructed to put the pager on his belt and was provided with an empty plate and silverware. When the participant again engaged in three correct responses, the participant was provided with a plate with food and was instructed to wait before taking a bite. Once the participants engaged in three consecutive correct responses with food present, training was considered to be complete. Praise was provided throughout training for correct responses, and if the participants attempted to take a bite prior to the pager vibrating, these attempts were blocked while the trainer instructed them to "wait."

After training participants were exposed to a pager plus verbal prompts treatment condition. This involved the participants wearing the pagers that were set to vibrate at 15 s intervals (the interval being based upon the previously described Lennox et al. study). Before sessions, the trainer reminded the participants not to take a bite until the pager went off, and additional verbal prompts to "wait" were provided whenever participants attempted to take a bite too early. In addition, praise was provided when the participants waited the length of the interval. This condition was followed by a condition with the pager alone, which was similar to the pager plus verbal prompts condition with the exception that there were no prompts to "wait" contingent upon attempts to take bites before the pager went off. Finally, a verbal prompts alone condition was implemented with one of the participants, which involved providing reminders to eat slowly, chew, etc., and praise each time the participant chewed his food before taking another bite (no pager was used in this condition). Follow-up data were also collected; participants were wearing pagers during the follow-up probe, and one of the participants was also receiving verbal prompts. Two measures of social validity were obtained. One group of staff who did not work with the participants was shown videos of the participants at the end of treatment, and they were asked to evaluate the participants' behavior. Additional social validity assessment involved asking three individuals who had experience working with the participants to evaluate their progress.

Results of the evaluation demonstrated that both the pager plus verbal prompts and pager alone conditions decreased the number of bites per minute for both participants. Of note, while the number of bites per minute decreased across both conditions, there was a slightly larger decrease in the pager + prompts condition (i.e., the pager alone was just slightly less effective). Given that the difference between these conditions was slight, and given the practical benefits associated with the pager alone condition (i.e., that a staff member does not have to consistently prompt), it is possible that the pager alone condition would remain preferred. Finally, the results of the social validity assessment, from both unfamiliar and familiar caregivers, were positive, indicating that the behavior change had at least some degree of social significance. Importantly, both of the social validity evaluations suggested that the behavior change achieved was appropriate for community settings.

More recently, Page, Griffith, and Penrod (2017) evaluated the use of a treatment package including a pager prompt, rule, and verbal prompts to decrease rapid eating with a 12-year-old female diagnosed with autism spectrum disorder. Using a multiple-baseline and reversal design, sessions were conducted in both a university-based feeding clinic and home-based setting. Each setting was similar in that sessions were conducted at a dinner table with typical items present. The participant's family and researchers sat at the table during all sessions and engaged in conversation unrelated to the participant's food consumption throughout all sessions. During baseline conditions the participant wore an inactive pager, and the researcher presented the meal by stating "Here's your dinner." There were no consequences related to food consumption. During the active pager with rule condition, the pager was activated (with the interval being set at 20 s), and the participant was given the following rule "Here's your dinner. When you take a bite, chew 20 times, and wait for the buzz to take another bite." (p. 89). There were no consequences related to the participant's compliance with the rule. The active pager with rule and vocal prompt condition was the same as the pager with rule condition with the exception that attempts to take a bite before the pager vibrated resulted in the researcher prompting the participant to "wait." Results demonstrated that the participants' mean IRT between bites did not increase during the active pager plus rule condition but did increase during the pager plus rule plus prompts condition. This effect was replicated in a reversal design in the clinic setting and in a multiple-baseline design in the home setting.

Finally, Valentino, LeBlanc, and Raetz (2018) recently evaluated strategies to fade the use of pagers in the reduction of rapid eating with a 10-year-old child with autism. After collecting baseline data, conducting pager training (similar to that described by Anglesea et al. (2008), and demonstrating the effectiveness of the activated pager, the researchers compared different strategies to fade the pager prompt to transfer stimulus control to the natural environment. Specifically, the researchers evaluated the extent to which the pager could be faded by reducing the intensity of the pager (i.e., by having it vibrate more softly) and the number of times the pager prompt was used (i.e., the frequency of prompts). Specifically, the authors evaluated a low intensity 100% condition (where the pager was used at regularly programmed intervals but at a lower intensity), lower intensity 50% of the intervals (where the pager was used at a lower intensity but only vibrated for half of the intervals), a muffled 100% condition (where the pager was at the lower intensity and also wrapped in bubble wrap and a kitchen dishtowel), and finally a no intensity, 0% condition (i.e., the pager was not activated).

Results indicated that the target IRT maintained during the lower intensity 100% conditions but that the IRT increased significantly during the lower intensity 50% conditions. The IRT maintained at the target level during the muffled condition and also during the final no intensity, 0% condition. The researchers hypothesized that it may be more difficult to pursue fading by reducing the frequency of the pager prompt because the participant learned to take bites after the pager vibrated. In other words, the participant learns to wait for the prompt and will simply wait longer. However, reducing the intensity of the prompt does seem like an option worth exploring further, as this method was associated with maintaining eating at target levels throughout the study, and these prompts were eventually faded entirely.

Recommendations for Research and Practice

Rapid eating is a socially important target behavior and warrants the attention from both researchers and clinicians alike. A number of interventions have been evaluated to reduce rapid eating within the behavioral literature, though the body of research on this topic remains small and is ripe with opportunities for further investigation. Of particular importance are studies that evaluate the long-term maintenance of interventions aiming to reduce rapid eating. While maintenance is always desirable, it may be particularly important with rapid eating given the many hazards associated with the persistence of this behavior. The study by Valentino et al. (2018) is an important step in this regard. More research is needed to better understand how to fade other prompts, such as the verbal prompts employed in some of the studies reviewed here. Similarly, systematic attempts at understanding caregiver preference for different interventions for rapid eating may also be useful, as it seems possible that caregiver's will need to make prolonged efforts to change rapid eating over time. Finally, it is possible that mindful eating interventions (e.g., Hanh & Cheung, 2011), or some components of them, will be useful for some individuals with intellectual disabilities. More research is needed to even begin to explore these possibilities.

Conclusion

Eating is a fundamental activity; it is needed for healthy growth and development, provides a context for social opportunities, impacts family dynamics, and more. We have provided a brief overview of the behavior analytic approach to feeding problems in general and to the areas of packing and rapid eating in particular. There are a number of interesting avenues for continued research and practice, and we hope we have highlighted some of these areas throughout. In particular, we note the need to continue to relate particular participant characteristics and unique contexts to the effects of various interventions. More refined pre-treatment assessment efforts will assist in this effort, and additional research in this area is also called for. Finally, as feeding problems impact families in general (e.g., Craig, Scambler, & Spitz, 2003; Didehbani, Kelly, Austin, & Wiechmann, 2011; Greer, Gulotta, Masler, & Laud, 2008), future research should consider incorporating assessment of parental stress before and after intervention to better understand the extent to which parental stress decreases after behavioral intervention.

References

Addison, L. R., Piazza, C. C., Patel, M. R., Bachmeyer, M. H., Rivas, K. M., Milnes, S. M., & Oddo, J. (2012). A comparison of sensory integrative and behavioral therapies as treatment for pediatric feeding disorders. *Journal of Applied Behavior Analysis, 45*, 455–471.

Ahearn, W. H. (2003). Using simultaneous presentation to increase vegetable consumption in a mildly selective child with autism. *Journal of Applied Behavior Analysis, 36*, 361–365. https://doi.org/10.1901/jaba.2003.36-361

Allison, J., Wilder, D. A., Chong, I., Lugo, A., Pike, J., & Rudy, N. (2012). A comparison of differential reinforcement and noncontingent reinforcement to treat food selectivity in a child with autism. *Journal of Applied Behavior Analysis, 45*, 613–617. https://doi.org/10.1901/jaba.2012.45-613

Anglesea, M. M., Hoch, H., & Taylor, B. A. (2008). Reducing rapid eating in teenagers with autism: Use of a pager prompt. *Journal of Applied Behavior Analysis, 41*, 107–111. https://doi.org/10.1901/jaba.2008.41-107

Azrin, N. H., Brooks, J., Kellen, M. J., Ehle, C., & Vinas, V. (2008). Speed of eating as a determinant of the bulimic desire to vomit. *Child & Family Behavior Therapy, 30*, 263–270.

Bachmeyer, M. H. (2009). Treatment of selective and inadequate food intake in children: A review and practical guide. *Behavior Analysis in Practice, 2*, 43–50.

Bachmeyer, M. H., Piazza, C. C., Fredrick, L. D., Reed, G. K., Rivas, K. D., & Kadey, H. J. (2009). Functional analysis and treatment of multiply controlled inappropriate mealtime behavior. *Journal of Applied Behavior Analysis, 42*, 641–658.

Baer, D. M., Wolf, M. M., & Risley, T. R. (1968). Some current dimensions of applied behavior analysis. *Journal of Applied Behavior Analysis, 1*, 91–97.

Benjasuwantep, B., Chaithirayanon, S., & Eiamudomkan, M. (2013). Feeding problems in healthy young children: Prevalence, factors, and feeding practices. *Pediatric Reports, 5*, 38–42. https://doi.org/10.4081/pr.2013.e10

Borrero, C. S. W., England, J. D., Sarcia, B., & Woods, J. N. (2016). A comparison of descriptive and functional analyses of inappropriate mealtime behavior. *Behavior Analysis in Practice, 9*, 364–379. https://doi.org/10.1007/s40617-016-0149-5

Borrero, C. S. W., Woods, J. N., Borrero, J. C., Masler, E. A., & Lesser, A. D. (2010). Descriptive analyses of pediatric food refusal and acceptance. *Journal of Applied Behavior Analysis, 43*, 71–88.

Britt, E., & Singh, N. N. (1985). Reduction of rapid eating by normal adults. *Behavior Modification, 9*, 116–125.

Brown, J. F., Spencer, K., & Swift, S. (2002). A parent training programme for chronic food refusal: A case study. *British Journal of Learning Disabilities, 30*, 118–121.

Buckley, S. D., & Newchok, D. K. (2005). An evaluation of simultaneous presentation and differential reinforcement with response cost to reduce packing. *Journal of Applied Behavior Analysis, 38*, 405–409. https://doi.org/10.1901/jaba.2005.71-04

Craig, G. M., Scambler, G., & Spitz, L. (2003). Why parents of children with developmental disabilities requiring gastronomy feeding need more support. *Developmental Medicine and Child Neurology, 45*, 183–188.

Crist, W., & Napier-Phillips, A. (2001). Mealtime behaviors of young children: A comparison of normative and clinical data. *Journal of Developmental and Behavioral Pediatrics, 22*, 279–286.

Dawson, J. E., Piazza, C. C., Sevin, B. M., Gulotta, C. S., Lerman, D., & Kelley, M. L. (2003). Use of the high-probability instructional sequence and escape extinction in a child with food refusal. *Journal of Applied Behavior Analysis, 36*, 105–108. https://doi.org/10.1901/jaba.2003.36-105

Didehbani, N., Kelly, K., Austin, L., & Wiechmann, A. (2011). Role of parental stress on pediatric feeding disorders. *Children's Health Care, 40*, 85–100. https://doi.org/10.1080/02739615.2001.564557

Echeverria, F., & Miltenberger, R. G. (2013). Reducing rapid eating in adults with intellectual disabilities. *Behavioral Interventions, 28*, 131–142.

Favell, J. E., McGimsey, J. F., & Jones, M. L. (1980). Rapid eating in the retarded. *Behavior Modification, 4*, 481–492.

Fernand, J., Saksena, K., Penrod, B., & Fryling, M. (in press). Feeding disorders. In J. Matson (Ed.), *Handbook of child psychopathology & developmental disabilities: treatment*. New York, NY: Springer.

Field, D., Garland, M., & Williams, K. (2003). Correlates of specific childhood feeding problems. *Journal of Paediatrics and Child Health, 39*, 299–304.

Greer, A. J., Gulotta, C. S., Masler, E. A., & Laud, R. B. (2008). Caregiver stress and outcomes of children with pediatric feeding disorders treated in an intensive interdisciplinary program. *Journal of Pediatric Psychology, 33*, 612–620. https://doi.org/10.1093/jpepsy/jsm116

Gulotta, C. S., Piazza, C. C., Patel, M. R., & Layer, S. A. (2005). Using food redistribution to reduce packing in children with severe food refusal. *Journal of Applied Behavior Analysis, 38*, 39–50. https://doi.org/10.1901/jaba.2005.168-03

Hanh, T. N., & Cheung, L. (2011). *Savor: Mindful eating, mindful life*. New York, NY: Harper.

Hendy, H. M., Williams, K. E., Camise, T. S., Eckman, N., & Hedemann, A. (2009). The parent mealtime action scale (PMAS). Development and association with children's diet and weight. *Appetite, 52*, 328–339.

Iwata, B. A., Dorsey, M. F., Slifer, K. J., Bauman, K. E., & Richman, G. S. (1994). Toward a functional analysis of self-injury. *Journal of Applied Behavior Analysis, 27*, 197–209. (Reprinted from *Analysis and Intervention in Developmental Disabilities, 2,* 3–20, 1982).

Kahng, S., Tarbox, J., & Wilke, A. E. (2001). Use of a multicomponent treatment for food refusal. *Journal of Applied Behavior Analysis, 34*, 93–96.

Kerwin, M. E. (1999). Empirically supported treatments in pediatric psychology: Severe feeding problems. *Journal of Pediatric Psychology, 24*, 193–214.

Kodak, T., & Piazza, C. C. (2008). Assessment and behavioral treatment of feeding and sleeping disorders in children with autism spectrum disorders. *Child & Adolescent Psychiatric Clinics of North America, 17*, 887–905. https://doi.org/10.1016/j.chc.2008.06.005

Lang, R., Mulloy, A., Giesbers, S., Pfeiffer, B., Delaune, E., Didden, R., … O'Reilly, M. (2011). Behavioral interventions for rumination and operant vomiting in individuals with intellectual disabilities: A systematic review. Research in Developmental Disabilities 32, 2193–2205. doi:https://doi.org/10.1016/j.ridd.2011.06.011.

Lennox, D. B., Miltenberger, R. G., & Donnelly, D. R. (1987). Response interruption and DLR for the reduction of rapid eating. *Journal of Applied Behavior Analysis, 20*, 279–284.

Levin, D. S., Volkert, V. M., & Piazza, C. C. (2014). A multi-component treatment to reduce packing in children with feeding and autism spectrum disorders. *Behavior Modification, 38*, 940–963. https://doi.org/10.1177/0145445514550683

Luiselli, J. K. (2015). Behavioral treatment of rumination: Research and clinical applications. *Journal of Applied Behavior Analysis, 48*, 707–711. https://doi.org/10.1002/jaba.221

Lukens, C. T., & Linscheid, T. R. (2008). Development and validation of an inventory to assess mealtime behavior problems in children with autism. *Journal of Autism and Developmental Disorders, 38*, 342–352.

Matson, J. L., & Kuhn, D. (2001). Identifying feeding problems in mentally retarded persons: Development and reliability of a screening tool of feeding problems (STEP). *Research in Developmental Disabilities, 21*, 165–172.

Meier, A. E., Fryling, M. J., & Wallace, M. D. (2012). Using high-probability foods to increase the acceptance of low-probability foods. *Journal of Applied Behavior Analysis, 45*, 149–153. https://doi.org/10.1901/jaba.2012.45-149

Munk, D. D., & Repp, A. C. (1994). Behavioral assessment of feeding problems of individuals with severe disabilities. *Journal of Applied Behavior Analysis, 27*, 241–250.

Najdowski, A. C., Wallace, M. D., Reagon, K., Penrod, B., Higbee, T. S., & Tarbox, J. (2010). Utilizing a home-based parent training approach in the treatment of food selectivity. *Behavioral Interventions, 25*, 89–107. https://doi.org/10.1002/bin.298

Najdowski, A. D., Wallace, M. D., Penrod, B., Tarbox, J., Reagon, K., & Higbee, T. S. (2008). Caregiver-conducted experimental functional analyses of inappropriate mealtime behavior. *Journal of Applied Behavior Analysis, 41*, 459–465. https://doi.org/10.1901/jaba.2008.41-459

Page, S. V., Griffith, K., & Penrod, B. (2017). Reduction of rapid eating in an adolescent female with autism. *Behavior Analysis in Practice, 10*, 87–91.

Patel, M., Reed, G. K., Piazza, C. C., Mueller, M., Bachmeyer, M. H., & Layer, S. A. (2007). Use of a high-probability instructional sequence to increase compliance to feeding demands in the absence of escape extinction. *Behavioral Interventions, 22*, 305–310. https://doi.org/10.1002/bin.251

Patel, M. R., Piazza, C. C., Layer, S. A., Coleman, R., & Swartzwelder, D. M. (2005). A systematic evaluation of food textures to decrease packing and increase oral intake in children with pediatric feeding disorders. *Journal of Applied Behavior Analysis, 38*, 89–100. https://doi.org/10.1901/jaba.2005.161-02

Patel, M. R., Reed, G. K., Piazza, C. C., Bachmeyer, M. H., Layer, S. A., & Pabico, R. S. (2006). An evaluation of a high-probability instructional sequence to increase acceptance of food and decrease inappropriate behavior in children with pediatric feeding disorders. *Research in Developmental Disabilities, 27*, 430–442.

Peterson, K. M., Piazza, C. C., & Volkert, V. M. (2016). A comparison of a modified sequential approach to an applied behavior-analytic approach in the treatment of food selectivity in children with autism spectrum disorder. *Journal of Applied Behavior Analysis, 49*, 485–511.

Piazza, C. C. (2008). Feeding disorders and behavior: What have we learned? *Developmental Disabilities Research Reviews, 14*, 174–181. https://doi.org/10.1002/ddrr.22

Piazza, C. C., Fisher, W. W., Brown, K. A., Shore, B. A., Patel, M. R., Katz, R. M., … Blakely-Smith, A. (2003). Functional analysis of inappropriate mealtime behavior. *Journal of Applied Behavior Analysis, 36*, 187–204.

Piazza, C. C., Patel, M. R., Gulotta, C. S., Sevin, B. M., & Layer, S. A. (2003). On the relative contributions of positive reinforcement and escape extinction in the treatment of food refusal. *Journal of Applied Behavior Analysis, 36*, 309–324.

Piazza, C. C., Patel, M. R., Santana, C. M., Goh, H., Delia, M. D., & Lancaster, B. M. (2002). An evaluation of simultaneous and sequential presentation of preferred and non-preferred food to treat food selectivity. *Journal of Applied Behavior Analysis, 35*, 259–270. https://doi.org/10.1901/jaba.2002.35-259

Reed, G. K., Piazza, C. C., Patel, M. R., Layer, S. A., Bachmeyer, M. H., Bethke, S. D., & Gutshall, K. A. (2004). On the relative contributions of noncontingent reinforcement and escape extinction in the treatment of food refusal. *Journal of Applied Behavior Analysis, 37*, 27–42.

Riordan, M. M., Iwata, B. A., Wohl, M. K., & Finney, J. W. (1980). Behavioral treatment of food refusal and selectivity in developmentally disabled children. *Applied Research in Mental Retardation, 1*, 95–112.

Rivas, K. M., Piazza, C. C., Roane, H. S., Volkert, V. M., Stewart, V., Kadey, H. J., & Groff, R. A. (2014). Analysis of self-feeding in children with feeding disorders. *Journal of Applied Behavior Analysis, 47*, 710–722. https://doi.org/10.1002/jaba.170

Schreck, K. A., Williams, K., & Smith, A. F. (2004). A comparison of eating behaviors between children with and without autism. *Journal of Autism and Developmental Disorders, 34*, 433–438.

Seiverling, L., Williams, K., & Sturmey, P. (2010). Assessment of feeding problems in children with autism spectrum disorder. *Journal of Developmental and Physical Disabilities, 22*, 401–413. https://doi.org/10.1007/s10882-010-9206-0

Seubert, C., Fryling, M. J., Wallace, M. D., Jimenez, A. R., & Meier, A. E. (2014). Antecedent interventions for pediatric feeding problems. *Journal of Applied Behavior Analysis, 47*, 449–453. https://doi.org/10.1002/jaba.117

Sevin, B. M., Gulotta, C. S., Sierp, B. J., Rosica, L. A., & Miller, L. J. (2002). Analysis of response covariation among multiple topographies of food refusal. *Journal of Applied Behavior Analysis, 35*, 65–68.

Sharp, W. G., Harker, S., & Jaquess, D. L. (2010). Comparing bite presentation methods in the treatment of food refusal. *Journal of Applied Behavior Analysis, 43*, 739–743.

Sharp, W. G., Jaquess, D. L., Morton, J. F., & Herzinger, C. V. (2010). Pediatric feeding disorders: A quantitative synthesis of treatment outcomes. *Clinical Child and Family Psychology Review, 13*, 348–365.

Silbaugh, B. C., Penrod, B., Whelen, C. M., Hernandez, D. A., Wingate, H. V., Falcomata, T. S., & Lang, R. (2016). A systematic synthesis of behavioral interventions for food selectivity of children with autism spectrum disorders. *Research Journal in Autism and Developmental Disorders, 3*, 345–347.

Silbaugh, B. C., Swinnea, S., & Penrod, B. (2018). Synthesis of applied behavior analytic interventions for packing in pediatric feeding disorders. *Behavior Modification, 42*, 249–272. https://doi.org/10.1177/0145445517724541

Twachtman-Reilly, J., Amaral, S. C., & Zebrowski, P. P. (2008). Addressing feeding disorders in children on the autism spectrum in school-based settings: Physiological and behavioral issues. *Language, Speech, and Hearing Services in Schools, 39*, 261–272.

Valentino, A. L., LeBlanc, L. A., & Raetz, P. B. (2018). Evaluation of stimulus intensity fading on reduction of rapid eating in a child with autism. *Journal of Applied Behavior Analysis, 51*, 177–182.

Vaz, P. C. M., Piazza, C. C., Stewart, V., Volkert, V. M., & Groff, R. A. (2012). Using a chase to decrease packing in children with feeding disorders. *Journal of Applied Behavior Analysis, 45*, 97–105. https://doi.org/10.1901/jaba.2012.45-97

Vazquez, M., Fryling, M. J., & Hernandez, A. (in press). Assessing the treatment acceptability of behavioral interventions for food selectivity. *Behavior Modification, 43*, 273. https://doi.org/10.1177/0145445517751435

Volkert, V. M., Peterson, K. M., Zeleny, J. R., & Piazza, C. C. (2014). A clinical protocol to increase chewing and assess mastication in children with feeding disorders. *Behavior Modification, 38*, 705–729. https://doi.org/10.1177/0145445514536575

Volkert, V. M., & Piazza, C. C. (2012). Pediatric feeding disorders. In P. Sturmey & M. Herson (Eds.), *Handbook of evidence-based practice in clinical psychology* (Child and adolescent disorders) (Vol. 1, pp. 323–337). Hoboken, NJ: Wiley.

Volkert, V. M., Piazza, C. C., Vaz, P. C. M., & Frese, J. (2013). A pilot study to increase chewing in children with feeding disorders. *Behavior Modification, 37*, 391–408. https://doi.org/10.1177/0145445512474295

Volkert, V. M., Vaz, P. C. M., Piazza, C. C., Frese, J., & Barnett, L. (2011). Using a flipped spoon to decrease packing in children with feeding disorders. *Journal of Applied Behavior Analysis, 44*, 617–621. https://doi.org/10.1901/jaba.2011.44.617

Wilder, D. A., Normand, M., & Atwell, J. (2005). Noncontingent reinforcement as treatment for food refusal and associated self-injury. *Journal of Applied Behavior Analysis, 38*, 549–553.

Wright, C. S., & Vollmer, T. R. (2002). Evaluation of a treatment package to reduce rapid eating. *Journal of Applied Behavior Analysis, 35*, 89–93.

35

Psychological and Educational Approaches to the Treatment of Aggression and Tantrums in People with Intellectual Disabilities

Russell Lang, Tonya Davis, Katherine Ledbetter-Cho, Laurie McLay, Patricio Erhard, and Mac Kenzie Wicker

Aggression and tantrumming are relatively common topographies of challenging behavior. Aggressive behaviors are acts with the potential to do physical harm to another person such as hitting, kicking, and biting. In some cases, verbal threats of harm and aggressive language – even when not accompanied by violent physical acts – may also be considered aggression (Crotty, Doody, & Lyons, 2014). The term tantrum can be used to describe some combination of screaming, crying, thrashing body on the floor, property destruction, and other similar behaviors that tend to be loud, disruptive, and potentially dangerous. Tantrums may include aggressive behavior: for example, a child may flail on the ground, scream, and attempt to hit caregivers when they approach.

Most people will engage in minor aggression or tantrumming at some point in life, probably during childhood. In people without intellectual disability (ID), however, these behaviors are more common, more likely to persist over time, and more likely to cause serious harm (Cooper et al., 2009; O'Dwyer et al., 2018; Ruddick, Davies, Bacarese-Hamilton, & Oliver, 2015; Schroeder et al., 2014). Indeed, severity of ID appears to be among the more reliable early predictors for the emergence of aggression and tantrumming (e.g., Arias, Gomez, Moran, Alcedo, Monsalve, & Fontanil, 2018; O'Dwyer et al., 2018; Powis & Oliver, 2014). Estimates of the percentage of individuals with ID that present with challenging behavior vary a great deal depending on sample characteristics (e.g., age, disability severity, gender), definitions of challenging behavior, and how challenging behavior is measured (e.g., direct observation or parent report) (Davies & Oliver, 2013; Powis & Oliver, 2014; Ruddick et al., 2015). For example, Hardiman and McGill (2018) completed a systematic review of 28 studies that estimated the prevalence of challenging behaviors – including aggression and tantrumming – in people with

R. Lang (✉) · P. Erhard · M. K. Wicker
Department of Special Education, Clinic for Autism Research Evaluation and Support, Texas State University, San Marcos, TX, USA
e-mail: russlang@txstate.edu

T. Davis
Department of Educational Psychology, Baylor University, Waco, TX, USA

K. Ledbetter-Cho
Department of Special Education, Clinic for Autism Research Evaluation and Support, Texas State University, San Marcos, TX, USA

The University of Texas at Austin, Austin, TX, USA

L. McLay
The University of Canterbury, Christchurch, New Zealand

J. L. Matson (ed.), *Handbook of Intellectual Disabilities*, Autism and Child Psychopathology Series, https://doi.org/10.1007/978-3-030-20843-1_35

fragile X syndrome (i.e., the leading genetic cause of ID; Mazzocco, 2000) and reported a range from 12.5% to 60.9%. Davies and Oliver (2013) conducted a meta-analysis of 11 studies wherein the prevalence of aggression was measured in samples of adults and/or children. They reported that the risk of aggression appears to increase with age until sometime between the late teenage years and early twenties, when prevalence tends to level off on average. Adults with ID and a history of sexual or physical victimization and comorbid mental illness appear to be at particular risk for emergence of aggression (Clark, Crocker, & Morin, 2016). In childhood, deficits in language and communication also appear to be notable risk factors for aggression, tantrumming, and other challenging behaviors (McClintock, Hall, & Oliver, 2003; Schroeder et al., 2014; Wacker, 1998).

Persistent severe aggression and tantrumming may limit a person's opportunities for social inclusion and detract from quality of life (Brosnan & Healy, 2011; Lloyd & Kennedy, 2014; Tranfaglia, 2011). Quality of life is conceptualized as a collection of factors including the extent to which a person is able to (a) exercise self-determination (e.g., set their own goals and make choices), (b) access typical social and community settings, and (c) achieve physical and emotional well-being (Schalock & Verdugo, 2002). All of these areas may be impeded by aggression and tantrumming at any point across an individual's life-span. Additionally, severe challenging behaviors – particularly aggression – may result in (a) injury to the individual with ID and their family or service providers, (b) higher rates of turnover and job dissatisfaction in teachers and direct care staff, and (c) increased family member stress (Hensel, Lunsky, & Dewa, 2012; Knotter, Stams, Moonen, & Wissink, 2016). Children who engage in these behaviors have an increased risk of poor educational outcomes in school, and, if unresolved by adulthood, adults with ID face an increased risk of incarceration (Taylor, 2002).

Given the prevalence of aggression and tantrumming among people with ID and the associated adverse outcomes, it is not surprising that parents, teachers, and others who work to educate and care for people with ID tend to identify the reduction of these behaviors to be among the more important treatment goals (e.g., Pittenger, Barahona, Cavalari, Parent, Luiselli, & DuBard, 2014). Fortunately, there are several intervention options for treating challenging behavior with a notable amount of support in the research base. This chapter covers the psychological and educational treatments for aggression and tantrumming in people with ID with the most supportive and robust evidence. Specifically, this chapter focuses on behavioral intervention consisting of functional assessment followed by a function-matched treatment.

Applied Behavior Analysis-Based Treatment Approaches

Applied behavior analysis (ABA) involves the application of findings from the experimental analysis of behavior to address problems of social significance (Baer, Wolf, & Risely, 1968). ABA-based intervention approaches emphasize the interaction between the individual and their environment as opposed to psychopharmacological approaches that can be said to focus on a person's internal biology. There have been a number of reviews and meta-analyses focused on the treatment of challenging behavior in general and of aggression specifically, but none focused exclusively on the treatment of tantrums. Taylor (2002) conducted a narrative review of studies related to the treatment of anger and aggression in adult offenders with ID and concluded that behavioral interventions appeared to be more effective than psychotropic medication. Brosnan and Healy (2011) reviewed 18 studies wherein behavioral interventions were used in an effort to reduce aggression in people 3 to 18 years old with ID and reported that all 18 studies resulted in a decrease in aggressive behavior. Similarly, Fossum, Handegard, Adolfsen, Vis, and Wynn (2016) conducted a meta-analysis of 56 studies involving the treatment of aggression in people with and without ID and found that a majority of studies ($n = 34$) utilized behavioral intervention approaches and that corpus of studies yielded a medium effect size for the reduction of aggression. Although there are no reviews or meta-analyses focused exclusively on tantrumming, reviews focused on challenging behavior in general have reached similar conclusions regarding the effectiveness of behavioral intervention for people with ID (e.g., Lloyd & Kennedy, 2014).

Operant Learning Theory

ABA arose from a translation of basic operant learning research, which is typically conducted in laboratory settings, to applied research conducted in naturalistic settings (Sidman, 2011). Specifically, the underlying basic research elucidated how a change in a behavior's probability is influenced by past consequences in the context of an individual's current environment (Skinner, 1953). Consequences that increase the probability of a behavior's future occurrence are referred to as reinforcers, and consequences that reduce that probability are termed punishers. Elements of the environment that signal when reinforcement is available are called discriminative stimuli (Cooper, Heron, & Heward, 2007). According to operant learning theory, behaviors persist when the immediate result is at least occasionally a reinforcing consequence (Skinner). In terms of challenging behavior, ABA-based interventions alter the antecedent environment and/or the reinforcement contingencies (i.e., consequences) associated with targeted challenging behaviors. Therefore, function-based treatment is a two-step process wherein the first step is to identify relevant interactions between the individual and their environment via functional behavioral assessment (FBA). The second step is to design an intervention that incorporates this information in an effort to eliminate the challenging behavior (Hurl, Wightman, Haynes, & Virues-Ortega, 2016; Lang et al., 2013).

Consequences that may function as reinforcement for challenging behavior can be grouped into three categories: (a) socially mediated positive reinforcement, (b) socially mediated negative reinforcement, and (c) automatic reinforcement. Socially mediated functions include all of the ways people respond to another person's challenging behavior. For example, a teacher may approach and reprimand a child who hits a peer in the classroom and, if that consequence (i.e., obtaining teacher attention) is reinforcing, then the likelihood of the child hitting again the next time he wants teacher attention increases. In such a case, aggression is maintained by the contingency between hitting and obtaining socially mediated positive reinforcement in the form of teacher attention. Even though the intent of the reprimand was to decrease aggression, the behavior will increase if the reprimand (a form of attention) is reinforcing (Piazza et al., 1999). Several types of social consequences like praise, reprimands, physical contact (e.g., hugs, high fives), delivery or removal of tangibles, and even closer physical proximity have the potential to function as reinforcers or punishers, and individualized FBA is often necessary to determine how specific contingencies influence behavior for a given individual.

Lang et al. (2014) provide an illustrative example in the treatment of tantrum behavior in two children between 4 and 5 years of age with ID and autism spectrum disorder. A modified pairwise preference assessment (Fisher et al., 1992) was conducted wherein picture cards depicting various forms of social interaction were presented to participants who then indicated their preference for a form of social interaction by touching the corresponding picture card. This process identified a high- and low-preferred form of social interaction for each child. The different topographies of contingent attention (e.g., "high five" and praise) where then compared during an educational intervention (i.e., discrete trial training) using an alternating treatment design. When the high-preferred form of attention was made contingent on appropriate behavior, correct responding on instructional tasks increased, and tantrumming behavior decreased relative to a condition where the lower-preferred topography of attention was contingent.

Socially mediated contingencies are referred to as positive reinforcement when behavior results in obtaining or accessing preferred stimuli (people, objects, activities) which in turn causes the behavior to occur more often in the future (Cooper et al., 2007). For example, a challenging behavior that results in obtaining a snack from mom or being allowed to sit near a preferred peer in school may be maintained by positive reinforcement. Socially mediated contingencies are referred to as negative reinforcement when undesirable stimuli are removed or reduced and the behavior increases. The term "negative," in this context, refers to the removal of stimuli, not a reduction in behavior. For example, a child who has a tantrum because she does not want to clean up, a man who hits staff in order to escape a noisy room, and a teenager who destroys their book to avoid homework are engaging in challenging behavior with a negative reinforcement function.

Automatic reinforcement differs from social-mediated reinforcement because other people are not involved in giving or removing of stimuli (Vollmer, 1994). Specifically, automatic reinforcement does not require action from another person because the behavior directly produces reinforcement. Automatic reinforcement may be in the form of a pleasant sensation or reduction of an aversive sensation. Although automatic reinforcement has been shown to have the potential to maintain self-injurious behavior (Iwata, Dorsey, Slifer, Bauman, & Richman, 1994, Iwata et al., 1994), there are far fewer instances of aggression and tantrums being maintained by automatic reinforcement contingencies in the research base (Brosnan & Healy, 2011; Kurtz, Chin, Robinson, O'Connor, & Hagopian, 2015). For additional discussion of challenging behaviors more likely to involve automatic reinforcement (e.g., stereotypy and self-injurious behavior), see Chapters 32 and 33. In the following sections of this chapter, approaches to assessing the functional properties of aggression and tantrums are presented. Next, a range of behavioral intervention options will be identified with emphasis on how interventions should be aligned with identified function(s).

Functional Behavior Assessment

The first step of a function-based approach to the treatment of aggression and tantrumming involves conducting a FBA. This is an essential first step because FBA results inform the selection and adjustment of specific treatment procedures (Anderson, Rodriguez, & Campbell, 2015). For example, a child with an FBA suggesting he hits peers in order to obtain attention (i.e., socially mediated positive reinforcement) would not receive the same intervention as a child with FBA results suggesting he hit peers in order to be removed from the classroom (i.e., socially mediated negative reinforcement). In the former case, some examples of function-matched treatment include (a) teaching the child other more appropriate ways to obtain attention (i.e., replacement behavior), (b) withholding teacher attention following aggression (extinction), (c) providing attention following periods without aggression, or (d) providing attention before the child hits (antecedent intervention). However, in the case of hitting maintained by escape from the classroom, examples of function-matched treatment might include (a) teaching a more appropriate way to request a break (replacement behavior), (b) not allowing the child to leave the room following hitting (escape extinction), (c) scheduling more work breaks, and (d) reducing the averseness of tasks to reduce motivation to escape. Although both cases involved the same topography of aggression (hitting), intervention procedures are tailored by alignment with the behavior's function.

There are a number of techniques for identifying function, and there is no standardized FBA process agreed to be best practice. Practitioners must select FBA procedures by considering characteristics of the individual being assessed and their specific context (e.g., assessment environment, type of challenging behavior, available resources). FBA assessment methods fall into three categories: indirect methods, direct observation methods, and experimental methods. Most FBAs will include a combination of these methods because each has specific strengths and limitations.

Indirect methods. Indirect FBA methods gather data from informants rather than via direct observation of the person engaging in challenging behavior (Anderson et al., 2015). Indirect methods include interviews, questionnaires, and rating scales that are completed by teachers, parents, and other caregivers who know the individual well. Specifically, the informant is asked to consider instances when they observed the person engaged in aggression, tantrumming, or some other challenging behavior and then endorse a degree of agreement or disagreement with statements on Likert scales. For example, a teacher might endorse strong agreement with a statement indicating the person "engages in the behavior to get attention" and strong disagreement with "engages in the behavior to escape work or learning situations" which would suggest that social-mediated positive reinforcement may be maintaining aggression. Advantages of indirect approaches are that questionnaires and similar tools are typically easy to administer and can be completed in a relatively short amount of time. These factors may be particularly appealing to professionals who lack the resources, expertise, or time to implement direct methods (Floyd, Phanuef, & Wilczynski, 2005).

Although research evaluating the technical adequacy of these instruments is somewhat scant (Dufrene, Kazmerski, & Labrot, 2017; Floyd et al., 2005), three semi-structured interviews have accumulated a notable amount of research support: the Functional Assessment Interview (FAI; O'Neil et al., 1997), the Functional Assessment Checklist: Teachers and Staff (FACTS), and the Functional Assessment Interview Record for Teachers (FAIR-T; Edwards, 2002). For school-age children, in particular, The FACTS and FAIR-T may be most appropriate because they were designed for school settings with teachers and school staff serving as informants. The FAI can be implemented across settings and, in addition to school personnel, other direct service staff and family members have served as informants (O'Neil et al., 1997). The FACTS and FAIR-T have been evaluated more extensively with typically developing children, while the FAI has been evaluated with people with and without an ID (Dufrene et al., 2017).

Similar to interviews, a variety of questionnaires and rating scales with empirical support are available. Options with the most research support include the Motivation Assessment Scale (MAS; Durand & Crimmins, 1988), the Questions About Behavioral Function (QABF; Matson, Tureck, & Rieske, 2012), and the Contextual Assessment Inventory for Problem Behavior (CAI; McAtee, Carr, & Shulte, 2004). All three of these rating scales were designed for individuals with intellectual and/or developmental disabilities (Dufrene et al., 2017). The MAS and QABF have the best available evidence, demonstrating sufficient, inter-rater reliability, internal consistency, and temporal stability (Dufrene et al., 2017).

Whenever possible, indirect methods should be utilized in combination with other FBA procedures, including both direct observations and experimental methods, because the likelihood of successfully identifying the function of aggression from a single indirect assessment is lower (Alter, Conroy, Mancil, & Haydon, 2008; Fee, Schieber, Noble, & Valdovinos, 2016). Although indirect methods alone may not be sufficient in some cases, they are still useful early in the overall FBA process. Specifically, responses from informants who know the person well (e.g., parents, direct care staff, and teachers) may inform timing and procedures used during direct observations and experimental analysis methods conducted later in the FBA process (Dufrene et al., 2017; Fee et al., 2016; Hall, 2005; O'Neil et al., 1997).

Direct observation. Direct observation involves watching the person in environments where aggression or tantrumming occurs and collecting data on (a) events that tend to precede occurrences of the challenging behavior (antecedents), (b) changes that occur contingently and immediately after the challenging behavior (consequences), and (c) frequency of occurrence (Anderson et al., 2015). Common direct observation methods include antecedent-behavior-consequence (ABC) recording, structural analysis, and scatterplots. ABC recording involves noting antecedent events observed immediately before and consequence events occurring immediately after the occurrence of challenging behavior. Practitioners then review this running record to identify potential patterns related to antecedents and consequences that suggest a specific function (Bijou, Peterson, & Ault, 1968). The ABC recording method is among the most frequently implemented direct observation FBA approaches (Anderson et al., 2015).

Another common direct observation method is a structural analysis (Anderson et al., 2015). To conduct a structural analysis, practitioners observe the targeted behavior in two or more predetermined, but naturally occurring, contexts (e.g., group work versus independent work). Similar to the ABC recording, practitioners then evaluate patterns arising from the data in an effort to identify the behavior's function (Anderson et al., 2015; Stitcher, Lewis, Johnson, & Trussell, 2004). Finally, a scatterplot involves recording occurrences of the behavior across time by dividing an observation time into equal intervals (e.g., 48 30-min intervals per day). The frequency of challenging behavior per interval can then be used to highlight the times of day challenging behavior is most common. The activity scheduled at that time of day, staff on duty, and other schedule and environmental factors can then be compared across intervals where challenging behavior is most frequent and when it is least likely. This approach may help inform the scheduling of future ABC recording observations, provide a baseline measure of the behavior's frequency, and, by noting environmental correlates with challenging behavior, provide clues regarding function (Touchette, MacDonald, & Langer, 1985). For example, a scatterplot that reveals tantrumming occurs most often right before dinner but is rare after meals would suggest that challenging behavior is maintained by access to food. To further explore that possibility, the practitioner could conduct ABC recording before and after dinner to note whether access to food may be the function. In such a case, the practitioner might note that the group home's cook began taking food from the refrigerator right before the tantrum started (discriminative stimuli) and, upon seeing the tantrum, gave the individual a snack (reinforcing consequence). Treatment for challenging behavior with this pattern of antecedent and consequence could include teaching the individual how to request food appropriately (i.e., functional communication training) and/or modification of the antecedent environment (e.g., increasing availability of snacks or moving dinnertime to earlier in the evening).

Direct observation methods require more time and expertise than indirect methods, but they tend to lead to more accurate conclusions than indirect methods, which are more susceptible to informant bias (Anderson & Long, 2002). However, beyond additional expertise and time, there are several other limitations associated with direct observation. Most importantly, direct observation alone can only provide correlational data, precluding practitioners from making causal statements regarding the environment-behavior relationship (Mace, Lalli, & Shea, 1992). Further, targeted challenging behavior may occur infrequently during observations, making correlational hypotheses difficult to develop (Lerman & Iwata, 1993). In many cases, a combination of indirect interviews and questionnaires in tandem with direct observation is sufficient to design a successful treatment for challenging behavior. However, when these approaches are insufficient or impractical due to resource constraints (e.g., limited time) and in cases where these methods do not produce clear results or when intervention fails to reduce the challenging behavior, a more rigorous FBA protocol that includes a functional analysis is indicated (Beavers, Iwata, & Lerman, 2013; Hanley, Iwata, & McCord, 2003).

Functional analysis. Instead of merely observing and attempting to identify patterns that hint at behavioral function, a functional analysis involves systematically programming conditions with contrived antecedents and contingencies and then measuring changes in behavior across these predetermined conditions (Anderson et al., 2015; Hanley et al., 2003). Iwata and colleagues (1982/1994) developed the most widely cited functional analysis procedures. Iwata et al.'s method involved a control condition (unstructured play) and three test conditions (i.e., attention/social disapproval, academic demands, alone). Although many variations and modifications to these conditions have been evaluated, those options are based on the conditions originally described by Iwata et al. and preserve the underlying operant logic (Lydon, Healy, O'Reilly & Lang, 2012). Procedures for each of Iwata et al.'s original FA condition are summarized in Table 35.1.

The procedures described in Table 35.1 have been modified in a number of ways in order to improve functional analysis efficiency or suitability for a specific individual (Lydon et al., 2012). For example, Iwata et al.'s (1982/1994) conditions were 15 min each, and the process involved running each condition multiple times. The overall duration of the functional analysis process can be decreased by reducing the duration of conditions (e.g., 15 min to 5 min) and is then referred to as a brief functional analysis (MacDonald et al., 2002; Northup et al., 1991). Other variations of functional analysis procedures include delivery of additional test conditions (e.g., tangible condition where preferred items are provided contingent on challenging behavior) and the use of protective equipment to prevent injury during assessment of aggressive or self-injurious behavior (see Beavers et al., 2013 and Lydon et al., 2012 for reviews of functional analysis procedural variations).

Unlike indirect and observation-only methods, a functional analysis allows for causal inferences regarding the function of a targeted behavior and is therefore more likely to yield accurate results (Didden, Duker, & Korzilius, 1997; Hurl et al., 2016). Although a large corpus of rigorous single-case design studies have demonstrated the success of treatments based on functional analysis results (Beavers et al., 2013), functional analysis methods are not without criticism. For example, they may be time-consuming and require rare expertise, and there is concern that occasioning and reinforcing targeted aggression during test conditions could pose additional risk of harm to the individual being assessed or the assessor. As with any such assessment, practitioners must weigh the risks and benefits when determining whether a functional analysis should be included in the FBA process for their client (Mueller & Nkosi, 2007). More recent approaches such as trial-based functional analyses (Sigafoos

Table 35.1 Overview of the functional analysis test and control conditions from Iwata et al. (1982/1994)

Condition	Purpose	Antecedent arrangement	Contingent consequence
Social disapproval	To measure the rate of challenging behavior that occurs when the contingency is socially mediated positive reinforcement	Assessor pretends to ignore the individual (withhold attention) by engaging in some other activity (e.g., reading a magazine)	Social interaction is made contingent on challenging behavior. Specifically, if the individual engages in challenging behavior (e.g., begins tantrumming), the assessor moves closer and reprimands. All other behavior is ignored
Academic demands	To measure the rate of challenging behavior that occurs when the contingency is socially mediated negative reinforcement	Assessor instructs the individual to complete a task that has been reported or observed to be non-preferred (e.g., doing homework)	The task materials and request to do the task are removed, and the assessor turns or moves away from the individual contingent on challenging behavior. All other behavior has no influence on requirement to complete task
Alone	To measure the rate of challenging behavior that occurs when there is no socially mediated reinforcement available	Assessor is not in the room, and the individual is alone	There are no socially mediated contingencies programmed for this condition. If the behavior occurs at high rate, it is likely maintained by automatic reinforcement
Unstructured play	Serves as a control condition to empower comparison of rates of challenging behavior across conditions	Assessor provides pleasant social interaction, does not make any demands, and provides free access to preferred items (e.g., toys)	There are no socially mediated contingencies programmed for this condition. If the behavior is maintained by socially mediated reinforcement, then challenging behavior should be low or absent in this condition

& Saggers, 1995) and interview-informed synthesized contingency analyses are appearing more often in the literature and may offer additional advantages, particularly for practitioners in complex applied settings (e.g., Slaton & Hanley, 2018). Regardless of FBA components utilized, an accurate and thorough assessment of a behavior's operant function is often critical to successful intervention to reduce aggression and tantrumming (Hurl et al., 2016; Mace, 1994).

Function-Based Intervention Components

After operant functions maintaining aggressive or tantrumming behavior are identified via FBA, behavioral intervention components aligned with the FBA results are selected (Smith, 2011). Therefore, in terms of selection of intervention components, a behavior's operant function is a more important consideration than the behavior's form or appearance (Dunlap & Carr, 2007; Petscher, Rey, & Bailey, 2009; Pinkelman & Horner, 2017). In general, function-based interventions can effectively reduce challenging behavior by (a) altering the antecedent environment to reduce motivation to engage in challenging behavior, (b) removing discriminative stimuli that signal reinforcement for challenging behavior, and (c) altering reinforcement contingencies such that people respond to challenging behavior differently (Dunlap & Fox, 2011).

Antecedent-Based Intervention Components

Antecedent interventions involve making changes to the environment where challenging behavior typically occurs in order to reduce the individual's motivation for reinforcers maintaining challenging behavior and remove or alter discriminative stimuli that occasion challenging behavior. When possible these changes are aligned with the results of the FBA process such that the function of the challenging behavior is incorporated. For example, Rispoli et al. (2011) conducted a functional analysis of tantrumming behavior (i.e., throwing work materials) of a 5-year-old boy with autism and ID and found that access to specific tangibles (crayons and paper) was the maintaining contingency. An alternating treatment design was then used to compare a condition in which the child had free access to those items prior to receiving academic instruction to a condition in which access was restricted. When the child had access to the items in the antecedent condition, the frequency of tantrumming was lower and rates of academic engagement were higher than in sessions following the restricted access condition.

Noncontingent reinforcement. Noncontingent reinforcement (NCR) involves the delivery of the reinforcer maintaining a challenging behavior on a time-based, response-independent schedule such that the individual is less motivated to engage in challenging behavior in order to access the reinforcer and the contingency between behavior and reinforcement is broken (Richman, Barnard-Brak, Grub, Bosch, & Abby, 2015; Vollmer, Iwata, Zarcone, Smith, & Mazaleski, 1993). Specifically, because the reinforcer is no longer contingent on the occurrence of the behavior, the behavior is rendered less useful to the individual, and the scheduled access may reduce the value of the reinforcer and abate behavior maintained by that reinforcer (Carr, Bailey, Ecott, Lucker, & Weil, 1998; Rispoli et al., 2011). In terms of aggression, the schedule of reinforcer delivery may need to be denser in the beginning of intervention and then gradually faded over time as aggression is reduced (Richman et al., 2015), particularly for older individuals capable of doing more physical damage.

Britton, Carr, Kellum, Dozier, and Weil (2000) implemented NCR with a 41-year-old male who engaged in grabbing, hair pulling, and biting. A functional analysis indicated Victor's aggression was maintained by provision of attention in the form of physical contact (e.g., hand-holding). Forms of physical contact identified as appropriate and reinforcing for Victor were provided at set time intervals independent of the occurrence of aggression. Initially, the NCR schedule was based on the mean latency to the first occurrence of aggression during baseline. However, after three consecutive sessions in which aggression occurred in less than 5% of intervals, the NCR schedule was thinned from every 50 s to delivery every 6 min. Results indicated that NCR successfully reduced aggression, and the schedule thinning procedure maintained low levels of aggression over time.

NCR can also be applied to challenging behaviors maintained by escape or avoidance of unwanted or unpleasant stimuli. For example, NCR to treat aggression maintained by socially mediated negative reinforcement involves removing the undesired stimulus or aversive demand on a time-based schedule. For example, scheduled breaks from undesired tasks (e.g., academic work, clean up, work demands) can be provided. This approach is potentially effective regardless of whether the individual engages in aggression or tantrumming as long as the challenging behavior's function is to avoid or escape.

Altering antecedent environments and discriminative stimuli. In addition to NCR, there are other antecedent approaches to reducing aggression or tantrums with notable support in the research base. Langthorne, McGill, and Oliver (2014) conducted a systematic review of 59 studies involving challenging behavior maintained by negative reinforcement. In terms of intervention studies targeting aggressive or tantrumming behaviors in people with ID, several antecedent-based strategies were identified including (a) adding discriminative stimuli associated with positive reinforcement to the antecedent environment, (b) reducing the difficulty or averseness of task demands, (c) offering choices for how or when to complete task demands, and (d) altering the mode of demand presentation.

McComas, Hoch, Paone, and El-Roy (2000) provide an example of reducing the disruptive tantrumming of an 8-year-old boy with autism and ID. In that study, the presentation of an academic task demand was found to occasion the occurrence of the challenging behavior, and escape from that task demand was identified as the maintaining consequence. The disruptive tantrums were then reduced, and compliance to task demands increased by offering more effective instructional strategies and academic supports (e.g., provided with calculator and number line). Improved academic instruction has also been found to reduce aggression maintained by negative reinforcement for older children with mild ID (Lali, Kates & Casey, 1999). An example of the potential benefit of altering schedule to include additional breaks is provided by Aikman, Garbutt, and Furniss (2003) who reduced the tantrumming (i.e., screaming and throwing materials) of an 8-year-old boy with severe ID by providing breaks for the child to play as opposed to a more continuous expectation for academic work. Crockett and Hagopian (2006) provide one of the few examples of this approach with an adult. Specifically, the disruptive tantrumming of a 19-year-old with mild ID and Nager's syndrome was reduced by providing a wider range of prompts.

Consequence-Based Intervention Components

Instead of focusing intervention procedures on the antecedent environment before aggressions or tantrumming occurs, consequence-based intervention components alter how the environment responds after challenging behavior occurs. Typically, these approaches involve some combination of ensuring reinforcement is not provided for challenging behavior (i.e., extinction) while also providing reinforcement contingent on an appropriate replacement behavior.

Extinction. Extinction is the discontinuation or withholding of reinforcement following a target behavior, and, when applied with fidelity, extinction can be used to reduce the frequency of the preceding behavior (Keller & Schoenfeld, 1950/1995). In order to be function-matched, extinction procedures differ based on the challenging behavior's socially mediated function. Aggression maintained by socially mediated positive reinforcement can be treated with extinction because practitioners and caregivers can control access to delivery of attention or tangibles. O'Reilly, Lancioni, and Taylor (1999) implemented extinction for aggression in a 10-year-old boy with ID who engaged in a variety of aggressive behaviors including biting, scratching, and hitting. A functional analysis indicated these behaviors were all maintained by provision of attention; therefore, treatment involved withholding attention; specifically no eye contact or verbal interactions were provided following aggression. During the first two sessions, aggression increased above baseline levels, but in subsequent sessions quickly decreased to zero levels.

Aggression maintained by socially mediated negative reinforcement can also be treated with extinction when the practitioner can control the presence of the aversive stimulus. This form of extinction is typically referred to as escape extinction (Cooper et al., 2007). To implement escape extinction, a practitioner would no longer remove or reduce task demands contingent upon aggression. Anderson and Long (2002) demonstrated escape extinction to reduce several problem behaviors, including aggression in an 8-year-old boy with autism and moderate to severe ID. Problem behaviors were maintained by escape and were highest during educational tasks with a speech pathologist. Escape extinction was therefore implemented within this context. Specifically, when the child engaged in aggression during the educational tasks, the speech pathologist continued to prompt him to complete the task and did not reduce or remove the demand. Escape extinction (i.e. persisting with the task demand) resulted in reduction of challenging behavior from three to four times per minute on average to less than once per minute across the last four sessions.

Several characteristics of a behavior's response to extinction have been discussed in the literature. These include concerns regarding extinction burst, spontaneous recovery, and undesirable indirect effects (Lerman & Iwata, 1996). An extinction burst is a temporary increase followed by a decrease in the frequency of a behavior when exposed to extinction (Lerman, Iwata, & Wallace, 1999). Although practitioners should consider the possibility of an extinction burst when using extinction to treat aggression, it is worth noting that extinction bursts do not occur in all instances of extinction (Lerman & Iwata, 1995). Further, when an extinction burst does occur, it is typically brief, lasting only a few sessions (Lerman & Iwata, 1996). Spontaneous recovery is the temporary reappearance of a behavior that has been previously reduced by extinction. Although spontaneous recovery has been demonstrated in basic research, much less evidence of spontaneous recovery is evident in the applied literature. In fact, instances of potential spontaneous recovery may actually reference other phenomena such as treatment that failed to generalize across other individuals or settings (Lerman & Iwata, 1996). Finally, indirect undesirable effects on non-targeted behaviors may occur. These other behaviors have been described as *agitated* or *emotional* behaviors and include topographies such as crying, disruption, and complaining. Although such side effects of extinction are possible and should be considered when planning treatment, these undesirable outcomes also tend to decrease with continued exposure to extinction (e.g., Goh & Iwata, 1994). Moreover, these phenomena may be mediated if extinction is utilized as a component of a treatment package, rather than a stand-alone treatment. Extinction is rarely utilized in isolation and is typically included within a multicomponent treatment package that also programs novel reinforcement contingencies (Vollmer & Athens, 2011).

Reinforcement Procedures

Differential reinforcement. Differential reinforcement is a two-component process in which the practitioner (a) provides reinforcement contingent upon an appropriate behavior targeted for increase but (b) withholds reinforcement for challenging behavior (i.e., extinction) with the same function. When used to decrease aggression, for example, differential reinforcement involves reinforcing occurrences of an alternative to aggression while placing aggression on extinction. If the behavior targeted for increase is physically incompatible with aggression such that the two behaviors cannot co-occur, the procedure is referred to as differential reinforcement of incompatible behavior (DRI). Friman and Altman (1990) treated high-rate disruptive behavior (tantrumming) in a 4-year-old boy with severe ID by teaching his parents to implement DRI. The child's tantrums included elopement and throwing objects. Therefore, reinforcement (i.e., praise and small edibles) was delivered contingent on the child staying in his seat for specified periods of time that gradually increased across intervention sessions. When the child left his seat, he was redirected back to the chair, and no programmed reinforcement was provided. Tantrumming (i.e., out of seat and throwing objects) reduced from multiple occurrences per minute in baseline to less than once per minute during initial intervention sessions and then eventually to zero in the last two sessions.

Differential reinforcement of alternative behavior (DRA) is similar to DRI except the behavior selected for increase is not physically incompatible with the targeted challenging behavior, and the behavior targeted for increase is aligned with the function of the challenging behavior. For example, if FBA results suggest that aggression is maintained by socially mediated positive reinforcement, the DRA procedure will involve teaching the individual to gain access to that consequence (e.g., tangible items, attention) via an alternative behavior. Roane, Fisher, Sgro, Falcomata, and Pabico (2004) implemented DRA with Carl, an 11-year-old boy with autism and ID who engaged in pinching, biting, and grabbing others. After a functional analysis concluded Carl's aggression was maintained by access to tangibles, the therapist taught Carl to give him a picture card as a replacement behavior to access preferred items while aggression was placed on extinction. This resulted in decreased rate of aggression and increased rate of picture card exchange.

If FBA results indicate aggression is maintained by socially mediated negative reinforcement (e.g., escape from undesired activities) a DRA may involve teaching the individual to emit a socially acceptable behavior to access a break. This is referred to as differential negative reinforcement of an alternative behavior (DRNA; Vollmer & Iwata, 1992). Vollmer, Roane, Ringdahl, and Marcus (1999) implemented DRNA with two participants with ID that engaged in aggression maintained by escape from demands. During DRNA, if the participant complied with a task demand (i.e., replacement behavior), the therapist provided a 30-second break from the task. During that 30-s break, the therapist moved away from the table and discontinued any prompts to engage in work. Aggression was placed on extinction. The DRNA procedure resulted in decreased aggression and increased task demand compliance.

When selecting replacement behaviors for differential reinforcement, practitioners should consider several factors (Dwyer, Rozewski, & Simonsen, 2012). First, the practitioner should select a replacement behavior that is already within the individual's repertoire, if possible. If not possible, then the replacement behavior should be one that is easy to teach. Similarly, the replacement behavior should require less effort than aggression. In other words, it should be physically easier for the individual to engage in the replacement behavior than to engage in aggression or tantrumming. Finally, the practitioner should select a replacement behavior that is likely to be reinforced in the natural environment. For example, hand raising is a behavior that is likely to be reinforced with attention in the classroom.

Functional communication training. Functional communication training (FCT) is a type of DRA in which the replacement behavior is a form of communication (Tiger et al., 2008). The communicative response may involve speech but may also involve augmentative or alternative communication (AAC) such as manual signs, picture exchange, or speech-generating devices (Tiger, Hanley, & Bruzek, 2008). Carr and Durand (1985) taught children with ID to produce vocal requests to access consequences maintaining challenging behavior. Specifically, children with challenging behavior maintained by attention were taught to request attention by asking the teacher, "Am I doing good work?" and children with challenging behavior maintained by escape from demands were taught to tell the teacher, "I don't understand". Praise and additional assistance with the task was then provided following these requests. Similarly, Hanley, Iwata, and Thompson (2001) implemented FCT with a 34-year-old man with an ID who engaged in several topographies of aggression: biting, pinching, and twisting limbs, along with self-injurious behavior. A functional analysis concluded these behaviors were maintained by access to preferred items. Experimenters successfully taught Jake to access those preferred items utilizing a manual sign while placing challenging behavior on extinction.

Radstaake et al. (2013) assisted a classroom teacher in implementing FCT with Amy, a 7-year-old girl with Angelman syndrome and an ID. Amy pinched others among other topographies of challenging behavior. A functional analysis concluded that these challenging behaviors were maintained by escape from tasks. As a result, the teacher successfully taught Amy a form of picture exchange communication (Bondy & Frost, 2001) to request a time-out from undesired tasks. In all of the above examples, challenging behavior decreased as the communicative replacement behavior increased. After replacement behaviors have been acquired and the aggression and tantrumming reduced, effort to thin reinforcement schedules such that the behavior is supported by natural reinforcement contingencies is necessary (for review of schedule thinning, see Hagopian, Boelter, & Jarmolowicz, 2011).

Conclusion

Aggression and tantrumming behaviors are common topographies of challenging behavior identified in samples of people with ID across the life-span. These behaviors pose considerable risk to the individual's quality of life, physical safety, and future opportunities. The psychoeducational treatments with the most support in the research base involve first conducting a FBA that includes some combination of indirect as well as direct measures and observations of how the individual interacts with their environment. After the function (reinforcing consequence) of challenging behavior is identified, intervention components are individualized accordingly. This two-step process has been found effective with individuals with a range of ID severity during childhood, adolescence, and adulthood. Although other educational and psychological approaches have been investigated and can be said to have a promising body of emerging evidence, interventions based on ABA are the only options that have accumulated sufficient scientific evidence to be considered evidence-based practices. Future research in this area should include replication and extension studies focused on older individuals because the overwhelming majority of studies have included participants in early adulthood or younger. Further, interventions that have been found effective with other populations but that lack sufficient evidence for people with ID (e.g., cognitive behavioral therapy, relaxation, and mindfulness) warrant additional consideration.

References

Aikman, G., Garbutt, V., & Furniss, F. (2003). Brief probes: A method for analyzing the function of disruptive behaviour in the natural environment. *Behavioural and Cognitive Psychotherapy, 31*, 215–220.

Alter, P. J., Conroy, M. A., Mancil, G. R., & Haydon, T. (2008). A comparison of functional behavior assessment methodologies with young children: Descriptive methods and functional analysis. *Journal of Behavioral Education, 17*, 200–219. https://doi.org/10.1007/s10864-008-9064-3

Anderson, C. M., & Long, E. S. (2002). Use of a structured descriptive assessment methodology to identify variables affecting problem behavior. *Journal of Applied Behavior Analysis, 35*, 13–154. https://doi.org/10.1901/jaba.2002.35-137

Anderson, C. M., Rodriguez, B. J., & Campbell, A. (2015). Functional behavior assessment in schools: Current status and future directions. *Journal of Behavioral Education, 24*, 338–371. https://doi.org/10.1007/s10864—015-9226-z

Arias, V. B., Gomez, L. E., Moran, L. M., Alcedo, A. M., Monsalve, A., & Fontanil, F. (2018). Does quality of life differ for children with autism spectrum disorder and intellectual disability compared to peers without autism? *Journal of Autism and Developmental Disorders, 48*, 123–136.

Baer, D. M., Wolf, M. M., & Risley, T. R. (1968). Some still-current dimensions of applied behavior analysis. *Journal of Applied Behavior Analysis, 20*, 313–327.

Beavers, G. A., Iwata, B. A., & Lerman, D. C. (2013). Thirty years of research on the functional analysis of problem behavior. *Journal of Applied Behavior Analysis, 46*, 1–21. https://doi.org/10.1002/jaba.30

Bijou, S. W., Peterson, R. F., & Ault, M. H. (1968). A method to integrate descriptive and experimental field studies at the level of data and empirical concepts. *Journal of Applied Behavior Analysis, 1*, 175–191. https://doi.org/10.1901/jaba.1968.1-175

Bondy, A., & Frost, L. (2001). The picture exchange communication system. *Behavior Modification, 25*, 725–744.

Britton, L. N., Carr, J. E., Kellum, K. K., Dozier, C. L., & Weil, T. M. (2000). A variation of noncontingent reinforcement in the treatment of aberrant behavior. *Research in Developmental Disabilities, 21*, 425–435. https://doi.org/10.1016/S0891-4222(00)00056-1

Brosnan, J., & Healy, O. (2011). A review of behavioral interventions for the treatment of aggression in individuals with developmental disabilities. *Research in Developmental Disabilities, 32*, 437–446.

Carr, E. G., & Durand, V. (1985). Reducing behavior problems through functional communication training. *Journal of Applied Behavior Analysis, 18*, 111–126. https://doi.org/10.1901/jaba.1985.18-111

Carr, J. E., Bailey, J. S., Ecott, C. L., Lucker, K. D., & Weil, T. M. (1998). On the effects of noncontingent delivery of differing magnitudes of reinforcement. *Journal of Applied Behavior Analysis, 31*, 313–321. https://doi.org/10.1901/jaba.1998.31-313

Clark, M., Crocker, A. G., & Morin, D. (2016). Victimization history and aggressive behavior among adults with intellectual disabilities: The mediating role of mental health. *International Journal of Forensic Mental Health, 15*, 301–311.

Cooper, J. O., Heron, T. E., & Heward, W. L. (2007). *Applied behavior analysis*. Upper Saddle River, NJ: Pearson.

Cooper, S. A., Smiley, E., Jackson, A., Finlayson, J., Allan, J., Mantry, D., & Morrison, J. (2009). Adults with intellectual disabilities: Prevalence, incidence and remission of aggressive behaviour and related factors. *Journal of Intellectual Disability Research, 53*, 217–232.

Crockett, J. L., & Hagopian, L. (2006). Prompting procedures as establishing operations for escape-maintained behavior. *Behavioral Interventions, 21*, 65–71.

Crotty, G., Doody, O., & Lyons, R. (2014). Aggressive behaviour and its prevalence within five typologies. *Journal of Intellectual Disabilities, 18*, 76–89.

Davies, L., & Oliver, C. (2013). The age related prevalence of aggression and self-injury in persons with intellectual disability: A review. *Research in Developmental Disabilities, 34*, 764–775.

Didden, R., Duker, P., & Korzilius, H. (1997). Meta-analytic study on treatment effectiveness for problem behaviors with individuals who have mental retardation. *American Journal on Mental Retardation, 101*(4), 387–399.

Dufrene, B. A., Kazmerski, J. S., & Labrot, Z. (2017). The current status of indirect functional assessment instruments. *Psychology in the Schools, 54*, 331–350. https://doi.org/10.1002/pits.22006

Dunlap, G., & Carr, E. (2007). Positive behavior support and developmental disabilities. In S. Odom, R. Horner, M. Snell, & J. Blacher (Eds.), *Handbook of developmental disabilities* (pp. 469–482). New York, NY: Guilford Press.

Dunlap, G., & Fox, L. (2011). Function-based interventions for children with challenging behavior. *Journal of Early Intervention, 33*, 333–343.

Durand, M., & Crimmins, D. B. (1988). Identifing the variables maintaining self-injurious behavior. *Journal of Autism and Developmental Disorders, 18*, 99–117.

Dwyer, K., Rozewski, D., & Simonsen, B. (2012). A comparison of function-based replacement behaviors for escape-motivated students. *Journal of Emotional and Behavioral Disorders, 20*, 115–125.

Edwards, R. P. (2002). A tutorial for using the functional assessment informant record for teachers. *Proven Practice, 4*, 31–33.

Fee, A., Schieber, E., Noble, N., & Valdovinos, M. G. (2016). Agreement between questions about behavior function, the motivation assessment scale, functional assessment interview, and brief functional analysis of children's challenging behaviors. *Behavior Analysis: Research and Practice, 16*, 94–102. https://doi.org/10.1037/bar000040

Fisher, W., Piazza, C. C., Bowman, L. G., Hagopian, L. P., Owens, J. C., & Slevin, I. (1992). A comparison of two approaches for identifying reinforcers for persons with severe and profound disabilities. *Journal of Applied Behavior Analysis, 25*, 491–498.

Floyd, R. G., Phanuef, R. L., & Wilczynski, S. M. (2005). Measurement properties of indirect assessment methods for functional behavioral assessment: A review of research. *School Psychology Review, 34*, 58–73.

Fossum, S., Handegard, B. H., Adolfsen, F., Vis, S. A., & Wynn, R. (2016). A meta-analysis of long-term outpatient treatment effects for children and adolescents with conduct problems. *Journal of Child and Family Studies, 25*, 15–29.

Friman, P. C., & Altman, K. (1990). Parent use of DRI on high-rate disruptive behavior: Direct and collateral benefits. *Research in Developmental Disabilities, 11*, 249–254.

Goh, H., & Iwata, B. A. (1994). Behavioral persistence and variability during extinction of self-injury maintained by escape. *Journal of Applied Behavior Analysis, 27*, 173–174. https://doi.org/10.1901/jaba.1994.27-173

Hagopian, L. P., Boelter, E. W., & Jarmolowicz, D. P. (2011). Reinforcement schedule thinning following functional communication training: Review and recommendations. *Behavior Analysis in Practice, 4*, 4–16. https://doi.org/10.1007/BF03391770

Hall, S. S. (2005). Comparing descriptive, experimental, and informant-based assessments of problem behaviors. *Research in Developmental Disabilities, 26*, 514–526. https://doi.org/10.1016/j.ridd.2004.11.004

Hanley, G. P., Iwata, B. A., & McCord, B. E. (2003). Functional analysis of problem behavior: A review. *Journal of Applied Behavior Analysis, 36*, 147–185. https://doi.org/10.1901/jaba.2003.36-147

Hanley, G. P., Iwata, B. A., & Thompson, R. H. (2001). Reinforcement schedule thinning following treatment with functional communication training. *Journal of Applied Behavior Analysis, 34*, 17–38. https://doi.org/10.1901/jaba.2001.34-17

Hardiman, R. L., & McGill, P. (2018). How common are challenging behaviours amongst individuals with fragile x syndrome. *Research in Developmental Disabilities, 76*, 99–109.

Hensel, J. M., Lunsky, Y., & Dewa, C. S. (2012). Exposure to client aggression and burnout among community staff who support adults with intellectual disabilities in Ontario, Canada. *Journal of Intellectual Disabilities Research, 56*, 910–915.

Hurl, K., Wightman, J., Haynes, S. N., & Virues-Ortega, J. (2016). Does a pre-intervention functional assessment increase intervention effectiveness? A meta-analysis of within-subject interrupted time-series of studies. *Clinical Psychology Review, 47*, 71–84.

Iwata, B. A., Dorsey, M. F., Slifer, K. J., Bauman, K. E., & Richman, G. S. (1994). Toward a functional analysis of self-injury. *Journal of Applied Behavior Analysis, 27*, 197–209. (Reprinted from *Analysis and Intervention in Developmental Disabilities, 2,* 3–20, 1982).

Iwata, B. A., Pace, G. M., Dorsey, M. F., Zarcone, J. R., Vollmer, T. R., Smith, R. G., … Willis, K. D. (1994). The functions of self-injurious behavior: An experimental epidemiological analysis. *Journal of Applied Behavior Analysis, 27*, 215–240.

Iwata, B. A., Dorsey, M. F., Slifer, K. J., Bauman, K. E., & Richman, G. S. (1994). Toward a functional analysis of self-injury. *Journal of Applied Behavior Analysis, 27*, 197–209.

Keller, F. S., & Schoenfeld, W. M. (1950/1995). *Principles of psychology*. Acton, MA: Copley Publishing Group.

Knotter, M. H., Stams, G. J. J. M., Moonen, X. M. H., & Wissink, I. B. (2016). Correlates of direct care staffs' attitudes towards aggression of persons with intellectual disabilities. *Research in Developmental Disabilities, 59*, 294–305.

Lalli, J. S., Kates, K., & Casey, S. D. (1999). Response covariation: The relationship between correct academic responding and problem behavior. *Behavior Modification, 23*, 339–357.

Lang, R., Sigafoos, J., van der Meer, L., O'Reilly, M. F., Lancioni, G. E., & Didden, R. (2013). Early signs and early behavioral intervention of challenging behavior. In R. Hastings & J. Rojahn (Eds.), *Challenging behavior. International review of research in developmental disability*, 44, (pp. 1–35). London: Elsevier Inc., Academic Press.

Lang, R., van der Werff, M., Verbeek, K., Didden, R., Davenport, K., Moore, M., … Lancioni, G. (2014). Comparison of high and low preferred topographies of contingent attention during discrete trial training. *Research in Autism Spectrum Disorders, 8*, 1279–1286.

Langthorne, P., McGill, P., & Oliver, C. (2014). The motivating operation and negatively reinforced problem behavior: A systematic review. *Behavior Modification, 38*, 107–159.

Lerman, D. C., & Iwata, B. A. (1993). Descriptive and experimental analysis of variables maintaining self-injurious behavior. *Journal of Applied Behavior Analysis, 26*, 293–319. https://doi.org/10.1901/jaba.1993.26-293

Lerman, D. C., & Iwata, B. A. (1995). Prevalence of the extinction burst and its attenuation during treatment. *Journal of Applied Behavior Analysis, 28*, 93–94. https://doi.org/10.1901/jaba.1995.28-93

Lerman, D. C., & Iwata, B. A. (1996). Developing a technology for the use of operant extinction in clinical settings: An examination of basic and applied research. *Journal of Applied Behavior Analysis, 29*, 345–382. https://doi.org/10.1901/jaba.1996.29-345

Lerman, D. C., Iwata, B. A., & Wallace, M. D. (1999). Side effects of extinction: Prevalence of bursting and aggression during treatment of self-injurious behavior. *Journal of Applied Behavior Analysis, 32*, 1–8. https://doi.org/10.1901/jaba.1999.32-1

Lloyd, B., & Kennedy, C. H. (2014). Assessment and treatment of challenging behavior for individuals with intellectual disability: A research review. *Journal of Applied Research in Intellectual Disabilities, 27*, 187–199.

Lydon, S., Healy, O., O'Reilly, M. F., & Lang, R. (2012). Sensory integration therapy for autism spectrum disorders: A systematic review. *Research in Autism Spectrum Disorders, 6*, 1004–1018.

MacDonald, J. E., Wilder, D. A., & Dempsey, C. (2002). Brief functional analysis and treatment of eye poking. *Behavioral Interventions, 17*, 261–270.

McComas, J. J., Hoch, H., Paone, D., & El-Roy, D. (2000). Escape behaviour during academic tasks: A preliminary analysis of idiosyncratic establishing conditions. *Journal of Applied Behavior Analysis, 33*, 479–493.

Mace, F. C. (1994). The significance and future of functional analysis methodologies. *Journal of Applied Behavior Analysis, 27*, 385–392. https://doi.org/10.1901/jaba.1994.27-385

Mace, F. C., Lalli, J. S., & Shea, M. C. (1992). Functional analysis and treatment in self-injury. In J. Luiselli, J. Matson, & N. Singh (Eds.), *Assessment, analysis, and treatment of self-injury* (pp. 122–152). New York: Springer-Verlag.

Matson, J. L., Tureck, K., & Rieske, R. (2012). The questions about behavioral function (QABF): Current status as a method of functional assessment. *Research in Developmental Disabilities, 33*, 630–634. https://doi.org/10.1016/j.ridd.2011.11.006

Mazzocco, M. M. (2000). Advances in research on the fragile X syndrome. *Mental Retardation and Developmental Disabilities Research Reviews, 6*, 96–106.

McAtee, M., Carr, E. G., & Shulte, C. (2004). A contextual assessment inventory for problem behavior: Initial development. *Journal of Positive Behavioral Interventions, 6*, 148–165. https://doi.org/10.1177/10983007040060030301

McClintock, K., Hall, S., & Oliver, C. (2003). Risk markers associated with challenging behaviours in people with intellectual disabilities: A meta-analytic study. *Journal of Intellectual Disability Research, 47*, 405–416.

Mueller, M. M., & Nkosi, A. (2007). State of the science in the assessment and management of severe behavior problems in school settings: Behavior analytic consultation to schools. *International Journal of Behavioral Consultation and Therapy, 3*, 176–201. https://doi.org/10.1037/h0100798

Northup, J., Wacker, D., Sasso, G., Steege, M., Cigrand, K., Cook, J., & DeRaad, A. (1991). A brief functional analysis of aggressive and alternative behavior in an outclinic setting. *Journal of Applied Behavior Analysis, 24*, 509–522. https://doi.org/10.1901/jaba.1991.24-509

O'Dwyer, C., McCallion, P., Burke, E., Carroll, R., O'Dwyer, M., & McCarron, M. (2018). Prevalence and associated factors of problem behaviours among older adults with intellectual disabilities in Ireland. *Research in Developmental Disabilities, 80*, 192–204.

O'Neil, R. E., Horner, R. H., Albin, R. W., Sprague, J. R., Storey, K., & Newston, J. S. (1997). *Functional assessment and program development for problem behavior: A practical handbook* (2nd ed.). Pacific Grove, CA: Brooks/Cole.

O'Reilly, M., Lancioni, G., & Taylor, I. (1999). An empirical analysis of two forms of extinction to treat aggression. *Research in Developmental Disabilities, 20*, 315–325. https://doi.org/10.1016/S0891-4222(99)00013-X

Petscher, E. S., Rey, C., & Bailey, J. S. (2009). A review of empirical support for differential reinforcement of alternative behavior. *Research in Developmental Disabilities, 30*, 409–425. https://doi.org/10.1016/j.ridd.2008.08.008

Piazza, C. C., Bowman, L. G., Contrucci, S. A., Delia, M. D., Adelinis, J. D., & Goh, H. (1999). An evaluation of the properties of attention as reinforcement for destructive and appropriate behavior. *Journal of Applied Behavior Analysis, 32*, 437–449.

Pinkelman, S. E., & Horner, R. H. (2017). Improving implementation of function-based interventions: Self-monitoring, data collection, and data review. *Journal of Positive Behavior Interventions, 19*, 228–238. https://doi.org/10.1177/1098300716683634

Pittenger, A., Barahona, C., Cavalari, R. N. S., Parent, V., Luiselli, J. K., & DuBard, M. (2014). Social validity assessment of job satisfaction, resources, and support among educational service practitioners with intellectual and developmental disabilities. *Journal of Developmental and Physical Disabilities, 26*, 737–745.

Powis, L., & Oliver, C. (2014). The prevalence of aggression in genetic syndromes: A review. *Research in Developmental Disabilities, 35*, 1051–1071.

Radstaake, M., Didden, R., Lang, R., O'Reilly, M., Sigafoos, J., Lancioni, G. E., … Curfs, L. M. G. (2013). Functional analysis and functional communication training in the classroom for three children with Angelman syndrome. *Journal of Developmental and Physical Disabilities, 25*, 49–63. https://doi.org/10.1007/s10882-012-9302-4

Richman, D. M., Barnard-Brak, L., Grub, L., Bosch, A., & Abby, L. (2015). Meta-analysis of noncontingent reinforcement effects on problem behavior. *Journal of Applied Behavior Analysis, 48*, 131–152. https://doi.org/10.1002/jaba.189

Rispoli, M., O'Reilly, M., Lang, R., Machalicek, W., Davis, T., Lancioni, G. E., & Sigafoos, J. (2011). Effects of motivating operations on problem and academic behavior in classrooms. *Journal of Applied Behavior Analysis, 44*, 187–192.

Roane, H. S., Fisher, W. W., Sgro, G. M., Falcomata, T. S., & Pabico, R. R. (2004). An alternative method of thinning reinforcer delivery during differential reinforcement. *Journal of Applied Behavior Analysis, 37*, 213–218. https://doi.org/10.1901/jaba.2004.37-213

Ruddick, L., Davies, L., Bacarese-Hamilton, M., & Oliver, C. (2015). Self-injurious, aggressive and destructive behaviour in children with severe intellectual disability: Prevalence, service need and service receipt in the UK. *Research in Developmental Disabilities, 46*, 3017–3315.

Schalock, R. L., & Verdugo, M. A. (2002). Moving us toward a theory of individual quality of life. *American Journal on Intellectual and Developmental Disabilities, 121*, 1–12.

Schroeder, S. R., Marquis, J. G., Reese, M., Richman, D. M., Mayo-Ortega, L., Oyama-Ganiko, R., … Lawrence, L. (2014). Risk factors for self-injury, aggression, and stereotyped behavior among young children at risk for intellectual and developmental disabilities. *American Journal of Intellectual and Developmental Disability, 119*, 351–370.

Sidman, M. (2011). Can an understanding of basic research facilitate the effectiveness of practitioners? Reflections and personal perspectives. *Journal of Applied Behavior Analysis, 44*, 973–991.

Sigafoos, J., & Saggers, E. (1995). A discrete-trial approach to the functional analysis of aggressive behavior in two boys with autism. *Australia & New Zealand Journal of Developmental Disabilities, 20*, 287–297. https://doi.org/10.1080/07263869500035621

Skinner, B. F. (1953). *Science and human behavior*. New York: Macmillan.

Slaton, J. D., & Hanley, G. P. (2018). Nature and scope of synthesis in functional analysis and treatment of problem behavior. *Journal of Applied Behavior Analysis, 51*, 1–31. early online.

Smith, R. G. (2011). Developing antecedent interventions for problem behavior. In W. W. Fisher, C. C. Piazza, & H. S. Roane (Eds.), *Handbook of applied behavior analysis* (pp. 297–316). New York: The Guildford Press.

Stitcher, J. P., Lewis, T., Johnson, N., & Trussell, R. (2004). Toward a structural assessment: Analyzing the merits of an assessment tool for students with E/BD. *Assessment for Effective Intervention, 30*, 25–40. https://doi.org/10.1177/153450841245521

Taylor, J. L. (2002). A review of the assessment and treatment of anger and aggression in offenders with intellectual disability. *Journal of Intellectual Disability Research, 46*, 57–73.

Tiger, J. H., Hanley, G. P., & Bruzek, J. (2008). Functional communication training: A review and practical guide. *Behavior Analysis in Practice, 1*, 16–23.

Touchette, P. E., MacDonald, R. F., & Langer, S. N. (1985). A scatter plot for identifying stimulus control of problem behavior. *Journal of Applied Behavior Analysis, 18*, 343–351. https://doi.org/10.1901/jaba.1985.18-343

Tranfaglia, M. R. (2011). The psychiatric presentation of fragile X: Evolution of the diagnosis and treatment of the psychiatric comorbidities of fragile X syndrome. *Developmental Neuroscience, 33*, 337–348.

Vollmer, T., Roane, H., Ringdahl, J., & Marcus, B. (1999). Evaluating treatment challenges with differential reinforcement of alternative behavior. *Journal of Applied Behavior Analysis, 32*, 9–23.

Vollmer, T. R. (1994). The concept of automatic reinforcement: Implications for behavioral research in developmental disabilities. *Research in Developmental Disabilities, 15*, 187–207.

Vollmer, T. R., & Athens, E. (2011). Developing function-based extinction procedures for problem behavior. In W. W. Fisher, C. C. Piazza, & H. S. Roane (Eds.), *Handbook of applied behavior analysis* (pp. 317–334). New York: The Guilford Press.

Vollmer, T. R., & Iwata, B. A. (1992). Differential reinforcement as treatment for behavior disorders: Procedural and functional variations. *Research in Developmental Disabilities, 13*, 393–417. https://doi.org/10.1016/0891-4222(92)90013-V

Vollmer, T. R., Iwata, B. A., Zarcone, J. R., Smith, R. G., & Mazaleski, J. L. (1993). The role of attention-maintained self-injurious behavior: Noncontingent reinforcement and differential reinforcement of other behavior. *Journal of Applied Behavior Analysis, 26*, 9–21. https://doi.org/10.1901/jaba.1994.27-331

Wacker, D. P., Berg, W. K., Harding, J. W., Derby, M. K., Asmus, J. M., & Healy, A. (1998). Evaluation and long-term treatment of aberrant behavior displayed by young children with disabilities. *Journal of Developmental and Behavioral Pediatrics, 19*, 260–266.

36

Use of Medications in the Treatment of Aggressive Behavior

Lauren Charlot, Rory Sheehan,
and Angela Hassiotis

Introduction

Psychotropic medications have been used to treat aggression in various populations, including youth with antisocial behaviors, people with traumatic brain injury, and elderly patients with dementia (Maher et al., 2011). However, individuals with intellectual and developmental disabilities (IDD) including people with an autism spectrum disorder (ASD) are perhaps the most medicated group of all of these populations (Esbensen, Greenberg, Seltzer, & Aman, 2009). The extensive use of psychotropic medications, especially antipsychotic medications, has been linked to the frequency with which aggressive and other challenging behaviors are part of the presenting problem that brings individuals with IDD to clinical attention (Tsakanikos, Costello, Holt, Sturmey, & Bouras, 2007). The topic is important given that aggressive behavior is associated with poor quality of life, can lead to out of home placement, and in general limits independence and the ability to be integrated into one's community (Koch et al., 2015). In this chapter, following a brief review of the general topic of aggressive behaviors of people with IDD, the use of psychotropic medications to treat aggressive challenging behaviors (ACB) is described including prescription patterns, research regarding efficacy, side effects, and major findings informing best practice guidelines.

Definition and Epidemiology of Aggressive Behavior

Aggressive behavior can take many forms, from self-directed aggression, i.e., self-injurious behavior, to outwardly directed behavior such as aggressive behavior aimed at people or destructive behavior toward objects. While not currently a diagnostic entity in the DSM-5 (American Psychiatric Association, 2013), it is nonetheless considered a potential comorbidity in people with IDD, and the ICD-10 does provide a diagnostic code for both "repetitive self-injury" and "aggression toward others" (World Health Organization, 1993). By definition challenging behavior "may threaten the quality of life and/or the physical safety of the individual or others and is likely to lead to responses that are restrictive,

---

L. Charlot
Center for S.T.A.R.T. Services, University of New Hampshire, Institute on Disabilities,
Concord, NH, USA

R. Sheehan
UCL Division of Psychiatry, Maple House,
London, UK

A. Hassiotis (✉)
UCL Division of Psychiatry, Maple House,
London, UK

Camden & Islington Foundation Trust, London, UK
e-mail: a.hassiotis@ucl.ac.uk

J. L. Matson (ed.), *Handbook of Intellectual Disabilities*, Autism and Child Psychopathology
Series, https://doi.org/10.1007/978-3-030-20843-1_36

aversive or result in exclusion" (Banks, Bush, & Baker, 2007), which is perhaps never more applicable than in the case of aggressive behavior. In addition to the personal, emotional, and quality of life costs, challenging behaviors and especially aggressive behaviors are associated with more expensive forms of care.

A few studies have attempted to establish the prevalence of challenging behavior and aggressive behavior specifically. Prevalence rates of overall challenging behavior lie at 10–15% (Emerson et al., 2001). In the community, in a population-based investigation, the prevalence of aggressive challenging behavior was reported as 9.8% and the 2-year incidence as 1.8% (Cooper et al., 2009). However, figures vary with the setting and specific clinical picture of the population observed. A Canadian study of 3165 clinically referred adults with intellectual disabilities receiving rehabilitation services in Canada provided a 12-month prevalence of 51.8% for overall aggressive challenging behavior as well as the prevalence of five subtypes of aggressive behavior: property damage (24%), verbal (37.6%), physical (24.4%; 4.9% leading to injury of another person), self-injurious (24.4%), and sexual aggression (9.8%) (Crocker et al., 2006). For people with multiple disabilities and profound ID, figures have been reported to be as high as 45% for destructive or aggressive behavior and 82% for self-injurious behavior (Poppes, Van der Putten, & Vlaskamp, 2010). In a more recent study, Crocker, Prokić, Morin, and Reyes (2014) reviewed the cases of 265 adults with IDD and found that a prevalence of overall challenging behavior was 18.1%, self-injurious behavior 7.5%, aggressive-destructive behavior 8.3%, and stereotyped behavior 10.9%.

Unfortunately, persistence levels for challenging behavior are high for adults, particularly those with comorbid autism spectrum disorder (Totsika & Hastings, 2009). This is particularly true for physical aggression where in a study population which is not known to have received any intervention, it remained stable at a persistence percentage of 70% over 11 years (Totsika, Toogood, Hastings, & Lewis, 2008). In children persistence of challenging behavior is moderate to high (Totsika & Hastings, 2009), and the presence of earlier challenging behavior, as reported by parents, represents a significant risk for ongoing behavioral problems (de Ruiter, Dekker, Verhulst, & Koot, 2007). When considering the wide-reaching potential consequences in the context of the above figures on the pervasiveness and chronic course of aggressive behavior in people with intellectual disability, the importance of fully understanding associations and the possible underlying root causes becomes clear.

Predictors of Aggression

Factors that appear to predict risk for aggressive behavior in neurotypical youth are not the same as those identified in studies of children with IDD (Hill et al., 2014). Hill et al. (2014), for example, concluded that "Sociodemographic correlates of aggressive behavior in typical populations such as gender, parent education, race, and ethnicity tended to be weaker or absent in children with ASD." This suggests that root causes of aggression may differ for individuals with and without IDD.

Matson and Hess (2011) conducted a large review of studies that had examined causes for aggression and noted that most of the time, an environmental cause can be established. Cooper et al. (2009) reported that factors independently associated with aggressive behaviors in their population-based survey included lower ability, female gender, living away from home, cause of ID other than Down syndrome, presence of ADHD, and urinary incontinence. Several risk factors for aggression were identified in a meta-analytic study of individuals with IDD: male gender, severe/profound intellectual disability, communication deficits for self-injurious behavior, as well as autism spectrum disorder for self-injurious, aggressive, and disruptive behavior (McClintock, Hall, & Oliver, 2003). Risk factors for aggression displayed by children and adolescents with ASD include self-injury, sleep problems, gastrointestinal problems, communication challenges, and challenges in social functioning (Mazurek, Kanne, & Wodka, 2013).

Other factors include comorbid mental conditions such as dementia, sensory impairment, physical health problems, and abusive or restrictive environments (Citation? – from text sent to LC). Anxiety likely plays a significant role in childhood aggression (Granic, 2014) and may be an important and frequently overlooked influence for both children and adults with IDD. Specific deficits in executive functioning, for example, challenges in set shifting, may be predictive of aggressive behavior through influence on mental flexibility and need for sameness seen in many children and adults with ASD (Visser, Berger, Prins, Lantman-De Valk, & Teunisse, 2014). A number of recent studies have demonstrated that subtypes of deficits in executive function may occur more frequently in association with specific syndromes seen in conjunction with intellectual disability such as Prader-Willi syndrome (PWS) and Fragile X syndrome (FRA-X). These limitations, in turn, may impact pathways to aggressive behaviors. Individuals with PWS, for example, become angry and aggressive related to changes in routines, and this correlates with lower ability to set shift more so than measures of overall cognitive ability. Though set-shifting challenges correlate with challenging behavior in individuals with FRA-X, in this case, anxiety is a critical associated feature.

Other genetic syndromes predispose to specific health problems, such as Cornelia de Lange syndrome (CdLS). Individuals with CdLS display self-injurious behavior at elevated rates compared to counterparts who also have IDD and with other genetic causes. Frequently this is secondary to pain associated with GERD (gastroesophageal reflux disease) (Oliver, Sloneem, Hall, & Arron, 2009). Numerous studies have linked pain to aggressive behaviors displayed by people with IDD (Carr & Owen-DeSchryver, 2007; Gardner & Whalen, 1996; Kennedy et al., 2007). In general, certain genetic syndromes are associated with higher reported rates of aggressive behavior. For example, Cri du Chat, Smith-Magenis, Prader-Willi, Angelman, Cornelia de Lange, and Fragile X syndromes have rates reported of about 70%, while other syndromes, such Williams and Down syndromes, have much lower associations (less than 15%) (Powis & Oliver, 2014).

Research to date clearly indicates that a wide array of factors may influence the occurrence of aggressive behavior in people with IDD; therefore, simple explanatory models are unlikely to be useful. Also, if there are multiple factors that contribute to the occurrence of aggression, there may be a need for multiple approaches to address them.

Aggression and Mental Illness

Various rationales are used to support treatment of challenging behaviors with psychotropic medication beyond the assertion that particular antipsychotic medications alleviate states of irritability and reduce aggression in children with ASD. Aggression and other disruptive behaviors are sometimes viewed as “behavioral equivalents” of mental illness symptoms or more accurately as actual symptom substitutes. For example, certain researchers have suggested that aggression could be an equivalent of depressed mood in the absence of sadness or anhedonia in classifications of psychiatric syndromes applied to people with IDD. However, in a detailed review of the topic, it was concluded that the practice is unsupported by the evidence base and should be avoided for a number of reasons (Sturmey, Laud, Cooper, Matson, & Fodstad, 2010).

The most critical reason is that externalizing behaviors, and even most signs of dysphoric mood, are diagnostically nonspecific and therefore unlikely to be due to mental illness (Matson & Konst, 2015).

Recent work by Melville et al. (2016) and Painter, Ingham, Trevithick, Hastings, and Roy (2018) presents evidence of current conceptualizations of challenging behavior as distinct from or equivalent with mental ill health. This is consistent with early investigations where irritability and aggression were found to be common to most mental health problems experienced by a person with IDD (Charlot, Doucett, & Mezzacappa, 1993). The finding that aggressive behaviors should not be used as symptom substitutes for people with IDD is supported by the above-reviewed findings that so many factors seem to contribute to the occurrence of aggression.

Most experts have agreed that the basic criteria from accepted classification systems should be retained with small adjustments, as seen in the DM-ID 2. In the DM-ID 2, for example, in reference to depressive disorders, it was emphasized that it is important to recognize challenging behaviors as nonspecific signs of distress and to determine if they are occurring in the setting of all of the other depressive disorder criteria (i.e., anhedonia, other signs of dysphoria such as crying, difficulty falling or staying asleep, appetite changes etc.) and have a history consistent with the syndrome. Psychiatric syndromes have a typical clinical course and history and do not generally arise abruptly or appear in only certain settings. When present, symptoms of depression co-occur and are persistent and clinically significant while representing a departure from usual functioning. Further, syndromes such as major depressive disorder and bipolar disorder are episodic. All of these factors need to be considered before a challenging behavior is seen as related to the psychiatric illness, and even then, the behavior itself would be the result rather than the cause of the illness. In addition, the DSM 5 suggests that developmental profile will impact the manifestation of the symptoms criteria in ways that clinicians should know, so as not to miss identifying syndromes when present.

Aggressive behavior can become a way to escape distressing situations when a person has a mental illness like depression or a phobia, especially when one is not able to otherwise convey that the situation is experienced as aversive (Lowry & Sovner, 1992). Many individuals have unique or individualized but characteristic displays of aggressive and disruptive behaviors. Caregivers will describe how the basic outburst is similar each time and that it is the frequency and intensity that primarily shifts. In behavioral terms, depression, anxiety, or even psychosis may be a form of a setting event that will increase the probability of occurrence of a targeted challenging behavior when present during exposure to a triggering factor (Hunter, Wilkniss, Gardner, & Silverstein, 2008).

Given the fact that many people with IDD demonstrate the same kind of agitated behaviors regardless of the source of distress or etiology of their challenging behaviors, it is conceivable too that the same rating on an aggression rating scale such as the *Irritability subscale* of the ABC might also be given in all of these different situations. Because of the lack of diagnostic specificity of surface features of challenging behaviors, a model to explain aggression must then consider multiple potential influences each time there is a "flare-up" or significant change in the frequency or intensity of aggressive behavior. It would not be sufficient to have previously found one or even two factors having impact and then to assume that these factors caused the present problem. This is why functional assessments tend to extend over a long period of time and are repeated many times over the course of treating individuals using a positive behavior support plan.

Pathways to Aggressive Behavior

Complex models are needed to better understand pathways to aggression, as even the most robust predictors account for less than 50% of the variance found. The impact of factors identified as contributing risk at one time is diminished when followed longitudinally. Functional behavioral assessments, even the most structured and intense ones, also fail to account for all of the influences that seem to impact the frequency and intensity with which a challenging behavior will occur (Charlot, 2016; Unwin, 2014). Though the identification of psychiatric syndromes in people with IDD may have improved, most agree there is a large gap between research-based diagnoses and that which occurs in practice. Further, irritability and aggression have been identified as occurring in the context of every type of psychiatric disorder, so that it has virtually no diagnostic specificity. In fact, the most suitable manner of conceptualizing aggressive and other challenging behaviors in people with IDD may be that it acts as final common pathway for distress among people who have fewer ways to demonstrate this than counterparts with neurotypical developmental histories. Additionally, and for similar reasons, aggressive acts may be the most effective means available to some people, to exert control over one's environment. The motivation to try to control one's environment is a universal and healthy human drive with only the means being the true problem, when negative or dangerous patterns of behavior develop over time.

As the result of the complexity of the causes of aggression, unsurprisingly, the most promising of approaches seems to be the one that requires the most "up-front" investment of resources and time. This would be the use of comprehensive multidisciplinary assessment in which attention is paid to environmental and contextual influences on probability of occurrence of aggression (Charlot, 2016; Hunter et al., 2008). In this type of model, "internal" person factors, sometimes referred to as vulnerabilities, transact with a large array of individually significant experiences to produce outcomes. Outcomes might include the development of specific psychiatric syndrome or the emergence of a pattern of behavior (Unwin, 2014). The model is actually similar to that applied in research in developmental psychopathology regarding pathways to psychopathology in general population studies of youth (Rutter & Sroufe, 2000).

Unlike a psychiatric or medical approach, a comprehensive multidisciplinary assessment yields a case formulation that includes a broad-based functional assessment, and not simply a diagnosis. The formulation includes description of what the person brings to a given situation and then all of the varied factors that have an impact on how the person responds to his or her environment. Individuals with IDD often have a variety of genetically associated or biologically influenced challenges to health, cognitive capacities, and psychological and social domains of functioning that will affect responses. At the same time, the environments and experiences to which a person is exposed will also exert influence. In the study of aggression of people with IDD, it is widely accepted that there are multiple pathways to aggressive behavior. There is therefore unlikely to be a universal set of influences or a single form of treatment that will address the causes of aggressive behavior of individuals with IDD. There are interventions that can reduce aggressive behavior without treating its causes at all, such as any intervention, for example, that induces fatigue or slows a person down or any intervention that restricts a person's actions. Clearly, short-term safety can be an important and necessary goal in responding to severe aggression, but in the longer term, the goal must be to identify critical influences and corresponding treatments. Treatments derived from application of a transactional multifactorial model will most always involve multiple modalities including interventions aimed at the system of care, and not just the individual. The latter is in recognition of the finding that in many cases, aggression is a response to ineffective or even abusive caretaking. There may be common contributing factors as suggested by Unwin, so that performing assessments that reveal what are the specific ones affecting a given individual is a critical component of a comprehensive assessment. For example, functional communication challenges and sensory sensitivities are often identified factors influencing distress-driven agitated behaviors in people with IDD. Assessments that seek to identify possible psychiatric syndromes using classification systems with guidelines for use with people with ID are also a component. Anxiety, as another example, is probably underdiagnosed as a factor that influences aggressive responding in people with IDD. Evaluation for all possible medical irritants and medication side effects are also critical. If a genetic syndrome can be identified, clues to frequently associated features may be helpful in guiding both assessment and treatment. Cognitive assessments that include evaluation of executive functions and social cognition will also often reveal factors that act as setting events for challenging behaviors, such as EF deficits associated with rigid patterns of thinking and resistance to change or biologically mediated tendencies which seem to cause heightened seeking of social interactions.

Treatment

Multiple forms of treatment for aggressive behaviors have been reported in the literature, and several major review studies provide information to clinicians regarding the evidence base for a number of these various interventions (Fitzpatrick, Srivorakiat, Wink, Pedapati, & Erickson, 2016). ABA-based interventions for aggressive behavior have been the most studied and have the largest evidence base (Matson & LoVullo, 2008). Much of the literature on ABA focuses on children with ASD but has been reported as also helpful when applied in treating aggression in children and adults with other IDD (see Ali, Blickwedel, & Hassiotis, 2014 for an excellent overview of treatments for individuals with IDD and challenging behaviors). The main focus in this chapter is specifically the use of medication for treating aggressive challenging behavior and is reviewed in detail below. Special attention is given to the risks as well as reports of benefits of pharmaceutical interventions aimed at reducing challenging behavior.

Rates of Psychotropic Medication Use

Increasing numbers of people with IDD are treated with psychotropic medications, and polypharmacy appears to be common (Esbensen et al., 2009; Häβler, Thome, & Reis, 2015; Lott et al., 2004; Mandell et al., 2008; Spina & Leon, 2017; Valenza et al., 2017). Community samples show that large numbers of both children and adults with IDD receive psychopharmacologic treatment, whether they have any psychiatric indications for these prescriptions or not (Brophy et al., 2018; Esbensen et al., 2009; Lunsky & Modi, 2017; Thalitaya, Reynolds, & Ismail, 2017). Unlike other patient groups, people with IDD are generally referred for acute mental health care when they display aggressive or other externalizing behaviors, and their pharmacotherapy is often aimed at its reduction. This is true (that medication is targeting reduction of aggression) even when a psychiatric disorder is diagnosed (de Kuijper & Hoekstra, 2017; Matson & Neal, 2009; Sheehan & Hassiotis, 2017; Tsakanikos et al., 2007; Valenza et al., 2017). Sheehan et al. (2015) reported that only 21% of a large cohort they reviewed including over 33,000 individuals with IDD had a record of mental illness at study entry, while 25% had a record of challenging behavior. Forty-nine percent of adults without a psychiatric illness documented in their records were, however, treated with psychotropic medications (Sheehan et al., 2015).

Antipsychotic drugs are usually the most frequently prescribed class of medications in studies of people with IDD taking psychotropic drugs (Bowring, Totsika, Hastings, Toogood, & McMahon, 2017; Mandell et al., 2008). Brophy et al. (2018) noted that youth with an ASD and ID were prescribed antipsychotic medications more often, at a younger age, and for longer periods of time than a neurotypical control group. De Kuijper et al. (de Kuijper & Hoekstra, 2017) reviewed the care of individuals with IDD in the Netherlands and concluded that the prevalence of antipsychotic drug use for off-label indications in people with IDD "remains high." More and more specialists are raising concerns specifically about the extent of antipsychotic drug prescriptions for challenging behavior in both children and adults with IDD (Brophy et al., 2018; Matson & Hess, 2011). Various best practice guidelines have been outlined, but apparently, having professional or best practice guidelines does not seem to alter the actual practice (Matson & Williams, 2015). In the United Kingdom, this has prompted centralized, government-supported actions aimed at addressing the problem though no similar activity seems to be occurring in the United States (US) (England, 2015).

Recent reviews show major increases in the number of antipsychotic drugs prescribed to youth with ID and ASD particularly since 2011 (Park et al., 2016), following the introduction of new or atypical antipsychotics over the preceding two decades. Adults have been affected by the trend as well. Bowring et al. (2017) found high doses and polypharmacy to be common in their review of the prevalence of psychotropic medication use for challenging behaviors in 265 adults with IDD. These individuals were treated with an average of 2.62 medications per person, with antipsychotic medications being the most frequently prescribed class. Factors influencing prescription of antipsychotic medication included presence of a psychiatric diagnosis, challenging behavior, older age, male gender, and type of residence (Bowring et al., 2017; Lunsky & Modi, 2017; Sheehan et al., 2015). Individuals with ASD may be more likely to receive psychotropic medications if they have a psychiatric diagnosis, ADHD diagnosis, or epilepsy (Houghton, Ong, & Bolognani, 2017). Other investigations have identified factors associated with medication use to include older age and type of residence, as well as gender (males usually receiving more antipsychotic medication in some but not all studies) (Bowring et al., 2017; Lunsky & Modi, 2017).

A recently published *Autism Treatment Network* sponsored study regarding youth with ASD and antipsychotic drugs (Lake et al., 2017) reviewed the care of over 5000 children, and among the children taking antipsychotics to treat externalizing behavior challenges, one third were not receiving any behavioral therapies. Autism Speaks sponsors for the research concluded that the drugs should not be used in isolation of other forms of intervention nor as first-line treatments. Risk factors contributing to antipsychotic drug use included older age (teens versus younger children), use of multiple psychotropic medications, prior ASD diagnosis, non-white Hispanic race/ethnicity, oppositional defiant problems, and physical health conditions such as gastrointestinal and sleep problems. They emphasized the need for thorough behavioral and medical assessment for children with ASD who have co-occurring challenging behaviors, prior to attempting medication interventions.

Factor Associated with Polypharmacy

Polypharmacy appears to be on the rise (Bowring et al. 2017) and is associated with higher rates of drug side effects (Matson & Neal, 2009). O'Dwyer et al. (2017) examined polypharmacy in a group of older adults with IDD. Factors found to be influencing polypharmacy included living in a residential institution, having a history of reporting a mental health condition and sleep problems. Older individuals with IDD may be taking more medications than younger peers, and longitudinal studies find that more drugs seem to be added over time once pharmacotherapy is initiated (Esbensen et al., 2009; Valenza et al., 2017). Schoufour et al. (2018) looked at 1050 older adults with ID. Individuals taking 5 or more medications and who had 4 or more health conditions were 2.3–2.6 times more likely to die during a 5-year follow-up study period than peers with fewer of these risk factors. They concluded that efforts must be made to reduce the occurrence of polypharmacy and to prevent "multimorbidity." Lunsky and Modi (2017) found polypharmacy of psychotropic medications among outpatients with IDD was associated with female gender, residing in a supervised living situation and multiple psychiatric diagnoses. Spina and Leon (2017) suggested that clinicians need to familiarize themselves with the potential for adverse events and especially, the "potential for dangerous combinations of the increasingly frequent co-prescription of AEDs and psychotropic agents."

A large number of the 198 psychiatric inpatients in a study by Charlot et al. (2011) suffered from constipation and GERD. About 40% of admissions that were primarily due to challenging behaviors were eventually found to have a major contributing factor that was medical in nature. This included a number of problems that were associated with medications. In general, the inpatients had both high rates of medical problems and high rates of medication use, taking about three psychotropic drugs per person and a total of an average of five drugs per person for physical and mental ill health combined. Frequent goals during the admissions to the unit were to simplify complex medication regimens while treating medical problems and introducing non-drug interventions (Charlot, 2016).

Evidence Base

Treatment with antipsychotic drugs targeting challenging behavior in adults with ID is considered "controversial"… "because of inconclusive evidence for efficacy in the absence of a comorbid psychiatric condition, and substantial concerns about adverse effects" (Lunsky & Modi, 2017). A small number of studies contributed to risperidone and aripiprazole receiving FDA approval for treating "irritability" in youth with an ASD (Hirsch & Pringsheim, 2016; Research Units on Pediatric Psychopharmacology Autism Network, 2005). Irritability in these studies is defined as a score on the Aberrant Behavior Checklist Irritability subscale. The ABC is a factor-derived instrument normed for use with people with IDD, and it includes an array of items rated by caregivers. Many items on the Irritability subscale are related to externalizing behavior challenges, and very few items actually address mood states. The ABC has proven to be an excellent outcome measure for assessing the impact of interventions aimed at challenging behaviors. The majority of studies that supported the antipsychotic drug indication for treating "irritability" in children with ASD were short term, and the evidence for longer-term use of such treatment and use *not* occurring with other proven nonmedical assessments and interventions is questionable though common (Matson & Hess, 2011). There was one 6-month-long study of risperidone in which the risperidone was stopped over a very short taper and placebo started during the crossover component of the investigation. However, it should be noted that some individuals who have sudden or brief taper of antipsychotic medication display irritability initially which might account for the apparent relapse when individuals are taken off the study drug and placed on a placebo (see discussion below).

It remains unclear whether or not sedation contributes to a slowdown of challenging behaviors reported in the studies cited above. Sedation was reported to occur frequently in children in investigations regarding both aripiprazole (72% in the Marcus et al., 2011 study) and risperidone (51% rate in the Aman, De Smedt, Derivan, Lyons, and Findling (2002) study). There may be some constraints in terms of how generalizable these findings are as well. In many controlled drug studies, a very significant component of the target population was excluded for a variety of reasons, e.g., patients with seizure disorder who had any recent anti-epilepsy drug changes or a seizure any time in the past 6 months and any "significant" medical comorbidities. During the study, participants could not be taking any other psychotropic medications. About 1/3 or more of children with an ASD have a seizure disorder, and youth with ASD have high rates of medical comorbidities. In fact, a follow-up study of children in the RUPP (Research Units in Pediatric Psychopharmacology) investigations demonstrated that the youth who had constipation or diarrhea were less responsive to the treatment drugs (either risperidone or methylphenidate) and that about 1/3 of participants in these studies had these medical problems (CHECK and CITE). Both of the FDA-approved antipsychotic drugs were approved only for short-term use in children with an ASD. In a Cochrane review, the one long-term, placebo discontinuation study of aripiprazole found no differences between placebo and drug conditions in terms of relapse after discontinuation, "suggesting that re-evaluation of aripiprazole use after a period of stabilisation in irritability symptoms is warranted" (Hirsch & Pringsheim, 2016). It should be noted that placebo effects are generally reported to be very high in studies including children with IDD.

It has been well established that despite reports of reduced aggression in short-term use with risperidone, core features of ASD are not improved with this or any other similar medications. A recent investigation addressing this topic with a sample of 184 children with ASD who had reports of behavioral benefit concluded that core ASD symptoms "failed to improve, and in fact subtly worsened over time" (p.516) (Marrus, Underwood-Riordan, Randall, Zhang, & Constantino, 2014).

A number of review papers examine the efficacy of using medications to treat challenging behaviors or aggression in individuals with IDD with similar findings but sometimes different conclusions. The most comprehensive reviews can be found through the National Institute for Health and Care Excellence (NICE) in the United Kingdom and in the Cochrane Library of systematic reviews and meta-analyses. NICE reviews of research regarding antipsychotic drugs, antidepressants, and other psychotropic medications have concluded that the quality of evidence in support of these medications in treating challenging behaviors in people with IDD is low to very low or insufficient. In a study that reviewed the use of psychotropic medications in children with IDD, evidence for use was considered to be low with only short-term benefit for the use of risperidone, with cautious use being advised following best practice guidelines in terms of first exhausting less intrusive non-drug approaches (McQuire, Hassiotis, Harrison, & Pilling, 2015). However, as Matson and Williams (2015) warned, parents may be making decisions about what forms of intervention they choose for their children with ASD based on very different rationales than that applied by researchers, and a wide array of variables likely impact this.

An important study published in the Lancet reported on the clinical effectiveness of antipsychotic drugs in treating challenging behaviors in adults with IDD (Tyrer et al., 2009). The methodology used mirrored that of the CATIE studies (Caroff, Hurford, Lybrand, & Campbell, 2011), investigating "real-world" patients in treatment at multiple sites in Europe. The impact of haloperidol was contrasted with risperidone and placebo. Placebo was found to be as effective in the reduction of challenging behaviors as either of the antipsychotic drugs in the study.

Weighing Risks Versus Benefits

Even in research regarding the use of drugs such as antipsychotic medications for treating actual psychosis, data from recent investigations suggest that findings of efficacy are not particularly robust. There is debate as to whether long-term antipsychotic drug use increases mortality rates in general, but it has been determined that using these agents in elderly dementia patients does and significantly so (reported rates almost three times that of controls). Another worry is that in crossover designs, when there is a switch to placebo or even when a trial is started following an abrupt discontinuation of a current medication, withdrawal effects likely amplify the positive response attributed to the intervention drugs' effects (Beumer & Maes-Festen, 2017). Rather, getting the medication probably mitigates withdrawal effects if you recently stopped taking it, and suddenly stopping an antipsychotic and then getting a placebo would likely uncover withdrawal-related effects in those prone to these (not all people seem to experience such effects). See additional discussion regarding placebo effects below.

There are no randomized clinically controlled trials demonstrating efficacy of polypharmacy in the treatment of challenging behaviors in people with IDD, despite this being quite a common practice. The risk of side effects rises with the addition of medications. It is then especially difficult to see any rationale for this practice, especially when patients are not given access to other forms of intervention that carry much less risk such as ABA-related interventions. Studies in which a drug trial is conducted and there is a direct comparison with an established non-medication intervention seem completely absent from the literature. One investigation contrasted findings from studies reporting on effects of various interventions for challenging behaviors in individuals with IDD (Heyvaert, Maes, & Onghena, 2010). No one form of treatment was superior, including methodologically rigorous investigations of medications and behavioral and other therapeutic strategies. The authors suggested that if these various interventions are equally efficacious, consideration of associated risk should become the focus in treatment selection.

Another consideration in the risk benefit analyses of the use of antipsychotic and other psychotropic medications to treat challenging behaviors is that individuals with IDD rarely control their own pharmacotherapy (Kroese, Dewhurst, & Holmes, 2001). Individuals with IDD often struggle to report internal states, including not only feelings but also physical effects. For example, one of the authors worked with a patient with ASD who reported that he had swallowed batteries.

It was eventually shown that he suffered from GERD, but did not know how to explain it. This was true even though he had only a mild degree of ID. His associated increased challenging behaviors at the time were seen as "treatment resistant" when evident during treatment with atypical antipsychotic medications. At the same time, he was having problems related to both weight gain and daytime sedation.

In addition to challenging behaviors, changes in eating and motor behavior may also be provoked by a physical source of distress, including medication side effects (Charlot et al., 2011). Mistaking surface features of distress such as irritability and aggression as evidence of an acute mental illness episode or as a condition that requires medication in the absence of a comprehensive assessment may inappropriately elevate risk for adverse drug events (Charlot, 2016; Mathews et al., 2005). As an example, it seems clear that antipsychotic drugs prescribed to treat irritability (manifested as negative mood coupled with externalizing behaviors) that is provoked by constipation may reduce the associated externalizing behavior transiently because of sedative effects. However, everyone would agree that this approach is likely to be ineffective over time and potentially harmful, and it would be best to treat the constipation. One strong indicator that the longer-term use of antipsychotic drugs to treat aggression and irritable mood states is ineffective is how often children are started on drugs like risperidone but eventually end up on three or more medications for the same problems that the antipsychotic was prescribed (Esbensen et al., 2009). The drugs may only be effective until the patient has fully accommodated to sedative effects, since there is no evidence that core features of ASD or the symptoms of irritability are diminished because of specific drug actions of antipsychotic medications (Marrus et al., 2014).

There may be differences related to psychotropic medication prescription practices in different countries. Despite the likelihood that rates of challenges are similar, more children and adolescents in the United States are prescribed psychotropic medications than their counterparts in the United Kingdom (Murphy, McCarthy, Baer, Zima, & Jellinek, 2014). One interesting observation is that European, Canadian, and Australian reviews of psychotropic medication use in people with IDD for challenging behaviors tend to advise caution in their use for treating CBs (in line with NICE findings and guidelines), while US reports seem to suggest drugs are not only helpful but that some should be seen as a first-line intervention (Matson & Hess, 2011). One group from the United States, for example, based their recommendation that using two antipsychotic drugs together provides clinical benefit and is "generally well tolerated" on a retrospective chart review at one clinic including 61 individuals with ASD (Wink, Pedapati, Horn, McDougle, & Erickson, 2017). One comprehensive review from the United Kingdom, in contrast, concluded there was a limited evidence base, expressed concern regarding adverse effects of antipsychotic drugs used to treat behavioral challenges, and offered suggestions more in line with best practice guidelines such as those provided by NICE (Sheehan et al., 2015). The view presented was balanced, as the authors noted that there are some individuals who benefit from short-term use of the drugs under specific conditions. In an Australian review of the subject, it was also concluded that evidence for the use of psychotropic medications to treat challenging behavior is weak, with the authors suggesting that such use should be "avoided unless the behaviour is severe and non-responsive to other treatments" (Trollor, Salomon, & Franklin, 2016). Canadian guidelines for children with disruptive behavior disorders endorsed conditional use of risperidone for severe challenges only after psychosocial interventions failed and then only after medications for ADHD also failed. These authors warned consumers of this area of research to cautiously interpret results, due to industry sponsors influencing much of the research on the topic (Gorman et al., 2015).

In another US-based review, the authors suggest that lithium and risperidone "may have efficacy for use in management of aggression in patients with ID," immediately after concluding that evidence for psychotropic medications treating aggression in the population is limited

(Roy, Hoffman, Dudas, & Mendelowitz, 2013). LeClerc and Easley (2015) concluded that "risperidone and aripiprazole remain the mainstays of ASD treatment…" (p. 41). It is unclear if these varying conclusive suggestions impact practice differences, though it is evident that concerns about overuse are more evident in research and review papers coming out of the United Kingdom, the Netherlands, and Canada compared with the United States with few exceptions. Papers providing summary reviews of psychotropic medication use for aggressive behavior of individuals with ASD and other developmental disabilities tend to downplay side effects, despite the large numbers affected by them in studies of antipsychotic medication use. Placebo rates in many studies are significantly high, and studies with children with disabilities are likely exquisitely open to such effects for several reasons (Sandler, 2005). First, parents and other caregivers may be extremely motivated to find help related to family stress. Second, in addition to expectancy effects, ratings that are subjective (such as the ABC scoring) done by the caregivers who are hopeful that an intervention will work are very subject to bias. Newer research suggests that placebo in developmental disability studies is a significant issue and a powerful factor that impacts medication intervention and other investigations using RCT formats (Curie et al., 2015). This may mean that efficacy of a medication that reaches statistical significance might be clinically relatively small (a few points on subscale that could be due to nonspecific drug effects like slowing people or making them tired). These considerations in turn amplify the importance of weighing the associated risks of interventions carefully, as well as adding objective measures of outcomes (actual frequency counts versus rating scales).

Another issue with significance in weighing risks versus benefits of medication use for aggressive behavior relates to the patient's role in treatment. When a patient has IDD, decisions are often made by others (not the patient) to continue medications or add medications and whether any apparent side effects are "well tolerated." In fact, individuals who refuse their psychotropic medications may be characterized as "non-compliant," without considering whether or not medication refusal is actually a wise choice at the time. This should be a very significant concern for anyone treating a person with IDD with psychotropic medications for off-label indications, given the finding from the CATIE studies indicating that even people who have psychosis will quit taking antipsychotic drugs more often than not for a variety of reasons, including the perception that the benefit is not worth the negative effects (Caroff et al., 2011). It is unknown how often people with IDD have long-term exposure to medications that have caused them to feel uncomfortable or unwell, because of a difficulty in conveying this to caregivers who are making decisions on their behalf.

Rationale for Use of Psychotropic Medications to Treat Challenging Behaviors

The concerns described above beg the question, why are the prescription rates for psychotropic medications so prevalent in this population? As noted by Matson and Williams (2015), availability of research and best practice guidelines for treatments in ASD suggest medications have a very limited role, yet the use has continued to be very high. Bhaumik and Michael (2004) speculated as to why psychiatrists may be prescribing antipsychotic drugs for challenging behaviors at such high frequencies, including limited availability of resources, "lack of clinical psychology input, inability to change environment meaningfully, lack of suitably trained staff to manage private residential homes, pressure from nursing staff and other professionals for immediate resolution of problems, lack of meaningful employment or day care opportunities." Thalitaya et al. (2017) emphasized that many people with IDD can be helped to come off of what may be unnecessary psychopharmacologic therapies, but adding this can likely only happen if "there are the right clinical supports in place." The latter suggestion is supported by studies that examined drug withdrawal.

De Kuijper and Hoekstra (2017) examined physicians' decisions to discontinue antipsychotic medications. An unexpected result was that their rationales not only depended on characteristics of their patients such as the presence of emotional or behavioral challenges "but also on environmental factors, such as inappropriate living circumstances, and on attitudes, knowledge and beliefs of staff, clients and their representatives towards the effects of antipsychotic drug use...". They summarized that the primary reasons given for not trying to eliminate antipsychotic medications are related to concerns about restlessness, ASD diagnoses, prior failed attempts to discontinue the medication, and the objections to discontinuation voiced by legal representatives.

Matson and Hess (2011) pointed to the absence of equivalent attention to the use of well-established and less risky interventions for challenging behaviors compared with reports about drugs and argued that there are no data supporting the view that medications aimed at treating irritability (operationalized as the ABC subscale score) are an evidence-based treatment for challenging behavior. Medication side effects and other medical problems may be one of many possible irritants that are often missed because people with IDD may not reliably report their own symptoms (Charlot, 2016). In behavioral terms, these and other factors are setting events that operate to increase the probability of occurrence of a targeted challenging behavior when they are present. Akathisia, muscle stiffness, constipation, or acid reflux may exacerbate a negative response even to a small triggering event.

Discontinuing Antipsychotic Medications

There are a number of studies in recent years that have examined efforts to reduce antipsychotic medications. Sheehan and Hassiotis (2017) reviewed studies in which attempts were made to discontinue the use of antipsychotic medications when prescribed for challenging behaviors. A significant proportion of people who were treated with antipsychotics for externalizing behaviors had successful tapers or complete discontinuation, with subsequent behavioral functioning that was either better or the same. Improvements in cognitive and adaptive functioning have been reported as the result of removing an antipsychotic drug, as have improvements in metabolic syndrome parameters. In one small case series cited by Sheehan and Hassiotis, individuals diagnosed with dementia were no longer presenting with symptoms of the syndrome after antipsychotic drug discontinuation.

Withdrawal-emergent movement symptoms were reported in a few studies, appearing or worsening when antipsychotic drug doses were reduced. In one discontinuation study, the emergence of withdrawal-related movement side effects was a factor predictive of reinstatement of antipsychotic drug treatment (or a lack of successful discontinuation). In another investigation, these types of symptoms appeared to be diminished at 6 months of follow-up, and Sheehan and Hassiotis speculated that over time, some movement disorders may remit. An older study of individuals with severe IDD actually identified this as occurring for a subset of individuals successfully taken off of antipsychotic drugs (May et al., 1995). These authors concluded that "working conditions, staff experience, and attributions about challenging behaviour can influence how behaviour is reported and how it is managed" (p. 8). Caregivers may need support and education to be open to drug tapers when patients present with aggressive behavior, given that their anxiety seems to contribute to an inability to complete efforts to remove the medication. It seems critical to raise awareness and knowledge of side effects, particularly movement disorders, among those treated with antipsychotic medications. It should be noted, however, that movements have been reported having been provoked by other classes of medication in addition to antipsychotic medications, including antidepressants and anti-epilepsy drugs, medications that individuals with IDD are prescribed often with an antipsychotic drug (Mahan et al., 2010).

Rates of tapering were contrasted with trials including 14 and 28 week protocols. Kuijper, Evenhuis, Minderaa, and Hoekstra (2014) found no advantage of a longer tapering period. However, people who could not tolerate the reduction of the drug included a disproportionate number of individuals with withdrawal-emergent movement disorder symptoms. This would indicate that the drug still may not have been needed to manage challenging behaviors, but more simply, that it was difficult to eliminate it. This is important since psychiatrists report past failed attempts to eliminate an antipsychotic medication is a reason they will not try to do this again. Though the de Kuijper group concluded that 14 weeks was adequate for a taper, it might be that a much lengthier one (more than 28 weeks) would help individuals who were experiencing uncomfortable emergent movement symptoms. In a similar finding, the individuals in an open clinical trial conducted by Ahmed et al. (2000) who were unable to complete a taper of antipsychotic medications had higher scores on a measure of drug-related movement disorder symptoms. Many people in the Ahmed group's study were successful in completing an antipsychotic drug taper, demonstrating improved activity levels and increased engagement. Staff and environmental variables also seemed to have a significant impact on outcomes in this investigation. The McNamara group (McNamara et al., 2017), however, used a 6-month tapering protocol, and although this RCT study of antipsychotic drug discontinuation had a small sample of adults with IDD, a 100% success rate in tapering risperidone was achieved without any behavioral decompensation. This may be because a very conservative taper is useful for individuals with movement disorder side effects, something that has received little attention but has been observed by others (Charlot, 2016; Silka & Charlot, 2002). Hypothetically, a long tapering period would allow more time for downregulation of previously sensitized receptors affecting neuromotor circuits, if in fact a pathway to developing dyskinesias and other movement problems is through upregulation of D2 receptors over the course of prolonged blockade. "The hypersensitivity of D2 receptors may cause them to respond abnormally to the dopamine reaching them," when the blockade ceases (Mathews et al., 2005).

Psychotropic Medication Side-Effect Ascertainment

Given all of the above, the small number of investigations focused on the occurrences of medication side effects of people with IDD is worrisome (Matson, Fodstad, Neal, Dempsey, & Rivet, 2010). There are some reports, but these are limited in frequency and scope. On a positive note, a screening tool was developed for use with individuals with IDD, the Matson Evaluation of Drug Side Effects or the MEDS (Matson et al., 1998). Its psychometric properties are sound, and it is easy to administer. The screen consists of subscales that include items representing common side effects of psychotropic medications. Some of the research noted here included use of the tool.

Individuals with IDD taking psychotropic medications, and specifically taking antipsychotic medications, are at elevated risk for extrapyramidal symptoms or EPS including acute dystonias, akathisia, Parkinsonism, and tardive dyskinesia (TD). TD was diagnosed in over 1/3 of children with ASD receiving haloperidol in one investigation cited by Matson and Hess (2011), who also noted that cases of TD have been reported with risperidone use as well. In an early study of the risk factors for TD in a population of individuals with IDD, Matson's group identified age, psychiatric diagnosis, level of ID, recent psychotropic medication use, lifetime exposure and duration of use, current treatment with an antipsychotic, and total daily dose were found significant predictors (Matson et al., 2010). In another investigation, the difference between treatment with atypical and typical antipsychotic on the MEDS movement disorder subscales was significant only for akathisia (Advokat, Mayville, & Matson, 2000).

In a retrospective review of a very large cohort of individuals with ID treated with antipsychotic medications, Sheehan et al. (2017) found that the recorded incidence rate of any movement disorder was 30% higher in people with ID compared with those without ID. This important study is the first large-scale investigation to demonstrate that ID confers elevated risk for these ADEs, confirming what many clinicians have observed in practice. Parkinsonism and akathisia showed the most elevation in risk comparatively. Neuroleptic malignant syndrome (NMS), a rare but potentially lethal side effect of antipsychotic drugs, was documented as occurring three times more often in the individuals with ID versus without who were taking antipsychotic medications.

Use of antipsychotic medications has also been found to be associated with increased risk for an epilepsy event, physical injury, diabetes, depression, and respiratory problems in children with ASD and ID (Brophy et al., 2018). Ramerman, Hoekstra, and de Kuijper (2018) found a measure revealing reduced health and wellness was associated with antipsychotic drug use, with specific effects related to occurrences of dysphagia, temperature regulation problems, and Parkinsonian symptoms. Individuals with IDD may be at an increased risk of experiencing EPS (extra pyramidal symptoms) (Matson et al., 2010). Risk factors for ADEs or drug side effects have been identified for people with IDD. Mahan et al. (2010) (using the MEDS screening tool) found that receiving medications from more than one class was associated with having higher MEDS scores. Scheifes et al. (2016) found that of the 103 individuals with IDD for whom a survey was completed, over 80% had at least one ADE reported while 46% had 3 or more. They also found that being on more medications was correlated with having more side effects.

Valdovinos and colleagues found that having multiple drug changes in a short period of time seemed to elevate risk for adverse events (Valdovinos, Caruso, Roberts, Kim, & Kennedy, 2005). Charlot et al. (2011) reviewed the care of 198 psychiatric inpatient adults with IDD who were prescribed close to three psychotropic medications per person on average and found that having more medical problems (including drug-induced symptoms and disorders) was associated with longer lengths of stay. These findings provide added support to the Schoufour's group's warning that more attention should be paid to "multimorbidity" (Schoufour et al., 2018).

Data trends indicate the increasing use of the newer or second-generation antipsychotics, though there is continued use of first-generation antipsychotics in the population. Since their release into the market, numerous published reports have suggested that atypicals confer less risk of extra pyramidal symptoms than typicals. However, studies looking more closely at this over time have asserted that early investigations were usually too short term (most lasting from only 6–12 weeks), while many side effects of antipsychotics emerge over the course of long-term treatment (Matson et al., 2010). In earlier studies, the side effects of risperidone, for example, were described as "well tolerated" and included mainly sedation and weight gain (de Kuijper et al., 2013; Pringsheim, Lam, Ching, & Patten, 2011). de Kuijper and colleagues concluded that "more recent evaluation of the evidence suggests that the initial enthusiasm for second-generation agents was misplaced and largely based on studies that made unequal comparisons between atypicals and high-potency typical antipsychotics (de Kuijper et al., 2013). In the Sheehan et al.'s investigation cited above, group differences in rates of movement disorder side effects were not found based on whether the individual took a typical or atypical antipsychotic drug (Sheehan et al., 2017). Divac, Prostran, Jakovcevski, and Cerovac (2014) note that atypicals differ in terms of their relative risk for drug-induced movement disorders, but "the fact is that these drugs have not lived up to the expectation regarding their tolerability" (p. 1). Reports from studies in schizophrenia have suggested that comparator selection and dosing likely impacted earlier findings of lower EPS rates in patients treated with atypicals (Caroff et al., 2011).

Bakker, de Groot, van Os, and van Harten (2013) predicted the rise of drug-induced movement disorders secondary to the extensive marketing of atypicals as indicated in the treatment of multiple disorders, beyond schizophrenia, and other psychotic disorders. Even if it is accepted that those drugs as a group may be associated with a somewhat lower risk of movement disorders, the absolute numbers of people now likely to develop them is much larger because more and more people are being prescribed antipsychotics. Ryu et al. (2015) who performed detailed chart reviews of 80 elderly patients with schizophrenia on antipsychotic medications noted that tardive dystonias as well as tardive dyskinesia will affect most patients who receive long-term antipsychotic drug therapy. These findings amplify the need for those of us in the field of IDD to be vigilant in regard to the assessment of neuromotor symptoms associated with medication and to keep in mind that cognitive disability itself is a risk factor for drug-induced movement disorder (Peluso, Lewis, Barnes, & Jones, 2012; Sheehan et al., 2017). These investigators, however, noted the rate of reported TD was lower than expected and speculated that drug-induced tardive dyskinesia "may be misinterpreted as part of the underlying ID and under-recorded, an example of 'diagnostic overshadowing'."

Differential diagnosis of drug-induced movement disorders can be complex (Burkhard, 2014). This is especially the case with patients who have an IDD who may not complain of subtle but uncomfortable problems like muscle stiffness or "inner restlessness." Ryu's group (2015) cited above conveyed that they were "surprised by the paucity of notes in the patient files about movement disorder side effects," supporting the contention that such problems may be overlooked by clinicians without special training or awareness of the frequency and seriousness of their occurrence in people with IDD. At the same time, a balanced approach or a rational psychopharmacology will be required so that people who are helped by certain medications and where the risk benefit demonstrates a medication is a good choice receive the treatment they need in the context of a multidisciplinary approach (Sheehan et al., 2017).

Sedation, one of the most commonly reported side effects of antipsychotic medications, can at times slow the rate or reduce the intensity of aggressive acts leading staff persons to view it as effective. In some cases, however, people are both irritable and tired, which may lower the threshold for (or in ABA terms "occasion") increased occurrence of externalizing behaviors. It is possible that the sedative side effects of risperidone and aripiprazole, for example, contributed to the difference between placebo and drug in some investigations, given a rate as high as 72% of subjects given these drugs reportedly experienced this side effect (Marcus et al., 2011). The onset of the metabolic syndrome (increased weight, hyperlipidemia, and hyperglycemia) and many of the attendant complications of obesity have been well documented in people with IDD receiving atypical antipsychotics (de Kuijper et al., 2013).

Summary and Suggestions for the Future

Given the extent of the use of psychotropic medications in the treatment of individuals with IDD who have mental health needs or challenging behaviors, it seems critical that more efforts be made to expand our knowledge base about the nature and correlates of side effects these individuals may be experiencing. It may be especially helpful to explore the extent of side effects reported for individuals in crisis who are exposed to polypharmacy and who are continuing to present with the same symptoms and challenges for which the medications they have been receiving were prescribed. In these cases, the presence and impact of side effects may be missed. Basch (2010) noted that clinicians may miss side effects that are then identified by patient self-report, noting that clinicians will "systematically downgrade the severity of patients' symptoms." This is a major concern for people with IDD who may not report their symptoms accurately or at all. López-Sendón et al. (2013) cautioned that clinicians need to be aware of and attend to the possible presence of drug-induced Parkinsonism, "particularly in the elderly, patients taking multiple drugs and those with genetic risk factors involved in Parkinson's Disease." The warning was based on their observations that these conditions were being overlooked despite being of significant clinical concern for many individuals. Some individuals with genetically mediated syndromes and who are born with challenges such as cerebral palsy have elevated risk for abnormal movements and may be especially vulnerable to medications that might worsen existing conditions involving neuromotor circuits in the brain and related to diminished neural plasticity (Ravin, 2016).

The association of various types of physical discomfort to challenging behavior in people with IDD has long been recognized (Carr & Owen-DeSchryver, 2007; Gardner & Whalen, 1996). In many cases, however, the presence of externalizing behaviors does not lead to a search for possible medical irritants but rather seems to lead to adding medications, including offending agents. Individuals with IDD who have high levels of emotional and behavioral support needs, and who have a history of requiring emergency services, are probably at the most risk for having medication change and additions that can elevate the risk for ADEs. One study found that individuals with IDD were more likely than peers without IDD to be given psychotropic medications but less likely to be offered simple pain relievers such as NSAIDs when showing nonspecific signs of discomfort (Axmon, Sandberg, Ahlström, & Midlöv, 2017). This too may be secondary to the concern raised by Sheehan and colleagues with regard to diagnostic overshadowing. Diagnostic overshadowing, attributing the symptoms observed to the IDD or even to an existing psychiatric label and missing alternative causative factors for a clinical problem, may be especially important with regard to ADE detection in this population. Of interest was a study that explored quality of life, psychopathology, and medications in a group of adults with mild ID. Koch et al. (2015) found that "proxy" or staff reports of quality of life were impacted by the presence of behavioral and psychiatric symptoms, while taking more psychotropic medications correlated with lower reported quality of life based on self-report of the individuals themselves.

The research reviewed here has many limitations and is varied in design. It is very challenging to design studies to provide clear evidence of cause and effect when looking at things such as the use of psychotropic medication subtypes or polypharmacy. It has been difficult for researchers in some countries like the United States, to secure major funding to support studies of variables such as drug side effects, while the studies that do receive the most support are RCTs that examine intended drug effects as part of the harms/benefits from the intervention. Even with these investigations, few include adults with IDD. There does seem to be more activity in this area in other countries, especially in Canada, the Netherlands, and the United Kingdom.

Future investigations that further examine side effects and their relationship to medications would be helpful. Also, RCTs in which medications are evaluated head to head with behavioral or other non-drug interventions would be informative, though design challenges would be significant. Despite the drawbacks to real-world studies, these seem important in improving our understanding of medication effects on challenging behavior. Since best practice guidelines seem to be ineffective in changing actual prescribing, other initiatives will be needed if there is a will to change what clearly seems to be an over-reliance on psychotropic medications to control behavior in the population such as current efforts in the United Kingdom. Examination of policies in places like the United States would be necessary, since there is currently no national approach nor funded initiatives that insure people with IDD who are given drugs for behavior have access to multidisciplinary assessment and alternative therapies. Research reviewed here would suggest many do not.

Acknowledgment We would like to thank Miss Jessica Blickwedel for having contributed to the initial draft of the introduction.

References

Advokat, C. D., Mayville, E. A., & Matson, J. L. (2000). Side effect profiles of atypical antipsychotics, typical antipsychotics, or no psychotropic medications in persons with mental retardation. *Research in Developmental Disabilities, 21*(1), 75–84.

Ahmed, Z., Fraser, W., Kerr, M. P., Kiernan, C., Emerson, E., Robertson, J., … Thomas, J. (2000). Reducing antipsychotic medication in people with a learning disability. *The British Journal of Psychiatry, 176*(1), 42–46.

Ali, A., Blickwedel, J., & Hassiotis, A. (2014). Interventions for challenging behaviour in intellectual disability. *Advances in psychiatric Treatment, 20*(3), 184–192.

Aman, M. G., De Smedt, G., Derivan, A., Lyons, B., & Findling, R. L.. Risperidone Disruptive Behavior Study Group(2002). Double-blind, placebo-controlled study of risperidone for the treatment of disruptive behaviors in children with subaverage intelligence. *American Journal of Psychiatry, 159*(8), 1337–1346.

American Psychiatric Association. (2013). *Diagnostic and Statistical Manual of Mental Disorders (DSM-5®)*. American Psychiatric Pub.

Axmon, A., Sandberg, M., Ahlström, G., & Midlöv, P. (2017). Prescription of potentially inappropriate medications among older people with intellectual disability: A register study. *BMC Pharmacology and Toxicology, 18*(1), 68.

Bakker, P. R., de Groot, I. W., van Os, J., & van Harten, P. N. (2013). Predicting the incidence of antipsychotic-induced movement disorders in long-stay patients: A prospective study. *Epidemiology and Psychiatric Sciences, 22*(4), 375.

Banks, R., Bush, A., & Baker, P. (2007). *Challenging behaviour: A unified approach*. London: Royal College of Psychiatrists, British Psychological Society and Royal College of Speech and Language Therapists. CR 144. *Google Scholar*.

Basch, E. (2010). The missing voice of patients in drug-safety reporting. *New England Journal of Medicine, 362*(10), 865–869.

Beumer, S., & Maes-Festen, D. A. M. (2017). Reduction or discontinuation of antipsychotics for challenging behaviour in adults with intellectual disability: Why does it fail? *The Lancet Psychiatry, 4*(3), e2.

Bhaumik, S., & Michael, D. M. (2004). Pharmacotherapy and pharmacovigilance in learning disability. *Learning Disability Psychiatry, 6*(3), 9–10.

Bowring, D. L., Totsika, V., Hastings, R. P., Toogood, S., & McMahon, M. (2017). Prevalence of psychotropic medication use and association with challenging behaviour in adults with an intellectual disability. A total population study. *Journal of Intellectual Disability Research, 61*(6), 604–617.

Brophy, S., Kennedy, J., Fernandez-Gutierrez, F., John, A., Potter, R., Linehan, C., & Kerr, M. (2018). Characteristics of children prescribed antipsychotics: analysis of routinely collected data. *Journal of Child and Adolescent Psychopharmacology, 28*(3), 180–191.

Burkhard, P. R. (2014). Acute and subacute drug-induced movement disorders. *Parkinsonism & Related Disorders, 20*, S108–S112.

Caroff, S. N., Hurford, I., Lybrand, J., & Campbell, E. C. (2011). Movement disorders induced by antipsychotic drugs: Implications of the CATIE schizophrenia trial. *Neurologic Clinics, 29*(1), 127–148.

Carr, E. G., & Owen-DeSchryver, J. S. (2007). Physical illness, pain, and problem behavior in minimally verbal people with developmental disabilities. *Journal of Autism and Developmental Disorders, 37*(3), 413–424.

Charlot, L. R. (2016). Multidisciplinary assessment. In *Health care for people with intellectual and developmental disabilities across the lifespan* (pp. 1677–1698). Cham: Springer.

Charlot, L. R., Abend, S., Ravin, P., Mastis, K., Hunt, A., & Deutsch, C. (2011). Non-psychiatric health problems among psychiatric inpatients with intellectual disabilities. *Journal of Intellectual Disability Research, 55*(2), 199–209.

Charlot, L. R., Doucett, A. C., & Mezzacappa, E. (1993). Affective symptoms of institutionalized adults with mental retardation. *American Journal of Mental Retardation, 98*(3), 408–416.

Cooper, S. A., Smiley, E., Jackson, A., Finlayson, J., Allan, L., Mantry, D., & Morrison, J. (2009). Adults with intellectual disabilities: Prevalence, incidence and remission of aggressive behaviour and related factors. *Journal of Intellectual Disability Research, 53*(3), 217–232.

Crocker, A. G., Mercier, C., Lachapelle, Y., Brunet, A., Morin, D., & Roy, M. E. (2006). Prevalence and types of aggressive behaviour among adults with intellectual disabilities. *Journal of Intellectual Disability Research, 50*(9), 652–661.

Crocker, A. G., Prokić, A., Morin, D., & Reyes, A. (2014). Intellectual disability and co-occurring mental health and physical disorders in aggressive behaviour. *Journal of Intellectual Disability Research, 58*(11), 1032–1044.

Curie, A., Yang, K., Kirsch, I., Gollub, R. L., des Portes, V., Kaptchuk, T. J., & Jensen, K. B. (2015). Placebo responses in genetically determined intellectual disability: A meta-analysis. *PLoS One, 10*(7), e0133316.

de Kuijper, G. M., & Hoekstra, P. J. (2017). Physicians' reasons not to discontinue long-term used off-label antipsychotic drugs in people with intellectual disability. *Journal of Intellectual Disability Research, 61*(10), 899–908.

de Kuijper, G. M., Mulder, H., Evenhuis, H., Scholte, F., Visser, F., & Hoekstra, P. J. (2013). Determinants of physical health parameters in individuals with intellectual disability who use long-term antipsychotics. *Research in Developmental Disabilities, 34*(9), 2799–2809.

de Ruiter, K. P., Dekker, M. C., Verhulst, F. C., & Koot, H. M. (2007). Developmental course of psychopathology in youths with and without intellectual disabilities. *Journal of Child Psychology and Psychiatry, 48*(5), 498–507.

Divac, N., Prostran, M., Jakovcevski, I., & Cerovac, N. (2014). Second-generation antipsychotics and extrapyramidal adverse effects. *BioMed Research International, 2014*.

Emerson, E., Kiernan, C., Alborz, A., Reeves, D., Mason, H., Swarbrick, R., … Hatton, C. (2001). The prevalence of challenging behaviors: A total population study. *Research in Developmental Disabilities, 22*(1), 77–93.

England N. H. S. (2015). Winterbourne medicines programme: Improving the use of medicines in people with learning disabilities–NHS Improving Quality Report Apr 2014–Apr 2015.

Esbensen, A. J., Greenberg, J. S., Seltzer, M. M., & Aman, M. G. (2009). A longitudinal investigation of psychotropic and non-psychotropic medication use among adolescents and adults with autism spectrum disorders. *Journal of Autism and Developmental Disorders, 39*(9), 1339–1349.

Fitzpatrick, S. E., Srivorakiat, L., Wink, L. K., Pedapati, E. V., & Erickson, C. A. (2016). Aggression in autism spectrum disorder: Presentation and treatment options. *Neuropsychiatric Disease and Treatment, 12*, 1525.

Gardner, W. I., & Whalen, J. P. (1996). A multimodal behavior analytic model for evaluating the effects of medical problems on nonspecific behavioral symptoms in persons with developmental disabilities. *Behavioral Interventions: Theory & Practice in Residential & Community-Based Clinical Programs, 11*(3), 147–161.

Gorman, D. A., Gardner, D. M., Murphy, A. L., Feldman, M., Bélanger, S. A., Steele, M. M., … Ustina, J. (2015). Canadian guidelines on pharmacotherapy for disruptive and aggressive behaviour in children and adolescents with attention-deficit hyperactivity disorder, oppositional defiant disorder, or conduct disorder. *The Canadian Journal of Psychiatry, 60*(2), 62–76.

Granic, I. (2014). The role of anxiety in the development, maintenance, and treatment of childhood aggression. *Development and Psychopathology, 26*(4pt2), 1515–1530.

Häβler, F., Thome, J., & Reis, O. (2015). Polypharmacy in the treatment of subjects with intellectual disability. *Journal of Neural Transmission, 122*(1), 93–100.

Heyvaert, M., Maes, B., & Onghena, P. (2010). A meta-analysis of intervention effects on challenging behaviour among persons with intellectual disabilities. *Journal of Intellectual Disability Research, 54*(7), 634–649.

Hill, A. P., Zuckerman, K. E., Hagen, A. D., Kriz, D. J., Duvall, S. W., Van Santen, J., … Fombonne, E. (2014). Aggressive behavior problems in children with autism spectrum disorders: prevalence and correlates in a large clinical sample. *Research in Autism Spectrum Disorders, 8*(9), 1121–1133.

Hirsch, L. E., & Pringsheim, T. (2016). *Aripiprazole for autism spectrum disorders (ASD)*. The Cochrane Library.

Houghton, R., Ong, R. C., & Bolognani, F. (2017). Psychiatric comorbidities and use of psychotropic medications in people with autism spectrum disorder in the United States. *Autism Research, 10*(12), 2037–2047.

Hunter, R. H., Wilkniss, S., Gardner, W. I., & Silverstein, S. M. (2008). The Multimodal Functional Model--Advancing case formulation beyond the "diagnose and treat" paradigm: Improving outcomes and reducing aggression and the use of control procedures in psychiatric care. *Psychological Services, 5*(1), 11.

Kennedy, C. H., Juárez, A. P., Becker, A., Greenslade, K., Harvey, M. T., Sullivan, C., & Tally, B. (2007). Children with severe developmental disabilities and behavioral disorders have increased special healthcare needs. *Developmental Medicine & Child Neurology, 49*(12), 926–930.

Koch, A. D., Vogel, A., Becker, T., Salize, H. J., Voss, E., Werner, A., … Schützwohl, M. (2015). Proxy and self-reported Quality of Life in adults with intellectual disabilities: Impact of psychiatric symptoms, problem behaviour, psychotropic medication and unmet needs. *Research in Developmental Disabilities, 45*, 136–146.

Kroese, B., Dewhurst, D., & Holmes, G. (2001). Diagnosis and drugs: help or hindrance when people with learning disabilities have psychological problems? *British Journal of Learning Disabilities, 29*(1), 26–33.

de Kuijper, G. M., Evenhuis, H. Minderaa, R. B., & Hoekstra, P. J. (2014). Effects of controlled discontinuation of long-term used antipsychotics for behavioural symptoms in individuals with intellectual disability. Journal of Intellectual Disability Research*,* 58(1)*,* 71–83.

Lake, J. K., Denton, D., Lunsky, Y., Shui, A. M., Veenstra-VanderWeele, J., & Anagnostou, E. (2017). Medical conditions and demographic, service and clinical factors associated with atypical antipsychotic medication use among children with an autism spectrum disorder. *Journal of Autism and Developmental Disorders, 47*(5), 1391–1402.

LeClerc, S., & Easley, D. (2015). Pharmacological therapies for autism spectrum disorder: A review. *Pharmacy and Therapeutics, 40*(6), 389.

López-Sendón, J., Mena, M. A., & de Yébenes, J. G. (2013). Drug-induced parkinsonism. *Expert Opinion on Drug Safety, 12*(4), 487–496.

Lott, I. T., McGregor, M., Engelman, L., Touchette, P., Tournay, A., Sandman, C., … Walsh, D. (2004). Longitudinal prescribing patterns for psychoactive medications in community-based individuals with developmental disabilities: Utilization of pharmacy records. *Journal of Intellectual Disability Research, 48*(6), 563–571.

Lowry, M. A., & Sovner, R. (1992). Severe behaviour problems associated with rapid cycling bipolar disorder in two adults with profound mental retardation. *Journal of Intellectual Disability Research, 36*(3), 269–281.

Lunsky, Y., & Modi, M. (2017). Predictors of psychotropic polypharmacy among outpatients with psychiatric disorders and intellectual disability. *Psychiatric Services*. appi-ps.

Mahan, S., Holloway, J., Bamburg, J. W., Hess, J. A., Fodstad, J. C., & Matson, J. L. (2010). An examination of psychotropic medication side effects: Does taking a greater number of psychotropic medications from different classes affect presentation of side effects in adults with ID? *Research in Developmental Disabilities, 31*(6), 1561–1569.

Maher, A. R., Maglione, M., Bagley, S., Suttorp, M., Hu, J. H., Ewing, B., … Shekelle, P. G. (2011). Efficacy and comparative effectiveness of atypical antipsychotic medications for off-label uses in adults: A systematic review and meta-analysis. *JAMA, 306*(12), 1359–1369.

Mandell, D. S., Morales, K. H., Marcus, S. C., Stahmer, A. C., Doshi, J., & Polsky, D. E. (2008). Psychotropic medication use among Medicaid-enrolled children with autism spectrum disorders. *Pediatrics, 121*(3), e441–e448.

Marcus, R. N., Owen, R., Manos, G., Mankoski, R., Kamen, L., McQuade, R. D., … Aman, M. G. (2011). Aripiprazole in the treatment of irritability in pediatric patients (aged 6–17 years) with autistic disorder: Results from a 52-week, open-label study. *Journal of Child and Adolescent Psychopharmacology, 21*(3), 229–236.

Marrus, N., Underwood-Riordan, H., Randall, F., Zhang, Y., & Constantino, J. N. (2014). Lack of effect of risperidone on core autistic symptoms: Data from a longitudinal study. *Journal of Child and Adolescent Psychopharmacology, 24*(9), 513–518.

Mathews, M., Gratz, S., Adetunji, B., George, V., Mathews, M., & Basil, B. (2005). Antipsychotic-induced movement disorders: Evaluation and treatment. *Psychiatry (Edgmont), 2*(3), 36.

Matson, J. L., Fodstad, J. C., Neal, D., Dempsey, T., & Rivet, T. T. (2010). Risk factors for tardive dyskinesia in adults with intellectual disability, comorbid psychopathology, and long-term psychotropic use. *Research in Developmental Disabilities, 31*(1), 108–116.

Matson, J. L., & Hess, J. A. (2011). Psychotropic drug efficacy and side effects for persons with autism spectrum disorders. *Research in Autism Spectrum Disorders, 5*(1), 230–236.

Matson, J. L., & Konst, M. J. (2015). Why pharmacotherapy is overused among persons with autism spectrum disorders. *Research in Autism Spectrum Disorders, 9*, 34–37.

Matson, J. L., & LoVullo, S. V. (2008). A review of behavioral treatments for self-injurious behaviors of persons with autism spectrum disorders. *Behavior Modification, 32*(1), 61–76.

Matson, J. L., Mayville, E. A., Bielecki, J., Barnes, W. H., Bamburg, J. W., & Baglio, C. S. (1998). Reliability of the Matson evaluation of drug side effects scale (MEDS). *Research in Developmental Disabilities, 19*(6), 501–506.

Matson, J. L., & Neal, D. (2009). Psychotropic medication use for challenging behaviors in persons with intellectual disabilities: An overview. *Research in Developmental Disabilities, 30*(3), 572–586.

Matson, J. L., & Williams, L. W. (2015). The curious selection process of treatments for autism spectrum disorders. *Research in Autism Spectrum Disorders, 9*, 21–25.

May, P., London, E. B., Zimmerman, T., Thompson, R., Mento, T., & Spreat, S. (1995). A study of the clinical outcome of patients with profound mental retardation gradually withdrawn from chronic neuroleptic medication. *Annals of Clinical Psychiatry, 7*(4), 155–160.

Mazurek, M. O., Kanne, S. M., & Wodka, E. L. (2013). Physical aggression in children and adolescents with autism spectrum disorders. *Research in Autism Spectrum Disorders, 7*(3), 455–465.

McClintock, K., Hall, S., & Oliver, C. (2003). Risk markers associated with challenging behaviours in people with intellectual disabilities: A meta-analytic study. *Journal of Intellectual Disability Research, 47*(6), 405–416.

McNamara, R., Randell, E., Gillespie, D., Wood, F., Felce, D., Romeo, R., … Meek, A. (2017). A pilot randomised controlled trial of community-led antipsychotic drug reduction for adults with learning disabilities. *Health Technology Assessment, 21*(47), 1–92.

McQuire, C., Hassiotis, A., Harrison, B., & Pilling, S. (2015). Pharmacological interventions for challenging behaviour in children with intellectual disabilities: A systematic review and meta-analysis. *BMC Psychiatry, 15*(1), 303.

Melville, C. A., Johnson, P. C., Smiley, E., Simpson, N., Purves, D., McConnachie, A., & Cooper, S. A. (2016). Problem behaviours and symptom dimensions of psychiatric disorders in adults with intellectual disabilities: An exploratory and confirmatory factor analysis. *Research in Developmental Disabilities, 55*, 1–13.

Murphy, J. M., McCarthy, A. E., Baer, L., Zima, B. T., & Jellinek, M. S. (2014). Alternative national guidelines for treating attention and depression problems in children: Comparison of treatment approaches and prescribing rates in the United Kingdom and United States. *Harvard Review of Psychiatry, 22*(3), 179–192.

O'dwyer, M., Peklar, J., Mulryan, N., McCallion, P., McCarron, M., & Henman, M. C. (2017). Prevalence, patterns and factors associated with psychotropic use in older adults with intellectual disabilities in Ireland. *Journal of Intellectual Disability Research, 61*(10), 969–983.

Oliver, C., Sloneem, J., Hall, S., & Arron, K. (2009). Self-injurious behaviour in Cornelia de Lange syndrome: 1. Prevalence and phenomenology. *Journal of Intellectual Disability Research, 53*(7), 575–589.

Painter, J., Ingham, B., Trevithick, L., Hastings, R. P., & Roy, A. (2018). Correlates for the risk of specialist ID hospital admission for people with intellectual disabilities: Development of the LDNAT inpatient index. *Tizard Learning Disability Review, 23*(1), 42–50.

Park, S. Y., Cervesi, C., Galling, B., Molteni, S., Walyzada, F., Ameis, S. H., … Correll, C. U. (2016). Antipsychotic use trends in youth with autism spectrum disorder and/or intellectual disability: A meta-analysis. *Journal of the American Academy of Child & Adolescent Psychiatry, 55*(6), 456–468.

Peluso, M. J., Lewis, S. W., Barnes, T. R., & Jones, P. B. (2012). Extrapyramidal motor side-effects of first-and second-generation antipsychotic drugs. *The British Journal of Psychiatry, 200*(5), 387–392.

Poppes, P., Van der Putten, A. J. J., & Vlaskamp, C. (2010). Frequency and severity of challenging behaviour in people with profound intellectual and multiple disabilities. *Research in Developmental Disabilities, 31*(6), 1269–1275.

Powis, L., & Oliver, C. (2014). The prevalence of aggression in genetic syndromes: A review. *Research in Developmental Disabilities, 35*(5), 1051–1071.

Pringsheim, T., Lam, D., Ching, H., & Patten, S. (2011). Metabolic and neurological complications of second-generation antipsychotic use in children. *Drug Safety, 34*(8), 651–668.

Ramerman, L., Hoekstra, P. J., & de Kuijper, G. (2018). Health-related quality of life in people with intellectual disability who use long-term antipsychotic drugs for challenging behaviour. *Research in Developmental Disabilities, 75*, 49–58.

Ravin, P. D. (2016). Movement disorders. In *Health care for people with intellectual and developmental disabilities across the lifespan* (pp. 985–994). Cham: Springer.

Research Units on Pediatric Psychopharmacology Autism Network. (2005). Risperidone treatment of autistic disorder: longer-term benefits and blinded discontinuation after 6 months. *American Journal of Psychiatry, 162*(7), 1361–1369.

Roy, D., Hoffman, P., Dudas, M., & Mendelowitz, A. (2013). Pharmacologic management of aggression in adults with intellectual disability. *Journal of Intellectual Disability-Diagnosis and Treatment, 1*(1), 28–43.

Rutter, M., & Sroufe, L. A. (2000). Developmental psychopathology: Concepts and challenges. *Development and Psychopathology, 12*(3), 265–296.

Ryu, S., Yoo, J. H., Kim, J. H., Choi, J. S., Baek, J. H., Ha, K., … Hong, K. S. (2015). Tardive dyskinesia and tardive dystonia with second-generation antipsychotics in non-elderly schizophrenic patients unexposed to first-generation antipsychotics: A cross-sectional and retrospective study. *Journal of Clinical Psychopharmacology, 35*(1), 13–21.

Sandler, A. (2005). Placebo effects in developmental disabilities: implications for research and practice. *Developmental Disabilities Research Reviews, 11*(2), 164–170.

Scheifes, A., Walraven, S., Stolker, J. J., Nijman, H. L., Egberts, T. C., & Heerdink, E. R. (2016). Adverse events and the relation with quality of life in adults with intellectual disability and challenging behaviour using psychotropic drugs. *Research in Developmental Disabilities, 49*, 13–21.

Schoufour, J. D., Oppewal, A., van der Maarl, H. J., Hermans, H., Evenhuis, H. M., Hilgenkamp, T. I., & Festen, D. A. (2018). Multimorbidity and polypharmacy are independently associated with mortality in older people with intellectual disabilities: A 5-year follow-up from the HA-ID study. *American Journal on Intellectual and Developmental Disabilities, 123*(1), 72–82.

Sheehan, R., & Hassiotis, A. (2017). Reduction or discontinuation of antipsychotics for challenging behaviour in adults with intellectual disability: A systematic review. *The Lancet Psychiatry, 4*(3), 238–256.

Sheehan, R., Hassiotis, A., Walters, K., Osborn, D., Strydom, A., & Horsfall, L. (2015). Mental illness, challenging behaviour, and psychotropic drug prescribing in people with intellectual disability: UK population based cohort study. *British Medical Journal, 351*, h4326.

Sheehan, R., Horsfall, L., Strydom, A., Osborn, D., Walters, K., & Hassiotis, A. (2017). Movement side effects of antipsychotic drugs in adults with and without intellectual disability: UK population-based cohort study. *British Medical Journal, 7*(8), e017406.

Silka, V. R., & Charlot, L. (2002, October). Neuroleptic withdrawal syndrome and cardiac risk of neuroleptics. In *Proceedings of the 19th Annual Conference, National Association for the Dually Diagnosed,* Denver, CO

Spina, E., & Leon, J. D. (2017). Potentially clinically relevant pharmacodynamic interactions between antiepileptic drugs and psychotropic drugs: An update. *Current Pharmaceutical Design, 23*(37), 5625–5638.

Sturmey, P., Laud, R. B., Cooper, C. L., Matson, J. L., & Fodstad, J. C. (2010). Challenging behaviors should not be considered depressive equivalents in individuals with intellectual disabilities. II. A replication study. *Research in Developmental Disabilities, 31*(5), 1002–1007.

Thalitaya, M. D., Reynolds, C., & Ismail, I. (2017). Study on the use of antipsychotic medication in an intellectual disability (ID) community caseload. *Psychiatria Danubina, 29*.(Suppl 3, 578–583.

Totsika, V., & Hastings, R. P. (2009). Persistent challenging behaviour in people with an intellectual disability. *Current Opinion in Psychiatry, 22*(5), 437–441.

Totsika, V., Toogood, S., Hastings, R. P., & Lewis, S. (2008). Persistence of challenging behaviours in adults with intellectual disability over a period of 11 years. *Journal of Intellectual Disability Research, 52*(5), 446–457.

Trollor, J. N., Salomon, C., & Franklin, C. (2016). Prescribing psychotropic drugs to adults with an intellectual disability. *Australian Prescriber, 39*(4), 126.

Tsakanikos, E., Costello, H., Holt, G., Sturmey, P., & Bouras, N. (2007). Behaviour management problems as predictors of psychotropic medication and use of psychiatric services in adults with autism. *Journal of Autism and Developmental Disorders, 37*(6), 1080–1085.

Tyrer, P., Oliver-Africano, P., Romeo, R., Knapp, M., Dickens, S., Bouras, N., … Hare, M. (2009). Neuroleptics in the treatment of aggressive challenging behaviour for people with intellectual disabilities: A randomised controlled trial (NACHBID). *Health Technology Assessment, 13*(21), i-73.

Unwin, G. L. (2014). *A longitudinal observational study of aggressive behaviour in adults with intellectual disabilities* (Doctoral dissertation, University of Birmingham).

Valdovinos, M. G., Caruso, M., Roberts, C., Kim, G., & Kennedy, C. H. (2005). Medical and behavioral symptoms as potential medication side effects in adults with developmental disabilities. *American Journal on Mental Retardation, 110*(3), 164–170.

Valenza, P. L., McGinley, T. C., Feldman, J., Patel, P., Cornejo, K., Liang, N., … McNaughton, N. (2017) Dangers of polypharmacy. In *Vignettes in patient safety* (Vol. 1). INTECH.

Visser, E. M., Berger, H. J. C., Prins, J. B., Lantman-De Valk, H. V. S., & Teunisse, J. P. (2014). Shifting impairment and aggression in intellectual disability and autism spectrum disorder. *Research in Developmental Disabilities, 35*(9), 2137–2147.

Wink, L. K., Pedapati, E. V., Horn, P. S., McDougle, C. J., & Erickson, C. A. (2017). Multiple antipsychotic medication use in autism spectrum disorder. *Journal of Child and Adolescent Psychopharmacology, 27*(1), 91–94.

World Health Organization. (1993). The ICD-10 Classification of mental and behavioural disorders: Diagnostic criteria for research (Vol. 2). World Health Organization.

Part IX

Skill Building and Intellectual Disabilities

Social Skills

37

Steven G. Little, Angeleque Akin-Little,
Margaret Gopaul, and Tom Nicholson

Libet and Lewinsohn (1973, p. 311) defined social skills as the "complex ability to maximize the rate of positive reinforcement and to minimize the strength of punishment elicited from others." Such a definition, while fundamentally accurate, is rather broad and does little to increase our understanding of the specific behaviors that are understood as comprising social skills. While current social skills definitions may vary somewhat, there is some basic agreement as to how social skills are developed. Specifically, social skills involve specific learned behaviors, are comprised of both initiation and response behaviors, and entail interactions with others. These skills are also socially reinforced and denote skills that are context specific (Little, Swangler, & Akin-Little, 2017). Basically, social skills are those skills that enable individuals to function competently at social tasks (Cook, Gresham, Barreras, Thornton, & Crews, 2008).

Gresham (1986, 1997) categorized social skills into three definitional areas: peer acceptance, behavioral, and social validity. Peer acceptance defines social skills by how readily an individual is accepted by his or her peers. In other words, it refers to an individual's popularity among peers (Oden & Asher, 1977). Such a definition defines social skills by their outcome and fails to identify the specific behaviors that lead to peer acceptance. In addition, while useful in a context such as a regular school classroom, it is less useful when applied to the ID population. The behavioral definition focuses on situation-specific behaviors that maximize the probability of reinforcement while decreasing the probability of punishment (Libet & Lewinsohn, 1973). This definition has greater relevance to ID as it allows social behavior to be specified and operationalized for measurement and intervention. It does not, however, ensure that the identified social behaviors are socially significant or socially relevant. The social validity definition defines social skills as exhibiting behaviors that predict important social outcomes in particular situations (Gresham, 1983; Kazdin, 1977; Wolf, 1978).

Related to social skills is the term social competence. While social skills consist of specific skills that form the basis for socially competent behavior, social competence reflects social judgment about the quality of an individual's performance (Hops, 1983). Social skills are considered to be the "most malleable of the components of social competence" (Elliott & Busse, 1991, p. 64). Semrud-Clikeman (2007) described social

---

S. G. Little (✉)
Walden University, Minneapolis, MN, USA

A. Akin-Little
Akin-Little & Little Behavioral Psychology Consultants, Malone, NY, USA

M. Gopaul
Liberty University, Lynchburg, VA, USA

T. Nicholson
Massey University, Auckland, New Zealand

J. L. Matson (ed.), *Handbook of Intellectual Disabilities*, Autism and Child Psychopathology Series, https://doi.org/10.1007/978-3-030-20843-1_37

competence as consisting of social, emotional, cognitive, and behavioral components needed for successful social adaptation. Social skills can be thought of as the behavioral component of Semrud-Clikeman's definition. Greenspan (1981) provided an interesting analogy that helps clarify the distinction between social skills and social competence. Competence as a golfer is usually defined by the outcome, the final score. This is analogous to social competence. The golf score, however, tells us nothing about the skills (e.g., driving, approach shots, putting) and how these contributed to the final score. These are analogous to social skills. As this chapter focuses on the ID population, attention will deal primarily with social skills and interventions directed at improving specific social behaviors.

Gresham (1986) categorizes social skills difficulty into four types: skills deficits, performance deficits, self-control skills deficits, and self-control performance deficits. Skills deficits are manifested when the individual lacks the skills needed for appropriate social interaction. Performance deficits are those where the individual possesses the capability of performing the behavior but is not performing the behaviors at an acceptable level due to factors such as motivation or lack of opportunity. Self-control skills deficits are identified when an individual has not learned a social skill. For example, in individuals with ID, their cognitive deficits may interfere with learning the social skill. Finally, self-control performance deficits also involve interference, but in this case it has not interfered with the acquisition of the behavior but rather with its performance. It is important to identify the type of social skill deficit when developing an intervention.

Constantino, Przybeck, Friesen, and Todd (2000) defined social skills as involving five components: social awareness, social cognition, social communication, social motivation, and autistic mannerisms. Social awareness involves the ability to recognize social cues. Social cognition is the ability to interpret social cues once they are identified. Social communication includes behaviors such as expressive social communication. Social motivation incorporates factors such as social anxiety, inhibition, and empathic orientation that influence the individual's motivation to respond in a socially responsive manner. Finally, autistic mannerisms include aspects of ASD and ID (e.g., stereotyped behaviors, highly restricted interests) that, while idiosyncratic, need to be addressed in any intervention designed to improve social functioning.

Social Skills and Intellectual Disability

The American Association on Intellectual and Developmental Disabilities (AAIDD) definition of ID states that "Intellectual disability is characterized by significant limitations both in intellectual functioning and in adaptive behavior as expressed in conceptual, social, and practical adaptive skills. This disability originates before age 18" (Schalock et al., 2010, p. 5). The DSM-5 (APA, 2013) definition includes three criteria: (a) deficits in intellectual functioning, (b) deficits in adaptive functioning that result in failure to meet developmental and sociocultural standards for personal independence and social responsibility, and (c) onset during the developmental period. The ICD-10 continues to use the term mental retardation and defines it as "A condition of arrested or incomplete development of the mind, which is especially characterized by impairment of skills manifested during the developmental period, skills which contribute to the overall level of intelligence, i.e. cognitive, language, motor, and social abilities" (World Health Organization, 2010). Adaptive behavior, which includes social skills and social functioning, is a fundamental component of each. Parmenter (2011) emphasized that social competence is the key factor for ID students making a successful transition into adult living, including successful employment. Without a doubt, individuals with ID have significant deficits in social skills, social skills are critical for effectively interacting with others, and the failure to acquire appropriate social skills can result in a multitude of other deficits (Matson, Dempsey, & LoVullo, 2009). The importance of social skills therefore cannot be underestimated.

Research has consistently supported the contention that social skills delays are a central component of ID. For example, de Bildt et al. (2005) examined social skills in 510 children with mild and moderate ID and found that all children in this study were delayed in their basic social skills, including communication skills, and the lower the level of ID, the larger the delay. De Bildt and colleagues, however, believe it is important to discriminate between limitations in social skills related to ID as opposed to social skills limitations in autism spectrum disorder (ASD) or other disorders with social skills deficits. Specifically in ID, the level of social skills functioning is usually commensurate with other areas of development and functioning with a relatively homogeneous profile (i.e., few relative strengths or weaknesses). In ASD, and with other disabilities with characteristic social skills deficits (e.g., ASD, attention–deficit/hyperactivity disorder, social anxiety disorder), limitations in social skills are often greater than in other areas of function, and there is greater heterogeneity in areas of social skills functioning.

Deficits in social functioning have been an essential component of ID/MR definitions as early as Doll (1941) where he defined ID as including "a state of social incompetence" (p. 215) and Greenspan (1999) has argued that social intelligence is the most important factor in ID. Greenspan makes it clear, however, that MR is not a global disorder of social competence, but involves significant deficits in social intelligence. He further elaborates that adaptive behavior instruments measure more "practical intelligence" (e.g., daily living skills) than social skills which are a component of social intelligence.

Specific Social Skills Deficits. It can be said that social skills involve specific learned behaviors, are comprised of both initiation and response behaviors, and entail interactions with others (Little et al., 2017). The Social Skills Scale of the *Social Skills Improvement System Rating Scales* (*SSIS*, Gresham & Elliott, 2008) provides a good framework for understanding the behaviors that comprise social skills. Social skills, as defined by the Social Skills Scale, consist of seven domains: communication, cooperation, assertion, responsibility, empathy, engagement, and self-control. Communication behaviors include making eye contact when talking and saying "please" and "thank you." Cooperation involves behaviors such as rule following and task completion. Assertion consists of behaviors such as asking for help and indicating when there is a problem. Examples of responsibility behaviors include respecting the property of others and acting in a well-behaved manner when unsupervised. Empathy involves behaviors such as trying to comfort others and feeling bad when others are sad. Engagement includes inviting others to join in activities, making friends easily, and introducing oneself to others. Finally, examples of self-control behaviors include staying calm when teased and acting appropriately when upset.

Communication. Communication skills are a frequent deficit in individuals with ID (Kurtz, Boetler, Jarmolowicz, Chin, & Hagopian, 2011). This is especially true for individuals at the lower end of the distribution (Matson, Terlonge, González, & Rivet, 2006). There is, however, a wide range of variability in communication skills in people with ID at all levels (van der Meer, Sigafoos, O'Reilly, & Lancioni, 2011). Light (1989) offered a definition of communicative competence as "… a relative and dynamic, interpersonal construct based on functionality of communication, adequacy of communication, and sufficiency of knowledge, judgment and skill in four interrelated domains: linguistic competence, operational competence, social competence, and strategic competence" (p. 137). The International Classification of Functioning, Disability and Health (ICF) divides communication into three areas: receiving communication (receptive communication), producing communication (expressive communication), and conversation (Schindler, Ruoppolo, & Barillari, 2010). This is very similar to the common communication subdomains of expressive, receptive, and written communication (Belva, Matson, Sipes, & Bamburg 2012) with written communication including expressive and receptive communication specific to reading and writing.

Cooperation. Cooperative behavior has been defined as involving both collaboration and prosocial behavior. Collaboration is usually defined as two or more individuals working together to achieve benefits and prosocial behavior defined as any behavior that benefits another, regardless of the benefit to the individual (Cronin, 2017). Lane, Givner, and Pierson (2004) surveyed elementary teachers utilizing items on the Social Skills Rating System and found that they viewed cooperation and self-control skills as the most important variables for success in school. Meier, DiPerna, and Oster (2006) confirmed Lane et al.'s research and found that specifically ignores peer distractions, follows directions, attends to instructions, uses free time acceptably, and uses time appropriately were all cooperation items identified critical for classroom success by a majority of elementary school teachers.

Assertion. This dimension is dominated by social skills that reflect a child or youth who might be called "outgoing or extroverted" by others. Skills such as initiating conversations with others, acknowledging compliments, and inviting others to interact are examples of assertion-based social skills (Caldarella & Merrell, 1997).

Responsibility. In the English language, "responsibility" can have multiple definitions including (a) the state or fact of having a duty to deal with something or of having control over someone, (b) the state or fact of being accountable or to blame for something, and (c) the opportunity or ability to act independently and take decisions without authorization. Operationalized on rating scales, responsibility includes behaviors such as respecting the property of others and being well-behaved when unsupervised.

Empathy. Empathy has been defined as sharing the emotional state or context of another (Eisenberg & Strayer, 1987). It has not, however, been used or measured in a consistent way, leading to confusion in exactly what it means (Cuff, Brown, Taylor, & Howat, 2016; Ralfs & Beail, 2012). In people with ID, the evidence on empathy is also equivocal with studies inconsistent about whether individuals with ID differ from individuals with higher levels of cognitive functioning on emotional and behavioral aspects of empathy (Kasari, Freeman, & Bass, 2003).

Engagement. In the context of social skills, engagement is usually defined as the time a child spends interacting with his/her environment in a developmentally and contextually appropriate manner (Nelson, Paul, Johnston, & Kidder, 2017) and is considered to be an important component of learning (Raspa, McWilliam, & Ridley, 2001). Engagement includes behaviors such as inviting others to join in activities, making friends easily, and introducing oneself to others.

Self-Control. Self-control can be conceptualized as "a set of goal-directed skills that develop from birth onward, enabling humans to act upon their personal and interpersonal aims; overcome difficulties relating to thoughts, emotions, and behaviors; delay gratification; and cope with distress, pain, and disturbing emotions while targeting both internal and external disturbing situations" (Orkibi, Hamama, Gavriel-Fried, & Ronen, 2018, p. 6). According to Gilbert (2006), people find it gratifying to exercise control, whereas the loss of control leads to unhappiness, helplessness, hopelessness, and depression. It has been operationalized by rating scales items such as "inviting others to join in activities," "making friends easily," and "introducing oneself to others." Research indicates that individuals with ID engage in meaningful activities or relationships to a lesser extent than the general population (Qian, Tichá, Larson, Stancliffe, & Wuorio, 2015).

Social Skills Training

Social skills training (SST) teaches specific skills that are targeted to improve social development and reduce behavioral problems (Gresham, Van, & Cook, 2006). In selecting a social skills intervention, it is best to ask a series of questions. First, does the child know how to perform the social skill in question? In other words, do they possess the prerequisite skills to be taught and/or the physical capability of engaging in the behavior? In addition, are there interfering responses present? Interfering responses consist of cognitive and verbal variables such as depressive thoughts, poor problem-solving ability, inadequate role-taking skill, or low self-efficacy. In planning interventions for individuals with ID, it is essential to adequately assess their cognitive functioning with particular focus on problem-solving and role-taking skills. It is also important to evaluate the overt and motoric variables such as disruptive behavior, aggressive behavior, excessive motor movement, or impulsive behavior. It is possible that interventions for these behaviors may need to be built into the social skills training package you plan on using or be dealt with prior to initiating direct social skills training. Similarly, if there are any emotional variables (e.g., anxiety, anger, depression, fear), these may need to be addressed before social skills training is initiated, or they need to be incorporated into the social skills training system.

In developing a social skills intervention program, it is also important to determine if the individual has skills acquisition or performance deficits (Gresham, 2016). Children with social skills deficits do not have the necessary social skills in their repertoires, or they may not know a critical step in the process. For example, a child may not know how to initiate a conversation. This is due to not having an appropriate model or never having the behavior shaped and reinforced. The test of whether this is the problem is to determine if the child has any history of performing the behavior. If there is no history of performing the behavior, it is likely a skill deficit. A subcategory of skills deficits is self-control skills deficit. This is when a child never learned a social skill because some type of interfering response was present which prevented skills acquisition. An example would be anxiety. A child may never learn peer interaction skills because social anxiety prevents social approach behaviors. Aggression also frequently interferes, as others avoid that child and he never has the opportunity to acquire social interaction skills. Performance deficits are when an individual knows how to perform a given behavior but does not perform the behavior at an acceptable level due to some form of interference. If an individual once performed a skill and does not now, or they are performing the behavior but at a very low rate, it is a performance deficit. Self-control performance deficits are a subcategory of performance deficit. Individuals with these have the social skill in their repertoire but do not perform the skill due to antecedent or consequent control. Here the child knows what to do and may even have intentions of doing it, but something gets in the way such as impulsivity (Gresham, 2016).

Gresham (2016) reports that since 1986, there have been 12 narrative reviews of social skills intervention literature with the following conclusions being derived: (a) a combination of modeling, coaching, behavioral rehearsal, and applied behavior analysis procedures appears to be the most effective approaches; (b) cognitive behavioral approaches are not as effective as the procedures mentioned above; and (c) a lack of generalization and maintenance of social skill gains are the greatest weakness of these interventions.

A number of meta-analyses focusing on social skills interventions (SSI) have been conducted over the past three decades. None, however, focused solely on an ID population. Gresham (2016) reviewed nine meta-analyses conducted between 1985 and 2011 and concluded that "approximately 65% of participants in social skills interventions improve relative to only 35% of individuals in control groups" (p. 329).

One meta-analysis that did include individuals with ID was conducted by Park, Kim, and Kim (2016). They conducted a meta-analysis on the effect of job-related social skills training (SST) for secondary students across different types of disabilities, outcomes, settings, and instruction. Regarding disability, the effect of SST was not significant for students with Down syndrome and learning disabilities. Other disabilities, such as intellectual disabilities, autism spectrum disorders, and emotional/behavioral disorders, showed moderate effect sizes. The effect sizes by subgroup analysis indicated the following effect sizes: intellectual disabilities ($ES = 0.487$), Down syndrome ($ES = 0.159$), autism ($ES = 0.445$), no specific ($ES = 0.856$), emotional and/or behavior disorder ($ES = 0.505$), and learning disabilities ($ES = 0.495$).

Gates, Kang, and Lerner (2017) conducted a meta-analysis on group-based social skills interventions (GSSIs) with youth with autism spectrum disorder (ASD). While the study did not include individuals with ID (mean IQ was 102), the results are useful in understanding the efficacy of GSSIs. Results indicated that the overall effect size was medium (g = 0.51). Effects were largest on self-report measures (g = 0.92), smaller for parent- (g = 0.47) and observer-report measures (g = 0.40), and smallest for teacher-report measures (g = 0.11). The authors concluded that social skills interventions appear modestly effective for youth with ASD, but may not generalize to school settings or self-reported social behavior. Research has shown that individuals with average or higher IQ and better developed verbal ability tend to benefit most from GSSIs (Herbrecht et al., 2009). Therefore, individuals with ID who have more highly developed cognitive and verbal abilities may show greater improvements from GSSIs than those individuals who have less-developed skills. Another important conclusion that may be drawn from this research involves the type of measure. Self-report measures had a large effect size, parent and observer measures moderate effect sizes, and teacher measures a minimal effect size. It is possible that rater bias may be influencing the scores on self- and other-report measures. Rater bias is a form of nonrandom measurement error or systematic variance that is attributable to the rater (Hoyt & Kerns, 1999), and rater bias may artificially inflate or suppress assessment scores relative to the actual frequency of behavior (Shapiro, Kim, Accomazzo, & Roscoe, 2016).

Wang, Cui, and Parrila (2011) conducted a meta-analysis using hierarchical linear modeling (HLM) to examine the effectiveness of peer-mediated and video modeling approaches, for social skills training of children with ASD. Both interventions (mean effect size = 1.27) were similarly effective and found to significantly impact the social performance of children with ASD. While level of cognitive functioning was not examined in this analysis, age functioned as a significant moderator in the effectiveness of the intervention with the interventions being slightly more effective for younger children. Similarly, Wang and Spillane (2009) conducted a meta-analysis on interventions to increase social skills for children and adolescents with ASD and attempt to determine whether interventions met the criteria for evidence-based practice. A total of 38 studies with 147 total participants were examined. Types of interventions examined using percentage of non-overlapping data points (PND) were social stories (n = 6), peer-mediated (n = 9), video modeling (n = 11), cognitive behavior training (n = 3), and others (n = 9). Results indicated the following: social stories (mean PND = 67.21%), peer-mediated (mean PND = 60.69%), and video modeling (mean PND = 84.25); leading the authors to conclude that only video modeling meets criteria as an evidence-based intervention. Due to the small n for cognitive behavior training studies and the variability in the others category, the authors concluded that while promising, firm conclusions could not be made as to the efficacy of these procedures.

Bellini, Peters, Benner, and Hopf (2007) conducted a meta-analysis examining the effectiveness of school-based social skills interventions for children and adolescents with ASD. Results indicated that social skills interventions produce low treatment and generalization effects and moderate maintenance effects for children with ASD. Using the percentage of non-overlapping data points (PND) as the metric, results indicated that just over half of the reviewed studies (52%) produced mean intervention PND scores above 70%. The results also suggested moderate maintenance effects (mean PND = 80%) and low generalization effects (mean PND = 53%). The authors concluded that results “suggest that school-based social skills interventions are minimally effective for children with ASD” (p. 159).

Bellini and Akullian (2007) conducted a meta-analysis analyzing 23 single-subject design research studies using the percentage of non-overlapping data points with children with ASD. Results indicated that both video modeling (VM) and video self-modeling (VSM) are effective intervention strategies to improve social communication, behavior, and functional skills in children with ASD. Additionally, results showed that VM and VSM not only promote skills acquisition but also have a lasting effect. While this study focused on children with ASD, it appears that VM and VSM may be promising approaches for social skills training in children with ID, provided they possess the prerequisite skills for these procedures. Note, these will be addressed later in this chapter.

Magee Quinn, Kavale, Mathur, Rutherford, and Forness (1999) conducted a meta-analysis on social skills interventions for students with emotional or behavioral disorders (EBD). Examining the data from 35 studies, their database yielded a pooled mean effect size of only 0.199, reflecting only a small gain as a result of a social skills intervention. These results are comparable to a meta-analysis examining the effects of social skills interventions on students with learning disabilities (Kavale & Forness, 1996) which found a mean effect size of 0.211. Magee Quinn et al. conclude that because developing appropriate social skills is critical to functioning in our society, social skills training should be refined and not eliminated.

Höher Camargo et al. (2016) examined behaviorally based interventions to teach social interaction skills for children with ASD in inclusive settings. All studies included in their analysis used peer or adult modeling, different prompt hierarchies and types (e.g., most to least or least to most hierarchy and verbal, visual, written, or tactile prompt), and/or positive reinforcement (e.g., praise, tangibles, and/or edibles) with a combination of these techniques used across the studies. Results indicated an overall effect size of 0.88 indicating a high magnitude of change. The authors concluded that "behaviorally based interventions were highly effective for improving social interaction skills of young children with ASD (ages 2–10) included in general education" (p. 240).

Whalon, Conroy, Martinez, and Werch (2015) performed a meta-analysis to evaluate the impact of school-based interventions to facilitate the peer-related social competence of children with ASD. Using results from 37 studies, target behaviors generally fit into one of four categories: initiations, responses, interactions, and engagement. Results indicated that studies using an adult-mediated intervention had a mean of 93% for non-overlapping data points and an effect size (Tau-U) of 0.85 indicating a positive impact on the social interactions of children with ASD. Studies using social narratives produced mixed findings with a mean overall of non-overlapping data points of 83% and an effect size of 0.65 (Tau-U). Targeting engagement was high (mean non-overlapping data points = 99%, *Tau-U* = 0.94) but variable for interactions (mean non-overlapping data points = 80%, *Tau-U* = 0.60), initiations (mean non-overlapping data points = 64%, *Tau-U* = 0.42), and responses (mean non-overlapping data points = 67%, *Tau-U* = 0.34). Results from VSM/VM studies were mixed (mean non-overlapping data points = 86% *Tau-U* = 0.71), but there was a large range between studies (non-overlapping data points = 57%–99%, *Tau-U* = 0.13–0.97). These results led the authors to conclude that "peer-related social competence interventions delivered in school settings produced a moderate to high impact regardless of intervention type (i.e., child-specific, peer-mediated, multi-component, and collateral skill), child characteristics (e.g., social initiations prior to intervention, cognitive ability, age), or outcome measure" (p. 1526).

Sukhodolsky and Butter (2007) identify five main techniques used in social skills training (SST) for children with intellectual disabilities: instruction, modeling, rehearsal, corrective feedback, and reinforcement for appropriate performance. Some SSTs rely exclusively on operant conditioning techniques (e.g., positive reinforcement), but most also include social learning models to aid in skill development. The goal of most SSTs with children with ID is usually the development of specific social skills as opposed to broader social interactional constructs. Depending on the developmental level of the child, targeted skills may differ in complexity from simple responses such as saying hello, making eye contact, and smiling to more complex behavioral repertoires used in interpersonal and vocational settings.

Promoting Skills Acquisition. Skills acquisition is the first thing that must be done when the child lacks the skill and there are no interfering behaviors. Modeling is frequently used in promoting skills acquisition. It is based on Bandura's

(1969) social learning theory. The basic premise is that by observing the successful demonstration of a task, the viewer learns to perform the task effectively. Modeling has consistently been shown to be an effective and efficient way to teach social behavior. The behavior to be modeled can be a simple discrete behavior or can involve an entire sequence of behaviors involved in a social skill. Coaching is another technique that can be used in skills acquisition. This is just direct verbal instruction where you provide rules, have the child rehearse behavior, and they you provide feedback.

Enhancing Skill Performance. The goal with this approach is to increase the likelihood of the social skill in a child who is not performing it by increasing the strength of the social skill and not by targeting the interfering behavior. In other words, by increasing the strength of the social skill, you indirectly weaken the interfering behavior. Techniques that can be used to enhance skill performance include behavioral rehearsal, reinforcement, peer initiation strategies, and cooperation learning strategies. Behavioral rehearsal involves having the child rehearse the behavior until they feel comfortable with it. Reinforcement-based techniques involve using contingency management, particularly positive reinforcement, to reinforce appropriate social skills at home and in classroom. Peer-initiation strategies incorporate other children in the class to initiate social contact with the target child as a cue to trigger the desired social behavior in the target child. In other words, you are providing the child with appropriate antecedents in the environment to signal particular social behaviors. Cooperation learning strategies are generally implemented through a group format where you use many of the other techniques described above while working on cooperative interactions. Role-playing is also used as a technique in cooperative learning.

Removing Interfering Behaviors. Unfortunately, many people overlook interfering behaviors and just try to teach positive behaviors without attempting to lessen the extent of any interfering behaviors that may exist. An assumption many people make is that the child is engaging in the inappropriate behaviors because he/she lacks appropriate social skills. This may be true sometimes, but many times work needs to first focus on the interfering behavior. Techniques such as response cost procedures, group contingencies where the entire class is targeted for behavior change (Skinner, Skinner, & Burton, 2009), or overcorrection in the form of positive practice or restitution are used.

Facilitating Generalization. One of the greatest weaknesses in social skills training is a lack of generalization and maintenance (Gresham, 2016). If you want the behavior to generalize to the natural environment, you must incorporate it into training. To facilitate generalization it is recommended that you take advantage of natural reinforcement. In other words, recognize what naturally occurring reinforcement exists in the environment, and use that as much as possible. You should also train diversely. That is, do not just take just one approach. Train the individual that there are different ways to handle situations. Emphasize that things do not proceed in real world exactly the way they did in the training environment. Make the individual aware of problems they may encounter. Of greatest importance with individuals with ID is to incorporate functional mediators into their environment. The narrow confines of the SST training environment, be it school or other controlled setting, simply may not give the individual enough training opportunities. Adults (e.g., teacher, parent), or possibly siblings or peers, can act as a mediator and coach in the natural environment. They can provide antecedent cues for engagement in certain behaviors and provide reinforcement (both verbal and tangible) on the occurrence of prosocial behaviors.

Discrete Trials

Discrete trials training is a practical instructional method used for implementing social and academic skills to students with ID and ASD. DTT incorporates the principle of ABA by focusing on a systematic way to facilitate learning, prompting accurate responding, correcting response errors, and delivering positive reinforcement (Leblanc, Ricciardi, & Luiselli, 2005). Anderson, Taras, and Cannon (1996) identify four distinct parts to DTT: (1) the trainer's presentation, (2) the child's response, (3) the consequence (e.g., reinforcement), and (4) a short pause between the consequence and the next instruction (between interval trials). Cooper, Heron, and Heward (2007) define a discrete trial as "any operant whose response rate is controlled by a given opportunity to emit the response. Each discrete response occurs when an opportunity to respond exists" (p. 694). DTT has established a history of effectiveness as the primary method used in early intensive behavioral interventions for ID and ASD students (Eikeseth, Smith, Jahr, & Eldevik, 2007). The use of DTT programs for students in schools is rare (Peters-Scheffer, Didden, Mulders, & Korzillus, 2010), primarily due to requirements for one-to-one instruction and the training required of staff (Dart, Radley, Furlow, & Murphy, 2017). Dart and colleagues tried to overcome some of the difficulties encountered in using DTT in schools by training high school students to implement DTT. Results indicated that while brief didactic training was not sufficient to produce acceptable levels of treatment integrity, a more intensive behavior skills package of training did produce acceptable levels of integrity.

Social Stories

Social stories are a growing topic in the education field. They are a behavioral intervention to improve children's social skills through a combination of visual and verbal cues. Their use is primarily with children who have been diagnosed with mild to severe disabilities and have been to teach appropriate skills such as interacting with others, prompting, joining-in behavior, sportsmanship, maintaining a conversation, specific skills acquisition, responding to cues, and describing settings (O'Hara, 2010). Barry and Burlew (2004) describe social stories as including empirically validated components such as modeling, task analysis, visual aids, practice and corrective feedback, and priming. When using social stories, the child or someone with whom he/she identifies reads a story that describes the character's behaviors, thoughts, and feelings as he/she accomplishes the behavioral goals outlined in the story (Barry & Burlew, 2004). Other people's perspectives can also be included in the story. McGill, Baker, and Busse (2015) conducted a meta-analysis of 27 single-subject research studies ($n = 77$) on social stories and found moderate to large treatment effects ($ES = 0.79$). They appear to be a promising treatment option for children with ID although the totality of the research indicates future research is still needed.

Role-Playing/Behavioral Rehearsal

It may be difficult to teach complex social behaviors using a strictly rule- and instruction-based approach (Bilich & Ciarrochi, 2009), and experience is a needed component to learn most social skills. Role-playing provides a venue in which individuals can practice behaviors which are emerging, or which they may find difficult, in a relatively safe setting. Role-playing/behavioral rehearsal has been used extensively to teach basic social interaction skills as well as other more complex social skill-related behaviors. It is an effective approach to teaching social skills that allows for the positive practice of skills (Gresham, 2010). Role-playing involves acting out situations or activities in a structured environment to practice newly acquired skills and strategies, or previously learned skills that the child is having difficulty performing. Role-plays can be either scripted or less structured with built in prompts. An important consideration in determining whether to use role-play in an intervention with an individual with an intellectual ability is the level of functioning and their ability to pay attention and take the perspective of another.

Video Self-Modeling

Video self-modeling (VSM) is a cognitive behavioral technique that provides participants with visual images of themselves performing a target behavior, such as on-task behavior, or interacting appropriately with peers. While it has been used primarily with individuals with ASD, it should be equally effective with individuals with ID. A video modeling intervention typically involves an individual watching a video demonstration of a model engaging in specific a behavior and then imitating the behavior in the video. The model can be a parent, teacher, therapist, another adult, age-level peer, or in the case of VSM, the model is the child him/herself (Bellini & Akullian, 2007; D'Ateno, Mangiapanello, & Taylor, 2003; Dowrick, Kim-Rupnow, & Power, 2006). The model is video recorded within a certain context (e.g., the classroom or playground). The recording is then edited to remove all instances of inappropriate behavior. The result is a video clip displaying the individual behaving appropriately and displaying the correct, targeted behavior.

Video self-modeling is a technique where individuals view themselves performing a desired behavior on video in order to increase the occurrence of that behavior or behaviors. Participants view their own positive behavior on edited video clips where all evidence of inappropriate and maladaptive behavior is removed. Bandura's (1977) social cognitive learning theory provided the theoretical basis for VSM. His view of learning emphasized the ability to learn through observation. Gul and Vuran (2010) write that the theory suggests that individuals gain knowledge and skills by observing behaviors which are displayed. The main advantage of this type of learning is that it provides the learner with clear information on how best to perform the skill. Bandura (1977) identifies four steps in the process of learning through modeling: (1) the individual must attend to the events being modeled, (2) modeled behavior must be retained, (3) symbolic representation of the behavior is converted into appropriate actions similar to the originally modeled behavior, and (4) the individual must be motivated to replicate the modeled behavior. Bandura goes on to explain that the success of the model being observed depends largely on the similarities between the observer and the model. The higher the number of similarities, the higher the success of the observer engaging in desired behavior. Gul and Vulcan concluded that video modeling interventions, where participants took part as the model, acquired target skills in a shorter time compared to participants who were not the model. VSM maximizes its chance of success by using the observer as the model of the desired behavior (Gelbar, Anderson, & McCarthy, 2012). Buggey (2007) writes that self-efficacy is at the heart of VSM with VSM helping to promote self-efficacy by providing participants with visual evidence that they can succeed or achieve the target.

Conclusion

Social skill excesses and deficits are a defining aspect of intellectual disabilities. Intervention targeting improving areas of deficit and decreasing excesses can aid in normalization and improve quality of life. This chapter reviewed definitions of social skills both in general and in the context of intellectual disabilities. It further broke down social skills into seven specific domains, communication, cooperation, assertion, responsibility, empathy, engagement, and self-control, and discussed the importance of each. A number of approaches to social skills training were also discussed with specific emphasis placed on interventions for individuals with intellectual disabilities ranging from discrete trials training which has been used to teach specific social behaviors to individuals with more severe intellectual disabilities to role-playing and video self-modeling which can be valuable treatment approaches for those with a higher level of functioning. Regardless of the approach used for SST, it is important to recognize the importance of incorporating procedures to increase the likelihood of generalization and maintenance of the learned behavior.

References

American Psychiatric Association. (2013). *Diagnostic and statistical manual of mental disorders* (5th ed.). Washington, DC: Author.

Anderson, S. R., Taras, M., & Cannon, B. O.'. M. (1996). Teaching new skills to young children with autism. In C. Maurice, G. Green, & S. C. Luce (Eds.), *Behavioral intervention for young children with autism: A manual for parents and professionals* (pp. 181–194). Austin, TX, US: PRO-ED.

Bandura, A. (1977). *Social learning theory*. Englewood Cliffs, NJ: Prentice Hall.

Barry, L. M., & Burlew, S. B. (2004). Using social stories to teach choice and play skills to children with autism. *Focus on Autism and Other Developmental Disabilities, 19*, 45–51. https://doi.org/10.1177/10883576040190010601

Bellini, S., & Akullian, J. (2007). A meta-analysis of video modeling and video self-modeling interventions for children and adolescents with autism spectrum disorders. *Exceptional Children, 73*, 264–287. https://doi.org/10.1177/001440290707300301

Bellini, S., Peters, J. K., Benner, L., & Hopf, A. (2007). A meta-analysis of school-based social skills interventions for children with autism spectrum disorders. *Remedial and Special Education, 28*, 153–162. https://doi.org/10.1177/07419325070280030401

Belva, B. C., Matson, J. L., Sipes, M., & Bamburg, J. W. (2012). An examination of specific communication deficits in adults with profound intellectual disabilities. *Research in Developmental Disabilities, 33*, 525–5299. https://doi.org/10.1016/j.ridd.2011.10.019

Bilich, L. L., & Ciarrochi, J. (2009). Promoting social intelligence using the experiential role-play method. In J. T. Blackledge, J. Ciarrochi, & F. P. Deane (Eds.), *Acceptance and commitment therapy: Contemporary theory, research and practice* (pp. 247–262). Bowen Hills, QLD: Australian Academic Press.

Buggey, T. (2007). A picture is worth…Video self-modeling applications at school and home. *Journal of Positive Behavior Interventions, 9*, 151–158.

Caldarella, P., & Merrell, K. W. (1997). Common dimensions of social skills of children and adolescents: A taxonomy of positive behaviors. *School Psychology Review, 26*, 264–278.

Constantino, J. N., Przybeck, T., Friesen, D., & Todd, R. D. (2000). Reciprocal social behavior in children with and without pervasive developmental disorders. *Journal of Developmental and Behavioral Pediatrics, 21*, 2–11. https://doi.org/10.1097/00004703-200002000-00002

Cook, C. R., Gresham, L. K., Barreras, R. B., Thornton, S., & Crews, S. D. (2008). Socials skills training for secondary students with emotional and/or behavioral disorders: A review and analysis of the meta-analytic literature. *Journal of Emotional and Behavioral Disorders, 16*, 131–144. https://doi.org/10.1177/1063426608314541

Cooper, J. O., Heron, T. E., & Heward, W. L. (2007). *Applied behavior analysis* (2nd ed.). Upper Saddle River, NJ: Pearson Education.

Cronin, K. A. (2017). Comparative studies of cooperation: Collaboration and prosocial behavior in animals. In J. Call, G. M. Burghardt, I. M. Pepperberg, C. T. Snowdon, & T. Zentall (Eds.), *APA handbook of comparative psychology: Basic concepts, methods, neural substrate, and behavior* (pp. 915–929). Washington: American Psychological Association.

Cuff, B. M. P., Brown, S. J., Taylor, L., & Howat, D. J. (2016). Empathy: A review of the concept. *Emotion Review, 8*, 144–153. https://doi.org/10.1177/1754073914558466

D'Ateno, P., Mangiapanello, K., & Taylor, B. A. (2003). Using video modeling to teach complex play sequences to a pre-schooler with autism. *Journal of Positive Behavior Interventions, 5*, 5–11.

Dart, E. H., Radley, K. C., Furlow, C. M., & Murphy, A. N. (2017). Using behavioral skills training to teach high school students to implement discrete trial training. *Behavior Analysis: Research and Practice, 17*, 237–249. https://doi.org/10.1037/bar0000075

de Bildt, A., Serra, M., Luteijn, E., Kraijer, D., Sytema, S., & Minderaa, R. (2005). Social skills in children with intellectual disabilities with and without autism. *Journal of Intellectual Disability Research, 49*, 317–328. https://doi.org/10.1111/j.1365-2788.2005.00655.x

Doll, E. A. (1941). The essentials of an inclusive concept of mental deficiency. *American Journal on Mental Deficiency, 46*, 214–219.

Dowrick, P. W., Kim-Rupnow, W. S., & Power, T. J. (2006). Video feedforward for reading. *Journal of Special Education, 39*, 194–219.

Eikeseth, S., Smith, T., Jahr, E., & Eldevik, S. (2007). Outcome for children with autism who began intensive behavioral treatment between ages 4 and 7: A comparison controlled study. *Behavior Modification, 31*, 264–278. https://doi.org/10.1177/0145445506291396

Eisenberg, N., & Strayer, J. (1987). *Empathy and its development*. New York: Cambridge University Press.

Elliott, S. N., & Busse, R. T. (1991). Social skills assessment and intervention with children and adolescents: Guidelines for assessment and training procedures. *School Psychology International, 12*, 63–83. https://doi.org/10.1177/0143034391121006

Gates, J. A., Kang, E., & Lerner, M. D. (2017). Efficacy of group social skills interventions for youth with autism spectrum disorder: A systematic review and meta-analysis. *Clinical Psychology Review, 52*, 164–181. https://doi.org/10.1016/j.cpr.2017.01.006

Gelbar, N. W., Anderson, C., & McCarthy, S. (2012). Video self-modeling as an Intervention strategy for individuals with autism spectrum disorders. *Psychology in Schools, 49*, 15–22.

Gilbert, D. T. (2006). *Stumbling on happiness*. New York, NY: Alfred A. Knopf.

Greenspan, S. (1981). Social competence and handicapped individuals: Practical implications and proposed model. *Advances in Special Education, 3*, 41–82.

Greenspan, S. (1999). What is meant by mental retardation? *International Review of Psychiatry, 11*, 6–18. https://doi.org/10.1080/09540269974474

Gresham, F. M. (1983). Social skills assessment as a component of mainstreaming placement decisions. *Exceptional Children, 49*, 331–336. https://doi.org/10.1177/001440298304900406

Gresham, F. M. (1986). Conceptual and definitional issues in the assessment of children's social skills: Implications for classification and training. *Journal of Clinical Child Psychology, 75*, 3–15. https://doi.org/10.1207/s15374424jccp1501_1

Gresham, F. M. (1997). Social competence and students with behavior disorders: Where we've been, where we are, and where we should go. *Education and Treatment of Children., 20*, 233–249.

Gresham, F. M. (2010). Evidence-based social skills interventions: Empirical foundations for instructional approaches. In M. Shinn & H. Walker (Eds.), *Interventions for achievement and behavior problems in a three-tier model including RTI* (pp. 337–362). Bethesda, MD: National Association of School Psychologists.

Gresham, F. M. (2016). Social skills assessment and intervention for children and youth. *Cambridge Journal of Education, 46*, 319–332. https://doi.org/10.1080/0305764X.2016.1195788

Gresham, F. M., & Elliott, S. N. (2008). *Social skills improvement system rating scales*. San Antonio, TX: Pearson Assessment.

Gresham, F. M., Van, M. B., & Cook, C. R. (2006). Social skills training for teaching replacement behaviors: Remediating acquisition deficits in at-risk students. *Behavioral Disorders, 31*, 363–377.

Gul, S. O., & Vuran, S. (2010). *An analysis of studies conducted video modeling in teaching social skills. Kuram ve Uygulanada Egitim Bilimleri* (Vol. 10, pp. 249–257).

Herbrecht, E., Poustka, F., Birnkammer, S., Duketis, E., Schlitt, S., Schmötzer, G., & Bölte, S. (2009). Pilot evaluation of the Frankfurt Social Skills Training for children and adolescents with autism spectrum disorder. *European Journal of Child & Adolescent Psychiatry, 18*, 327–335. https://doi.org/10.1007/s00787-008-0734-4

Höher Camargo, S. P., Rispoli, M., Ganz, J., Hong, E. R., Davis, H., & Mason, R. (2016). *Journal of Behavioral Education, 25*, 223–248. https://doi.org/10.1007/s10864-015-9240-1

Hops, H. (1983). Children's social competence and skill: Current research practices and future directions. *Behavior Therapy, 14*, 3–18. https://doi.org/10.1016/s0005-7894(83)80084-7

Hoyt, W. T., & Kerns, M.-D. (1999). Magnitude and moderators of bias in observer ratings: A meta-analysis. *Psychological Methods, 4*, 403–424. https://doi.org/10.1037/1082-989X.4.4.403

Kasari, C., Freeman, S. F. N., & Bass, W. (2003). Empathy and response to distress in children with Down syndrome. *Journal of Child Psychology and Psychiatry and Allied Disciplines, 44*, 424–431.

Kavale, K. A., & Forness, S. R. (1996). Social skills deficits and learning disabilities: A meta-analysis. *Journal of Learning Disabilities, 29*, 226–237. https://doi.org/10.1177/002221949602900030

Kazdin, A. E. (1977). Assessing the clinical or applied importance of behavior change through social validation. *Behavior Modification, 1*, 427–451. https://doi.org/10.1177/014544557714001

Kurtz, P. F., Boetler, E. W., Jarmolowicz, D. P., Chin, M. D., & Hagopian, L. P. (2011). An analysis of functional communication training as an empirically supported treatment for problem behavior displayed by individuals with intellectual disabilities. *Research in Developmental Disabilities, 32*, 2935–2942. https://doi.org/10.1016/j.ridd.2011.05.009

Lane, K. L., Givner, C. C., & Pierson, M. R. (2004). Teacher expectations of student behavior: Social skills necessary for success in elementary school classrooms. *Journal of Special Education, 38*, 104–111.

Leblanc, M., Ricciardi, J., & Luiselli, J. (2005). Improving discrete trial instruction by paraprofessional staff through an abbreviated performance feedback intervention. *Education & Treatment of Children, 28*, 76–82.

Libet, J., & Lewinsohn, M. (1973). Concept of social skills with special reference to the behavior of depressed patients. *Journal of Consulting and Clinical Psychology, 401*, 304–312.

Light, J. (1989). Toward a definition of communicative competence for individuals using augmentative and alternative communication systems. *Augmentative and Alternative Communication, 5*, 137–144.

Little, S. G., Swangler, J., & Akin-Little, A. (2017). Defining social skills. In J. Matson (Ed.), *Handbook of social behavior and skills in children* (pp. 9–17). New York: Springer. ISBN 978-3-319-64592-6.

Magee Quinn, M., Kavale, K. A., Mathur, S. R., Rutherford, R. B., & Forness, S. R. (1999). A meta-analysis of social skill interventions for students with emotional or behavioral disorders. *Journal of Emotional and Behavioral Disorders, 7*, 54–64. https://doi.org/10.1177/106342669900700106

Matson, J. L., Dempsey, T., & LoVullo, S. V. (2009). Characteristics of social skills for adults with intellectual disability, autism and PDD-NOS. *Research in Autism Spectrum Disorders, 3*, 207–213. https://doi.org/10.1016/j.rasd.2008.05.006

Matson, J. L., Terlonge, C., González, M. L., & Rivet, T. (2006). An evaluation of social and adaptive skills in adults with bipolar disorder and severe/profound intellectual disability. *Research in Developmental Disabilities, 27*, 681–687. https://doi.org/10.1016/j.ridd.2005.10.001

McGill, R. J., Baker, D., & Busse, R. T. (2015). Social Story™ interventions for decreasing challenging behaviours: A single-case meta-analysis 1995–2012. *Educational Psychology in Practice, 31*, 21–42. https://doi.org/10.1080/02667363.2014.975785

Meier, C. R., DiPerna, J. C., & Oster, M. M. (2006). Importance of social skills in the elementary grades. *Education & Treatment of Children, 29*, 409–419.

Nelson, C., Paul, K., Johnston, S. S., & Kidder, J. E. (2017). Use of a creative dance intervention package to increase social engagement and play complexity of young children with autism spectrum disorder. *Education and Training in Autism and Developmental Disabilities, 52*, 170–185.

O'Hara, M. (2010). Using social stories to teach social skills: A professional's guide. University of Pittsburgh. Avaiable at: https://www.sbbh.pitt.edu/files/Powerpoint%20Presentations%202524%20Spring%202010/USING%20SOCIAL%20STORIES%20TO%20TEACH%20SOCIAL%20SKILLS.pdf.

Oden, S., & Asher, S. R. (1977). Coaching children in social skills for friendship making. *Child Development, 48*, 495–506. https://doi.org/10.2307/1128645

Orkibi, H., Hamama, L., Gavriel-Fried, B., & Ronen, T. (2018). Pathways to adolescents' flourishing: Linking self-control skills and positivity ratio through social support. *Youth & Society, 50*, 3–25. https://doi.org/10.1177/0044118X15581171

Park, E., Kim, J., & Kim, S. (2016). Meta-analysis of the effect of job-related social skill training for secondary students with disabilities. *Journal of Vocational Rehabilitation, 44*, 123–133. https://doi.org/10.3233/JVR-150785

Parmenter, T. R. (2011). What is Intellectual Disability? How is it assessed and classified? *International Journal of Disability, Development, and Education, 58*, 303–319. https://doi.org/10.1080/1034912X.2011.598675

Peters-Scheffer, N., Didden, R., Mulders, D., & Korzillus, H. (2010). Low intensity behavioral treatment supplementing preschool services for young children with autism spectrum disorders and severe to mild intellectual disability. *Research in Developmental Disabilities, 31*, 1678–1684. https://doi.org/10.1016/j.ridd.2010.04.008

Qian, X., Tichá, R., Larson, S. A., Stancliffe, R. J., & Wuorio, A. (2015). The impact of individual and organizational factors on engagement of individuals with intellectual disability living in community group homes: A multilevel model. *Journal of Intellectual Disability Research, 59*, 493–505. https://doi.org/10.1111/jir.12152

Ralfs, S., & Beail, N. (2012). Assessing components of empathy in sex- offenders with intellectual disabilities. *Journal of Applied Research in Intellectual Disabilities, 25*, 50–59. https://doi.org/10.1111/j.1468-3148.2011.00648.x

Raspa, M. J., McWilliam, R. A., & Ridley, S. (2001). Child care quality and children's engagement. *Early Education and Development, 12*, 209–224.

Schalock, R. L., Borthwick-Duffy, S. A., Bradley, V. J., Buntinx, W. H. E., Coulter, D. I., Craig, E. M., … Yeager, M. H. (2010). *Intellectual disability. Definition, classification, and systems of support*. Washington, DC: AAIDD.

Schindler, A., Ruoppolo, G., & Barillari, U. (2010). Communication and its disorders: Definition and taxonomy from a phoniatric perspective. *Audiological Medicine, 8*, 163–170. https://doi.org/10.3109/1651386X.2010.530023

Semrud-Clikeman, M. (2007). *Social competence in children*. New York: Springer.

Shapiro, V. B., Kim, B. K. E., Accomazzo, S., & Roscoe, J. N. (2016). Predictors of rater bias in the assessment of social-emotional competence. *The International Journal of Emotional Education, 8*(2), Special Issue: SEL Training, Intervention and Research Worldwide), 25–44.

Skinner, C. H., Skinner, A. L., & Burton, B. (2009). Applying group-oriented contingencies in the classroom. In A. Akin-Little, S. G. Little, M. Bray, & T. Kehle (Eds.), *Behavioral Interventions in Schools: Evidence-Based Positive Strategies* (pp. 157–170). Washington, DC: APA Books.

Sukhodolsky, D., & Butter, E. M. (2007). Social skills training for children with intellectual disabilities. In J. W. Jacobson, J. A. Mulick, & J. Rojahn (Eds.), *Handbook of intellectual and developmental disabilities* (pp. 601–618). New York: Springer.

van der Meer, L., Sigafoos, J., O'Reilly, M. F., & Lancioni, G. E. (2011). Assessing preferences for AAC options in communication interventions for individuals with developmental disabilities: A review of the literature. *Research in Developmental Disabilities, 32*, 1422–1431. https://doi.org/10.1016/j.ridd.2011.02.003

Wang, P., & Spillane, A. (2009). Evidence-based social skills interventions for children with autism: A meta-analysis. *Education & Training in Developmental Disabilities, 44*, 318–342.

Wang, S., Cui, Y., & Parrila, R. (2011). Examining the effectiveness of peer-mediated and video-modeling social skills interventions for children with autism spectrum disorders: A meta-analysis in single-case research using HLM. *Research in Autism Spectrum Disorders, 5*, 562–569. https://doi.org/10.1016/j.rasd.2010.06.023

Whalon, K. J., Conroy, M. A., Martinez, J. R., & Werch, B. L. (2015). School-based peer-related social competence interventions for children with autism spectrum disorder: A meta-analysis and descriptive review of single case research design studies. *Journal of Autism and Developmental Disorders, 45*, 1513–1531. https://doi.org/10.1007/s10803-015-2373-1

Wolf, M. M. (1978). Social validity: The case for subjective measurement or how behavior analysis is finding its heart. *Journal of Applied Behavior Analysis, 11*, 203–214. https://doi.org/10.1901/jaba.1978.11-203

World Health Organization. (2010). *Mental retardation (F70-F79). International statistical classification of diseases and related health problems* (10th ed.). Revisions (ICD-10). Retrieved from http://apps.who.int/classifications/icd10/browse/2010/en#/F70-F79

38

Coping Skills Training in Individuals with an Intellectual Disability

Renae Beaumont, Amy Lemelman,
Jennifer Schild, and Karen Tang

Introduction

The mental health and well-being of individuals with an intellectual disability (ID) have received scant empirical attention until the past 20 years. This chapter provides an overview of therapy approaches that have been evaluated for children, adolescents, and adults with an ID, including cognitive behavioral therapy and mindfulness-based interventions. A critical analysis of contemporary research evaluating the effectiveness of these treatments for a range of mental health issues is presented, together with a description of how the content and process of therapy can be appropriately tailored to meet the needs of clients with an ID. Core principles for adapting therapy are also illustrated with a case example. The chapter concludes with recommendations for future research to improve the quality and accessibility of mental health services to this underserved population.

Until the twenty-first century, the mental health and well-being of individuals with intellectual disabilities (ID) received little empirical attention (Whitehouse, Tudway, Look, & Kroese 2006). Arthur (2003) largely ascribes this to institutionalization, which, for many, led to an attitude of "out of sight, out of mind" (p 26). More recent research shows that individuals with ID experience full emotional lives (Jones, Miller, Williams, & Goldthorpe 1997), have the ability to reflect on and describe their emotional states (Stenfert Kroese, Dagnan, & Loumidis 1998), and suffer from the same mental health problems as the general population, often at higher rates (Lunksky et al. 2012). Smiley (2005) reported the prevalence of mental illness in people with ID to be between 30% and 50%, with Cooper, Heron, and Heward (2007) reporting a 40.9% prevalence rate in a population study. However, "diagnostic overshadowing" often results in mental health issues being underdiagnosed in individuals with an ID (Hemmings, Deb, Chaplin, Hardy, & Mukherjee 2013). This refers to presenting concerns being attributed to patients' ID or clinicians' attention being focused on reducing overt challenging behaviors, rather than investigating and treating mental health issues that may contribute to them.

Historically, individuals with ID have had very limited access to interventions for psychological

R. Beaumont (✉) · J. Schild
Weill Cornell Medicine, Child and Adolescent Psychiatry, New York, NY, USA
e-mail: rbb2002@med.cornell.edu

A. Lemelman · K. Tang
Weill Cornell Medicine, Center for Autism and the Developing Brain, New York, NY, USA

J. L. Matson (ed.), *Handbook of Intellectual Disabilities*, Autism and Child Psychopathology Series, https://doi.org/10.1007/978-3-030-20843-1_38

problems (Dodd & McGinnity 2003; Lynch 2004). Bender (1993) attributes this to the reluctance of many mental health professionals to treat this client group, perhaps due to assumptions about these individuals being unable to effectively participate and collaborate in treatment due to their disability. Heifetz and Dyson (2017) also note a tradition in the ID field for professionals to overly rely on behavior modification techniques and medication to address patient psychological or behavioral issues. These methods appear to show limited concern for the client's well-being (Stenfert Kroese 1998) and, in many cases, may be more aligned with meeting the needs of service providers or carers (Waitman & Reynolds 1992).

Encouragingly, in more recent times, studies examining the effectiveness of psychotherapeutic interventions for individuals with ID have begun to emerge. These have predominantly included evaluations of psychotherapy for anxiety (e.g., Douglass, Palmer, & O'Connor 2007; Lindsay, Neilson, & Lawrenson 1997; Marwood & Hewitt 2012), depression (e.g., Hassiotis et al. 2013; Unwin, Tsimopoulou, Stenfert Kroese, & Azmi 2016), and anger (e.g., Nicoll, Beail & Saxon, 2013; Willner et al. 2013). Most studies have been conducted with adults who have mild-to-moderate ID and have involved evaluations of CBT and, more recently, mindfulness-based interventions. This chapter provides an overview of psychological intervention techniques that have some empirical basis for children, adolescents, and adults with an ID, together with a critical analysis of the evidence supporting these approaches. The chapter also describes how the content and process of therapy can be successfully adapted for clients with an ID, including a case example. Lack of clinician knowledge, skills, and confidence in how to effectively tailor therapy to meet the needs of individuals with an ID is purported to be a major contributor to the lack of service availability for this client population (Whitehouse et al. 2006). This handbook is an important step in bridging this professional practice gap.

Cognitive Behavior Therapy for Adults with an ID

A central premise of CBT is that psychological disorders are characterized by distorted or dysfunctional thinking (Beck 1976). As such, clients learn to develop more helpful thinking patterns and behaviors by changing their thoughts, attitudes, and beliefs, in addition to making more realistic or adaptive assessments of events that cause distress (Beck 1976; Willner 2009). Success in CBT requires the client to engage in cognitive and behavioral tasks, such as identifying automatic thoughts; cognitive monitoring; identifying relationships between thoughts, feelings, and behaviors; relaxation skills training; cognitive restructuring; and engaging in behavioral experimentation (Beck 2011; Cooney, Tunney, & O'Reilly, 2018).

Case studies, small sample trials, and systematic reviews evaluating CBT for adults with an ID have demonstrated successful outcomes in some individuals with mild-to-moderate ID with a comorbid diagnosis. These include those with anxiety (Douglass et al., 2007; Lindsay et al., 1997; Marwood & Hewitt, 2012; Willner, 2005), depression (James, 2017; McCabe, McGillivray & Newton, 2006; McGillivray, McCabe & Kershaw, 2008; Unwin et al., 2016), anger (Hamelin, Travis & Sturmey, 2013; Nicoll et al., 2013; Taylor, Novaco, Gillmer, Robertson & Thorne, 2005), post-traumatic stress disorder (Carrigan & Allez; 2017; Kroese et al., 2016), obsessive compulsive disorder (Willner & Goodey, 2006), and psychosis (Kirkland, 2005; Lindsay, 1999; Willner, 2005, 2009). Recently, a small number of between-group comparison studies and randomized controlled trials have also supported the effectiveness of CBT for individuals with mild-to-moderate ID who experience issues with anger (Willner et al., 2013), anxiety (Cooney, Jackman, Coyle & O'Reily, 2017), and/or depression (Hartley et al., 2015; Hassiotis et al., 2013; Lindsay et al., 2015; McGillivray & Kershaw, 2015). New research also suggests that computerized cognitive behavioral therapy is an engaging approach to increase the accessibility of effective CBT interventions for individuals with ID (Cooney et al., 2017; Cooney, Jackman, Tunney, Coyle & O'Reily, 2018).

Lindsay's (1999) work with adults with an ID demonstrated that the core techniques used in CBT, such as agenda setting and homework assignments, could be implemented with adults with an ID if clinicians made modifications in their delivery of treatment. Stenfert Kroese et al. (1998) stated that modifications to CBT for adults with an ID may also include facilitating valid self-reports and accommodating the client's level of comprehension and expression of abstract concepts. However, published studies typically do not provided details on how CBT was implemented (Prout, Chard, Nowak-Drabik, & Johnson, 2000; Prout & Nowak-Drabik, 2003), so it remains unclear to what extent modifications can be made while also maintaining the integrity of the original psychotherapeutic model (Flynn, 2012) and which elements of treatment are integral to treatment success (Whitehouse et al., 2006; Willner, 2005). Cooney, Tunney, et al. (2018) conducted a systematic review of 18 studies evaluating CBT skills for adults with an ID, including emotion regulation; discriminating between thoughts, feelings, and behaviors; connecting events to emotions; and cognitive mediation (i.e., recognizing the mediating role of a thought in the thought-feeling-behavior relationship). A description of their findings regarding the appropriateness and value of each of these skills for adults with an ID is provided below.

Emotion Regulation

Adults with an ID were more effective in identifying emotions if they had a larger receptive language vocabulary and were able to point to a visual depiction of an emotion rather than having to verbally describe it (Cooney, Tunney, et al., 2018; Willner, 2006). Cooney, Tunney, et al. (2018) recommend that clinicians use photographs of faces expressing emotions with context, rather than photographs without context or line drawings, to guide emotion recognition by adults with an ID (especially those with moderate cognitive impairment).

Discriminating Between Thoughts, Feelings, and Behaviors

Willner (2006) found that adults with an ID were able to identify their feelings and behaviors but struggled with identifying their thoughts in isolation, even with training. A study by Vereenooghe, Gega, Reynolds, and Langdon (2016) showed that although adults' ability to identify thoughts within the context of a thought-feeling-behavior relationship increased with training, this improvement did not translate to situations where they were asked to identify thoughts in isolation.

Connecting Events to Emotions

Willner (2006) found that adults with ID were able to make a connection between events and emotions. Moreover, as an individual's level of expressive language ability increased, so did their ability to make the connection between events and experienced emotions.

Cognitive Mediation

Cognitive mediation is the ability to identify the mediating role of a thought in a thought-feeling-behavior relationship (Beck, 1976). Cooney, Tunney, et al.'s (2018) systematic review showed that adult participants with an ID were better able to understand and engage in cognitive mediation when presented with a congruent scenario (i.e., situations, feelings, and behaviors were all positive or all negative) relative to an incongruent scenario, where a negative situation was paired with a positive emotion or belief or vice versa. Cognitive mediation ability in adults with an ID was also boosted when the stimulus was presented in pictures rather than verbally alone. A study by Richardson et al. (2016) also suggested that individuals with an ID may be better able to identify mediating cognitions in personally relevant situations, relevant to arbitrary scenarios.

Cognitive Behavior Therapy for Children with an ID

Much of the research evaluating CBT for children with ID has focused on the treatment of fears and anxiety. For example, early research (Obler & Terwilliger, 1970) used a modified version of systematic desensitization to address specific phobias (e.g., fear of using a public bus or fear of the sight of a live dog) in 15 “neurologically impaired” children aged 7–12. The authors noted that their modified version of systematic desensitization differed in that there was no training in relaxation provided to the participants, participants were not required to create a fear hierarchy, and the participants were not expected to verbally indicate their anxiety level during exposure sessions. Specifically, treatment involved having the participants look at pictures of the fear-inducing stimulus instead of using imaginal exposure paired with progressive muscle relaxation. Once it was determined that the participants could tolerate looking at pictures of the stimulus, they were exposed in vivo. Instead of the participants using relaxation methods while viewing the fear-inducing stimulus, the therapist would act as a buffer between the participant and the stimulus and would praise the participant for successfully tolerating the exposure. High-potency reinforcement (toys, candy) were provided to the participants for eventually tolerating the stimulus without the therapist present. All children with IQs 75 or lower, as measured by the Wechsler Intelligence Scale for Children, were at a minimum able to tolerate the feared stimulus in the presence of another individual, with some participants able to independently tolerate the stimulus by the end of treatment. This early study demonstrated methods for modifying systematic desensitization for use with children with ID.

Davis, Kurtz, Gardner, and Carman (2007) also examined the utility of CBT to treat specific phobia in children with ID through a case study. This case study examined the effectiveness of a variant of CBT, one-session treatment (OST; Öst, 1989), to address the specific phobias (i.e., fear of water and heights) of a 7-year-old verbal child with “mild cognitive delays.” OST was delivered without adaptations and consisted of a 3-h session of graduated, in vivo exposure. The OST incorporated participant modeling, verbal and physical praise for approaching the feared stimulus, psychoeducation, and cognitive challenging. The child was never forced to approach the feared stimulus; however, the child and therapist negotiated how to move up the steps in his fear hierarchy. By the end of treatment for each feared stimulus, the child was able to complete the last steps in his fear hierarchies. At follow-up assessments, the child no longer met diagnostic criteria for either specific phobia. This research presented preliminary evidence for the use of one type of CBT, OST, to treat specific phobias in children with ID. However, since the participant was verbal and had “mild cognitive delays,” it is unclear what the effectiveness of this treatment is for nonverbal, more cognitively impaired children.

Another variant of CBT, cognitive behavioral play therapy (CBPT) has been examined for its ability to improve self-esteem in children with an ID (Bana, Sajedi, Mirzale, & Rezasoltani, 2017). Specifically, Bana et al. (2017) compared children aged 8–12 with IQs between 50 and 70 who received CBPT to a control group that was age- and IQ-matched. CBPT consisted of 12 1-h sessions administered in groups of five children each. During each CBPT session, children played with toys and participated in group games targeting predetermined objectives. Each session had different objectives such as identifying feelings, communicating with body language, focusing on children’s personal strengths, increasing self-esteem, and improving communication and cooperation among children. For example, in one session, children played with Lego blocks to increase their imagination abilities. Following treatment, comparisons between the treatment and control groups revealed that children who received the CBPT made significant improvements on a self-report measure of their self-esteem. The authors noted that previous research (Baggerly, 2004) has shown that improvements in self-esteem after play therapy also led to reduced anxiety and depression in a population of homeless children, although no information was given regarding the intellectual functioning of these children. Therefore, conclusions cannot be drawn regarding the effectiveness of CBPT as a mood enhancement intervention for children with ID.

Other researchers have focused on adapting manualized CBT programs for children, such as the Coping Cat program (Kendall & Hedtke, 2006). This program is recommended for anxious youth aged between 7 and 13 years with an IQ of at least 80, although it has been noted that the treatment can be modified for children with ID (Beidas, Benjamin, Puleo, Edmunds & Kendall, 2010). An example of this is illustrated in a 2006 case study by Suveg, Comer, Furr, and Kendall (2006). The authors described the treatment of an 8-year-old female with cognitive abilities in the borderline range (Full Scale IQ = 78) and learning difficulties in most academic areas. Initial assessment indicated that she met criteria for three anxiety diagnoses, social phobia, generalized anxiety disorder, and selective mutism. Her treatment plan which was based on the Coping Cat program (Kendall & Hedtke, 2006) was individualized to ensure that CBT methods were compatible with her abilities. Additionally, treatment had a focus on physical movement, such as dancing or active games, and placed less emphasis on metacognitive content. Further, increased repetition and drawing activities helped the patient understand and retain concepts. Some concepts were emphasized, such as understanding the physical symptoms of anxiety, and some were deemphasized, such as identifying maladaptive cognitions. The patient participated in 20 sessions of this modified CBT treatment and showed significant improvement across multiple domains of anxiety posttreatment, including no longer meeting criteria for generalized anxiety disorder or selective mutism. Although only a case study, this publication is important as it describes the specific modifications made to a widely accessible manualized treatment that clinicians can use with their own clients.

Much of the recent research examining the effectiveness of CBT for children with ID has focused on children with comorbid autism spectrum disorder (ASD). Similar to the ID literature on children without ASD, researchers have used strategies such as systematic desensitization to address phobias in children with ASD and ID. For example, Luscre and Center (1996) used systematic desensitization paired with video modeling and reinforcement to decrease fear of the dentist in three children aged six to nine who were functioning at the "severely mentally retarded level." Another study (Ricciardi, Luiselli, & Camare, 2006) used contact desensitization procedures, which involved shaping of approach responses, to gradually expose an 8-year-old child with ASD and cognitive delays to his feared stimulus (i.e., electronic animated toys). Both of these studies provide additional evidence for the use of graduated exposure-based interventions to address phobias in children with ID. However, they, like so many studies in the ID literature, fail to provide a clear indication of the IQs and functional abilities of the participant sample.

A case study by Mullins and Christian (2001) utilized progressive muscle relation to reduce disruptive behaviors in a 12-year-old boy with ASD and "mild mental retardation." The authors used adapted progressive muscle relaxation procedures to create a training book for the child to use during sessions. The therapist provided a series of most to least intrusive prompts to encourage the child to practice the steps of the relaxation procedures in the book. After 11 sessions, the child was able to perform the steps of progressive muscle relaxation independently with only the use of the book as a guide. After this training phase, the child was cued to complete the progressive muscle relaxation prior to engaging in a leisure activity. The child's disruptive behavior (i.e., loud screams, other vocal sounds, repetitive statements, hand and toe chewing, nose picking, and finger picking) was monitored during the leisure activities, and results indicated that the child engaged in less disruptive behavior during leisure activities posttreatment as compared to pretreatment. However, the child's disruptive behavior was not completely eliminated, and the child still had to be cued to engage in the progressive muscle relaxation, limiting its generalizability to other settings when the child was not cued to use this strategy.

Moskowitz et al. (2017) described a combination of CBT and applied behavior analysis/positive behavior supports (ABA/PBS) therapy to treat anxiety in three children aged six to nine with adaptive or intellectual abilities in the "low" range. Each child's treatment package was individualized based on the parent's report of which strategies would best fit their child. The CBT components used were graduated exposure, psychoeducation, and cognitive restructuring, while the ABA/PBS strategies used were increasing predictability, visual schedules, social stories, priming/video priming, providing choices, reinforcement/counterconditioning, and incorporating perseverative interests. Additionally, consequence-based strategies used were positive reinforcement contingent on approach, differential reinforcement of alternative behavior, and escape extinction. Following the application of the intervention procedures, all three children had significant reductions in anxious behaviors, as coded by blind raters.

Similarly, Parent, Birtwell, Lambright, and DuBard (2016) combined CBT and behavioral techniques to address verbal and physical aggression in a 12-year-old boy with ASD who had cognitive abilities at the first percentile. CBT strategies taught included taking deep breaths, reaching out to a familiar adult for help, cognitive restructuring for maladaptive thoughts, identifying triggers for aggression, and asking for a break when upset. These strategies were taught using behavior analytic principles, such as creating a task analysis for each technique and teaching smaller steps until they were mastered. Additionally, visual materials were used to help teach these techniques. The child participant's rate of physical and verbal aggression significantly reduced following intervention. Additionally, at post-intervention, he was using coping skills more than 80% of the time without prompting.

Gobrial and Raghavan (2017) examined the effectiveness of a parent-delivered anxiety management program, the Calm Child Programme (Gobrial & Raghavan, 2017), for seven 5- to 14-year-old children with a diagnosis of ASD and mild-to-moderate ID. The program featured a manual that taught parents to use proactive strategies (e.g., visual schedules, talking and explaining, relaxation, and physical activities) to prevent their child's anxiety, communication strategies (e.g., anxiety thermometer) to help their child to self-regulate, and reactive strategies (e.g., distraction, quiet time, having fun, comfort strategies) to manage their child's anxiety. Children's anxiety scores from pre- to posttreatment significantly decreased based on a parent report questionnaire. However, as parents were both the program evaluators and deliverers, rater bias may have influenced these results.

Collectively, the findings from the studies described above suggest that CBT holds promise for children with ID (and comorbid ASD), especially when used to treat anxiety and when supplemented with other behavioral supports and teaching strategies. However, large-scale controlled studies are needed to evaluate the effectiveness of CBT for upskilling children with ID in how to cope with a range of emotions, beyond anxiety.

Cognitive Behavior Therapy for Adolescents with an ID

Research suggests that adolescents with mild-to-moderate ID have the potential to greatly benefit from CBT when appropriate adaptations are made (Hronis, Roberts, & Kneebone, 2017). Teens with mild-to-moderate ID often have more severe depression, anxiety, and antisocial behavior than teens with more profound ID (Einfeld & Tonge, 1996), which in part may be due to a greater awareness of the challenges that they face (White et al., 2010). However, this same insight can help them to optimally engage in and benefit from CBT. For example, Singh et al. (2013) demonstrated the effectiveness of 20 sessions of exposure with response prevention over 4 weeks in reducing the OCD symptoms of an 18-year-old man with mild intellectual disability. A differential

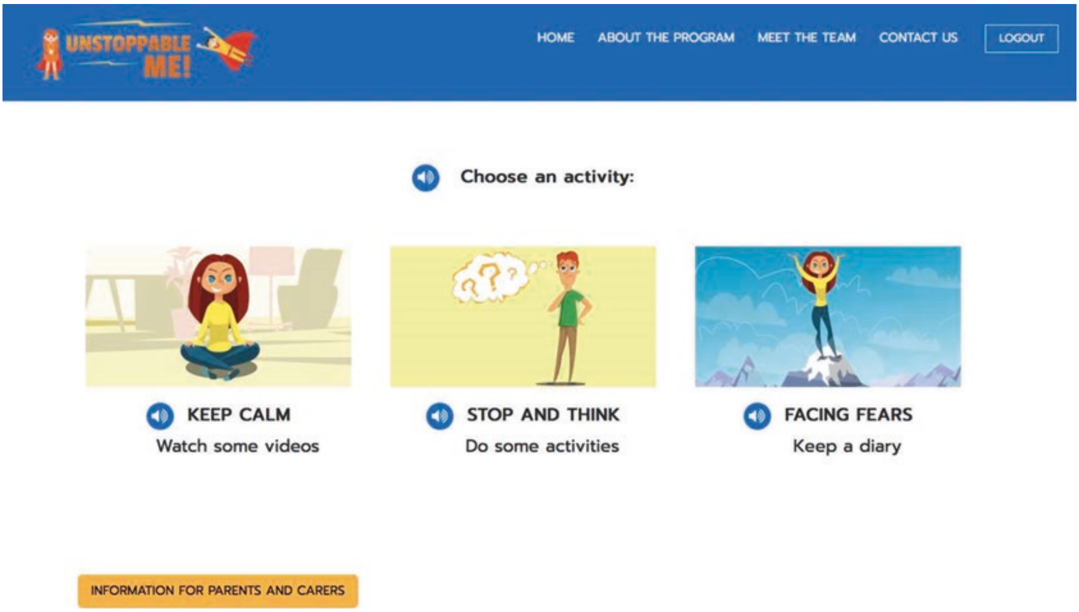


Fig. 38.1 Screenshot from Hronis et al.'s (2018) Fearless Me! online anxiety treatment program for youth. Reproduced with authors' permission

reinforcement system was used to motivate the patient to engage in therapeutic tasks, and his mother was trained as a co-therapist. Treatment gains were maintained at 1-year follow-up. However, as the patient was concurrently taking fluoxetine and clomipramine, the unique contribution that therapy made to his improved functioning could not be determined. Hronis, Roberts, Roberts, and Kneebone (2018) have also developed an online CBT anxiety treatment program (Fearless Me!) for 8- to 18-year-olds with mild-to-moderate ID (www.unstoppableme.com.au), which was being evaluated at the time of this publication. The intervention teaches relaxation strategies, helpful thinking approaches, and the importance of exposure in overcoming fears, as shown in the screenshot illustrated in Fig. 38.1.

Conclusions Regarding Cognitive Behavioral Therapy for Individuals with an ID

While there is a growing evidence base supporting the effectiveness of CBT for children, adolescents, and adults with an ID, the majority of studies have been uncontrolled and performed with small sample sizes and lack follow-up data examining the durability of treatment gains. Furthermore, it is not clear which ingredients in CBT treatment packages are the most powerful in producing positive change: the behavioral strategies, cognitive techniques, or both (Sturmey, 2006). More recently, empirical attention has turned to a third generation of behavioral therapies for individuals with ID: mindfulness-based approaches.

Mindfulness-Based Interventions for Individuals with an ID

Acceptance and commitment therapy (ACT) is a behavioral therapy model that focuses on changing one's relationship with the meaning of an event through psychotherapeutic strategies such as mindfulness, experiential engagement, and cognitive defusion (Hayes, Luoma, Bond, Masuda, & Lillis, 2006; Leoni, Corti & Cavagnola, 2015). In ACT, mental health issues are perceived to stem from avoiding unwanted internal thoughts and emotions. By distancing oneself from these thoughts and reconnecting people with their values, ACT aims to help clients to live the life they want to live, rather avoiding uncomfortable or unwanted aspects of it (Leoni et al., 2015). In one of the first studies examining ACT with an individual with an ID, Brown and Hooper (2009) found that CBT was not appropriate for an 18-year-old female participant with moderate-to-severe ID due to her limited language skills and poor verbal reasoning abilities. However, the participant was able to engage in experiential activities to learn to recognize and separate herself from anxious and obsessive thoughts, resulting in decreased functional impairment (Brown & Hooper, 2009).

Mindfulness is one of the fundamental techniques used in ACT. It refers to an individual's ability to nonjudgmentally pay attention to their thoughts, feelings, and perceptions as they occur (Hayes et al., 2006; Leoni et al., 2015). For individuals with ID, mindfulness-based therapies offer a way of accommodating new ways of seeing problems, thereby changing the relationship they have with unhelpful thoughts about themselves, and past, present, or feared future experiences of exclusion or rejection (Idusohan-Moizer, Sawicka, Dendle & Albany, 2015). Mindfulness aims to reduce experiential avoidance by helping individuals to become aware of and normalize unpleasant emotions and body sensations without engaging in futile efforts to avoid or escape them (Idusohan-Moizer et al., 2015). In contrast to CBT, which aims to change or challenge unhelpful thoughts, mindfulness focuses on being aware of these thoughts and letting them go without getting caught up (fused) with them (Idusohan-Moizer et al., 2015). Such concepts are simplified for individuals with ID through visual metaphors and analogies such as "dropping the rope" in a "tug of war" (Hayes, Strosahl & Wilson, 1999).

Emerging literature shows some evidence supporting the effectiveness of mindfulness-based procedures for individuals with an intellectual disability. For example, Singh et al. (2014) conducted a randomized controlled trial evaluating the effectiveness of a mindfulness-based smoking cessation program with 51 participants with mild ID. The program included basic concentration meditation, setting a daily intention, mindful observation of thoughts, and Meditation on the Soles of the Feet (shifting focus from emotionally arousing thoughts to the soles of one's feet). Results showed that participants in the mindfulness condition showed a significantly greater reduction in smoking relative to those in the treatment as usual condition, with gains maintained at 1-year follow-up.

Further evidence for the effectiveness of mindfulness techniques for individuals with ID, their parents, and paid carers is provided by a systematic review conducted by Chapman et al. (2013). An analysis of 11 studies showed improvements in aggression, sexual arousal, and psychological well-being in participants with mild-to-moderate IDs following mindfulness training, decreased use of physical restraint for aggressive behavior by care staff and improved parental well-being, parent satisfaction, and parent-child interactions (Chapman et al., 2013). Participants in the reviewed studies were predominantly adults with ID, although they did include individuals from White, African-American, and Hispanic populations, suggesting that mindfulness is acceptable and effective for people from diverse cultural backgrounds. Feedback from participants suggested that optimal mindfulness training involved clear instructions, regular practice, the use of concrete examples, and role-play.

Beauchemin, Hutchins, and Patterson (2008) demonstrated that mindfulness meditation can also be valuable for adolescents with ID. Following a 5-week intervention, adolescents aged 13–18 ($n = 34$) showed a significant improvement in their state and trait anxiety, academic performance, and social skills, based on teen and parent report measures. Adolescents also reviewed the program favorably. Furthermore, a case study by Singh et al. (2008) demonstrated that the Meditation on the Soles of the Feet program helped a 17-year-old with Prader-Willi syndrome and mild ID to self-manage his hunger to prevent overeating.

Idusohan-Moizer et al. (2015) examined the effectiveness of a 9-week group mindfulness-based CBT program for 15 adults with borderline, mild, or moderate ID who had recurrent depression and/or anxiety and a history of deliberate self-harm behavior. Results showed the intervention to be effective in improving participants' experience of self-reported depression, anxiety, self-compassion, and compassion for others, with improvements maintained at 6-week follow-up. However, there was a higher dropout rate among participants with a moderate ID relative to those with borderline or mild ID, and participant motivation appeared to be influenced by carer/support worker engagement in group sessions. A unique feature of Idusohan-Moizer et al.'s (2015) mindfulness-based CBT program was a focus on compassion and kindness. Specifically, the authors acknowledged the role that shame, self-loathing, and self-criticism can play in depression and anxiety in individuals with ID and taught participants how to develop self-compassion: to "let go" of self-blame and painful memories to ease their suffering.

Heifetz and Dyson (2017) also demonstrated the potential of mindfulness-based group therapy with a sample of eight adolescents with mild ID and their parents. Results showed that an 8-week mindfulness-based group program for teens (with intermittent joint and parallel parent sessions) resulted in improvements in teens' mood and social behavior, as reported by their parents. There was also a trend for adolescents to report feeling more happy, relaxed, and less worried at the end of each mindfulness session relative to the beginning. However, some adolescents were reluctant to practice mindfulness skills outside of sessions, despite parental support (which was sometimes resisted) and the introduction of a points reward system.

Conclusions Regarding Mindfulness-Based Interventions for Individuals with an ID

Much like the CBT literature, many of the studies examining the effectiveness of mindfulness-based treatments for individuals with ID consist of reports on single case studies or small group pre-/post-treatment evaluations. Therefore, while this class of interventions holds promise, it is premature to conclude that it is appropriate and effective for individuals with ID. Furthermore, the comparative effectiveness of mindfulness-based interventions relative to more traditional CBT approaches is unknown. Future research is also needed to identify predictors of treatment outcome for both CBT and mindfulness-based programs, in addition to determining which individuals with ID respond best to individual versus group therapy.

Treatment Modifications for Individuals with an Intellectual Disability

Irrespective of the treatment approach, it is important to understand common barriers to treatment engagement for individuals with an ID and how these can be overcome. Individuals with an ID have, to varying degrees, impairments in how they acquire and process information, as well as possible challenges in episodic and/or prospective memory, such as projecting the self into past or future scenarios (Willner, 2009). Moreover, these cognitive impairments may also be associated with sensory differences, impaired attention, and poor executive functioning (Willner, 2009; Willner & Goodey, 2006). Another potential treatment barrier is impaired verbal abilities. Individuals with ID may lack experience in reporting on their emotions, giving their opinions, or making decisions (Willner, 2009). Lastly, client motivation may be a treatment barrier, which may arise from the psychological presentation, maladaptive beliefs, and cognitively challenging framework of the psychotherapeutic treatment (Willner, 2009).

With a better understanding of common barriers to treatment for individuals with an ID, clinicians are better equipped to modify therapy to better meet their clients' needs. Hurley, Tomasulo, and Pfadt (1998), together with other researchers in this field, suggest the following modifications to psychotherapeutic treatment for individuals with an ID.

Simplification

When working with individuals who have an ID, it is recommended to simplify treatment by shortening activities and sessions, segmenting the intervention into smaller regular parts over a longer period of time, and using simple language during discussion of the client's thoughts and feelings (Hernadez-Halton, Hodges, Miller, & Simpson, 2000; Reddy, 2015). It is also advised that clinicians be more directive and less collaborative to reduce the executive functioning demands placed on clients (Hronis et al., 2017).

Language

The language abilities of clients with an ID should be considered when tailoring therapy to meet their needs. It is suggested that clinicians use shorter sentences and simpler words and be more succinct in sharing their thoughts and more concise when setting the agenda at the beginning of each session (Hurley et al., 1998; Lindsay et al., 1997; Willner, 2009). It is also recommended that supplemental tools such as photographs, video clips, and illustrations are used to facilitate the client's understanding of core concepts and ideas, rather than text-dense written materials (Morin, Cobigo, Rivard & Lepine, 2010). Clinicians need to provide explanations and interpretations at a level that matches the client's level of understanding (Hwang & Kearney, 2013).

Activities and Supports

It is highly recommended that clinicians include supplemental activities and supports to promote client learning, understanding, and change (Hurley et al., 1998). These activities should include experiential learning exercises such as practicing mindfulness in session with therapist support, role-playing new skills, completing interactive worksheets that provide simple visual illustrations of skills and concepts, and capturing details of how skills were used between sessions via drawings or smartphone voice memos, photos, or videos if writing is challenging. Digital calendar reminders can also be scheduled to prompt skill usage in daily life. Supplemental activities not only deepen the client's understanding and skill in applying therapeutic concepts but also help the clinician to gauge the client's comprehension and capacity to use these techniques (Summers & Witts, 2003).

Developmental Level

It is of the upmost importance that clinicians consider the client's developmental level when presenting topics and material in session (Hurley et al., 1998). If necessary, materials such as cartoon strips (hand drawn or computer-generated), emotion masks, dolls, and games can be used to provide concrete illustrations of emotions and event sequences to patients. Caution is warranted when using materials such as these, however, as some adolescents and adults may consider them to be developmentally inappropriate.

Directive Approach

Clinicians working with individuals with an ID often need to take a more directive than collaborative approach (Hronis et al., 2017). A directive approach typically includes outlining the goals of therapy, discussing treatment progress, and providing supplemental materials to guide sessions (Hurley et al., 1998; Lindsay et al., 1997). Moreover, Stavrakaki and Klein (1986) recommend that clinicians use more direct strategies, such as suggestion, persuasion, and reassurance, when working with individuals with an ID relative to typically developing clients.

Flexible Methods

Clinicians working with clients with an ID are encouraged to be flexible in their delivery of treatment, including flexibility in the structure and length of sessions and treatment, incorporating techniques from different treatment modalities, and a willingness to frequently repeat core concepts and ideas (Drury & Alim, 2014; Leoni et al., 2015). Being flexible and creative in the delivery of treatment allows therapy to be aligned with a client's level of cognitive and language functioning. Research conducted by Whitehouse et al. (2006) showed that flexibility was the most common treatment modification used by clinicians when delivering CBT to clients with ID.

Involve Caregivers

Caregiver involvement often supports treatment progress for individuals with an ID (Willner, 2006, 2009). Caregivers can function as co-therapists, prompting, modeling, and/or rewarding patients' use of emotion regulation skills when needed in daily life (Heifetz & Dyson, 2017; Hurley et al., 1998). While care must be taken to give clients the opportunity to discuss personal information with the therapist in private, caregivers also serve a vital role in providing therapists with feedback on patients' progress.

Transference/Countertransference

Hurley et al. (1998) argue that it is particularly important for clinicians working with clients who have an ID to establish firm boundaries and have consistent peer supervision, as stronger attachments often quickly develop with this clientele relative to typically developing patients. Clinicians also need to be cognizant of negative countertransference feelings that may develop from the client's sensitivity to nonverbal communication due to their impaired language abilities (Hernadez-Halton et al., 2000).

Disability/Rehabilitation Approaches

The disability experienced by clients with ID needs to be sensitively explored in therapy (Hurley et al., 1998). This includes the stigma that is oftentimes associated with having an ID (Haddock, Lobban, Hatton & Carson, 2004). It is recommended that clinicians help clients to accept limitations that cannot be changed and to focus on working toward goals that are important and achievable (Idusohan-Moizer et al., 2015).

Case Example

T was a 13-year-old Hispanic boy with mild intellectual impairment (Full Scale IQ = 71) who lived in a shelter with his mother and attended a school for special needs students. He presented to therapy with a diagnosis of oppositional defiant disorder. T was frequently irritable and angry at home and at school and refused to follow his mother's instructions to tidy his things and complete his homework. He also became frustrated in response to his teacher's requests to complete schoolwork at school. T would often complain about schoolwork being "boring" and "stupid." T's mother indicated that his oppositionality was causing significant distress to herself and other shelter residents. T's teacher believed that his noncompliance was contributing to him underachieving at school and being disliked by his peers.

T's mother and teacher reported that they had tried to use a points chart in the past to encourage T to follow instructions at home and at school. T's mother described this as being largely unsuccessful at home, as she did not have the financial resources to provide T with rewards that he was motivated to earn. T's teacher described some initial success with the points chart; however, she indicated that T quickly lost interest in it.

Individual therapy with T initially focused on rapport building. T was angry that his mother had forced him to come to therapy, insisting that there was "nothing wrong" with him. During the first four therapy sessions, the clinician informally chatted with T and enquired about his interests (basketball, soccer). During these conversations, the therapist took turns shooting hoops with T using a mini basketball set and kicked a mini soccer ball with him. She found this informal approach of talking with T while doing another activity helped him to relax and open up.

In Session 5, the therapist started exploring with T what life at home and at school was like. T indicated that he often didn't understand what his mom and teacher were asking him, which annoyed him. T and the therapist took a photo of T with the therapist's iPad and digitally drew body clues on the photo to illustrate what "annoyed" felt like for T (tightness in his stomach, feeling energetic, thinking "What are they talking about?", shouting, and swearing). The therapist printed this out and, with T's help and permission, discussed the picture with T's mother at the end of session. The therapist asked T and his mom to be on high alert for when these body clues occurred during the week and to make a voice memo on the mom's smartphone of what was happening when they spotted them.

At the beginning of the next session, T and his mom shared three voice memos with the therapist: one made by T's mom and two by T. Two of the voice memos described T's frustration when trying to do homework, and one related to him not getting ready to go to school when asked. To help address the homework issue, the therapist asked T for permission to contact his teacher to request him to complete homework after school at homework club with school staff support. T reluctantly agreed. The therapist also told T that she would talk to the teacher about simplifying his schoolwork and homework to make it easier for him to understand. The therapist spoke to both the teacher and T's mom about giving simple instructions to T, one step at a time. The teacher also agreed to provide T with handouts showing worked examples of concepts taught in class, rather than verbally describing them or writing them up on the whiteboard for him to copy.

T's mother agreed to set up a visual schedule for T at home with photos illustrating the daily tasks he needed to complete (e.g., getting dressed, brushing his teeth, etc.). A simple monitoring chart was set up at school, whereby if T tried his best to complete activities at school without complaining during three periods (morning, middle, afternoon), he was awarded 5-min increments of iPad time at the end of the school day. A response cost system was set up at home, whereby 5 min was deducted from T's 30 min per day iPad play time for each daily task that he did not complete (eating breakfast and dinner at the table, showering, dressing for the day and bed). T initially argued with his mother when this system was implemented. However, his mother was able to remove herself from him at these times and consistently use the response cost system. Over a period of 2 weeks, she started to see an improvement in his compliance with the daily routine tasks.

Over subsequent sessions, T reported that while things were "a bit better" at school, he still did not want to do schoolwork and found it annoying when he did. T indicated that he did not like asking for help when he didn't understand something, as the other kids would think that he was "stupid and a loser." As T and the therapist were shooting hoops in session, the therapist asked him how he was likely to perform if he thought "I'm stupid and a loser." He said he didn't know, so she suggested they test it out. She asked him to say "You can't do this. You're a loser at B-Ball" as she shot hoops. She didn't make any shots and showed an exaggerated disappointed face and body posture. The therapist asked T to identify how she was feeling. He laughed and replied, "Bad. Like a loser." She then asked him if she was likely to want to keep playing basketball when she had these thoughts, and he said no. Next, the therapist asked him what he could say to help her feel better and keep trying, and he said, "You've got this. Just practice." She said these helpful statements to herself (T joined in) and landed two of the next five baskets. She asked T what he learned from this activity, and he said, "You did better when I cheered you on." The therapist asked T if he could cheer himself on. She repeated the basketball activity, this time with T shooting hoops and initially saying unhelpful statements to himself and then being his own cheerleader. At the end of the activity, she asked him what he learned. He responded, "I feel better when I say nice things to myself." The therapist invited T's mother into the room at the end of the session and asked her to film him shooting hoops and being his own cheerleader with her smartphone. She asked both mother and son to review the video each day before school to remind T that he can also be his own cheerleader when doing difficult tasks at school.

The therapist reached out to T's teacher and asked if it would be possible for T to use an inconspicuous code word or card to signal when he needed help in class, so as not to draw peers' attention to him. The teacher agreed that if T was getting frustrated in class he could ask to go and get a drink (a maximum of five times per day), which signaled to her and the teaching assistant to offer him help. This was explained to T in the next therapy session, and he reluctantly agreed to give it a go. In this and subsequent sessions, T was asked to bring in schoolwork to session, and his mother filmed him saying helpful thoughts (e.g., "I'll give it a go. I'll get better with practice, like B-ball."), while he attempted it. T also practiced asking for a drink of water when he didn't understand something and walking to the other side of the therapy room and having a drink from his water bottle. T was initially encouraged to focus on the sensations in his body while walking as a mindfulness activity. However, this repeatedly made him agitated, so he was encouraged to squeeze a basketball-shaped stress ball in his pocket instead and to be receptive to help with his schoolwork when he returned to the desk. T was rewarded with NBA collector cards for calmly practicing this process over eight therapy sessions.

With his mother's support, T reviewed the filmed footage from therapy each morning before school, and over a period of 2 months, both his mother and teacher reported a marked improvement in his willingness to try his best at schoolwork and to ask for help. By the end of therapy, T's mother also reported that T completed his daily routine tasks with minimal prompting and that they were fighting a lot less at home. This case example illustrates how using a combination of environmental modifications, a behavioral reinforcement system, enlisting carer and teacher support, and promoting client skill building by taking therapy at a slower pace, using action-based learning, illustrating concepts with personally relevant examples, and consolidating skills through repeated roleplays, practice, and creating and reviewing video clips reduced the oppositional behavior and irritability of an adolescent client with mild ID.

Conclusion

CBT and mindfulness-based interventions appear to hold promise in improving the mental health and well-being of individuals with mild-to-moderate ID across the life span. However, as most of the research to date has involved single case study or uncontrolled evaluations with small sample sizes, large-scale randomized controlled trials are needed to conclude which interventions are most effective, for whom and in what format, and dosage. Future research is also needed to identify the active ingredients of different therapeutic approaches and what procedural modifications are optimal for different clinical populations who have an ID. Given the cognitive limitations of individuals with an ID, empirical attention should also be focused on the durability of treatment gains over time and the transferability of skills across settings. The dissemination of this knowledge to clinicians will help to improve their confidence and competence in working with clients who have an ID, enhancing service accessibility for this often-neglected sector of the population. The potential benefits of such efforts are far-reaching, including improved well-being, independence, and community and workforce participation for individuals with ID.

References

Arthur, A. R. (2003). The emotional lives of people with intellectual disabilities. *British Journal of Learning Disabilities, 31*, 25–30.

Baggerly, J. (2004). The effects of child-centered group play therapy on self-concept, depression, and anxiety of children who are homeless. *International Journal of Play Therapy, 13*(2), 31.

Bana, S., Sajedi, F., Mirzaie, H., & Rezasoltani, P. (2017). The efficacy of cognitive behavioral play therapy on self esteem of children with intellectual disability. *Iranian Rehabilitation Journal, 15*(3), 235–242.

Beauchemin, J., Hutchins, T. L., & Patterson, F. (2008). Mindfulness meditation may lessen anxiety, promote social skills, and improve academic performance among adolescents with learning disabilities. *Complementary Health Practice Review, 13*(1), 34–45.

Beck, A. T. (1976). *Cognitive therapy and the emotional disorders*. New York, NY: International Universities Press.

Beck, J. S. (2011). *Cognitive behavior therapy: Basics and beyond*. New York, NY: Guilford Press.

Beidas, R. S., Benjamin, C. L., Puleo, C. M., Edmunds, J. M., & Kendall, P. C. (2010). Flexible applications of the coping cat program for anxious youth. *Cognitive and Behavioral Practice, 17*(2), 142–153.

Bender, M. (1993). The unoffered chair: The history of therapeutic disdain towards people with a learning disability. *Clinical Psychology Forum, 54*, 7–12.

Brown, F. J., & Hooper, S. (2009). Acceptance and Commitment Therapy (ACT) with a learning disabled young person experiencing anxious and obsessive thoughts. *Journal of Intellectual Disabilities, 13*(3), 195–201. https://doi.org/10.1177/1744629509346173

Carrigan, N., & Allez, J. (2017). Cognitive behaviour therapy for post-traumatic stress disorder in a person with an Autism Spectrum Condition and intellectual disability: A case study. *Journal of Applied Research in Intellectual Disabilities, 30*, 326–335.

Chapman, M. J., Hare, D. J., Caton, S., Donalds, D., McInnis, E., & Mitchell, D. (2013). The use of mindfulness with people with intellectual disabilities: A systematic review and narrative analysis. *Mindfulness, 4*, 179–189.

Cooney, P., Jackman, C., Coyle, D., & O'Reily, G. (2017). Computerised cognitive-behavioural therapy for adults with intellectual disability: Randomised controlled trial. *The British Journal of Psychiatry, 211*, 95–102. https://doi.org/10.1192/bjp.bp.117.198630

Cooney, P., Jackman, C., Tunney, C., Coyle, D., & O'Reily, G. (2018). Computer-assisted cognitive behavioural therapy: The experiences of adults who have an intellectual disability and anxiety or depression. *Journal of Applied Research in Intellectual Disabilities, 31*, 1–14. https://doi.org/10.1111/jar.12459

Cooney, P., Tunney, C., & O'Reilly, G. (2018). A systematic review of the evidence regarding cognitive therapy skills that assist cognitive behavioural therapy in adults who have an intellectual disability. *Journal of Applied Research in Intellectual Disabilities, 31*(1), 23–42. https://doi.org/10.1111/jar.12365

Cooper, J. O., Heron, T. E., & Heward, W. L. (2007). *Applied behavior analysis* (2nd ed.). Upper Saddle River, NJ: Pearson.

Davis, T. E., III, Kurtz, P. F., Gardner, A. W., & Carman, N. B. (2007). Cognitive-behavioral treatment for specific phobias with a child demonstrating severe problem behavior and developmental delays. *Research in Developmental Disabilities, 28*(6), 546–558.

Dodd, P., & McGinnity, M. (2003). Psychotherapy and intellectual disabilities. *Irish Journal of Psychological Medicine, 20*, 38–40.

Douglass, S., Palmer, K., & O'Connor, C. (2007). Experiences of running an anxiety management group for people with learning disability using a cognitive behavioural intervention. *British Journal of Learning Disabilities, 35*(4), 245–252. https://doi.org/10.1111/j.1468-3156.2007.00443.x

Drury, H., & Alim, N. (2014). Integrative psychotherapy in intellectual disabilities: Using cognitive-behavioural and psychodynamic formulations. *Advances in Mental Health and Intellectual Disabilities, 8*(3), 197–209. https://doi.org/10.1108/AMHID-10-2012-0001

Einfeld, S. L., & Tonge, B. J. (1996). Population prevalence of psychopathology in children and adolescents with intellectual disability: II Epidemiological findings. *Journal of Intellectual Disability Research, 40*(2), 99–109.

Flynn, A. G. (2012). Fact or faith? On the evidence for psychotherapy for adults with intellectual disability and mental health needs. *Current Opinion in Psychiatry, 25*(5), 342–347. https://doi.org/10.1097/YCO.0b013e328355e196

Gobrial, E., & Raghavan, R. (2017). Calm child programme: Parental programme for anxiety in children and young people with autism spectrum disorder and intellectual disabilities. *Journal of Intellectual Disabilities, 22*(4), 315–327. https://doi.org/10.1177/1744629517704536

Haddock, G., Lobban, F., Hatton, C., & Carson, R. (2004). Cognitive–behaviour therapy for people with psychosis and mild intellectual disabilities: A case series. *Clinical Psychology & Psychotherapy, 11*(4), 282–298. https://doi.org/10.1002/cpp.414

Hamelin, J., Travis, R., & Sturmey, P. (2013). Anger management and intellectual disabilities: A systematic review. *Journal of Mental Health Research in Intellectual Disabilities, 6*(1), 60–70. https://doi.org/10.1080/19315864.2011.637661

Hartley, S. L., Esbensen, A. J., Shalev, R., Vincent, L. B., Mihaila, I., & Bussanich, P. (2015). Cognitive behavioral therapy for depressed adults with mild intellectual disability: A pilot study. *Journal of Mental Health Research in Intellectual Disabilities, 8*(2), 72–97. https://doi.org/10.1080/19315864.2015.1033573

Hassiotis, A., Serfaty, M., Azam, K., Strydom, A., Blizard, R., Romeo, R., . . . King, M. (2013). Manualised individual cognitive behavioural therapy for mood disorders in people with mild to moderate intellectual disability: A feasibility randomized controlled trial. *Journal of Affective Disorders, 151*, 186–195.

Hayes, S. C., Luoma, J. B., Bond, F. W., Masuda, A., & Lillis, J. (2006). Acceptance and commitment therapy: Model, processes and outcomes. *Behaviour Research and Therapy, 44*(1), 1–25. https://doi.org/10.1016/j.brat.2005.06.006

Hayes, S. C., Strosahl, K., & Wilson, K. G. (1999). *Acceptance and commitment therapy: An experiential approach to behavior change*. New York, NY: Guilford Press.

Heifetz, M., & Dyson, A. (2017). Mindfulness-based group for teens with developmental disabilities and their parents: A pilot study. *Mindfulness, 8*(2), 444–453.

Hemmings, C., Deb, S., Chaplin, E., Hardy, S., & Mukherjee, R. (2013). Review of research for people with ID and mental health problems: A view from the United Kingdom. *Journal of Mental Health Research in Intellectual Disabilities, 6*(2), 127–158. https://doi.org/10.1080/19315864.2012.708100

Hernadez-Halton, I., Hodges, S., Miller, L., & Simpson, D. (2000). A psychotherapy service for children, adolescents and adults with learning disabilities at the Tavistock Clinic, London, UK. *British Journal of Learning Disabilities, 28*(3), 120–124. https://doi.org/10.1046/j.1468-3156.2000.00064.x

Hronis, A., Roberts, L., & Kneebone, I. I. (2017). A review of cognitive impairments in children with intellectual disabilities: Implications for cognitive behaviour therapy. *British Journal of Clinical Psychology, 56*(2), 189–207.

Hronis, A., Roberts, R., Roberts, L., & Kneebone, I. I. (2018). *Fearless Me!© Cognitive behaviour therapy for children with intellectual disability and anxiety*. Sydney, Australia: University of Technology.

Hurley, A. D., Tomasulo, D. J., & Pfadt, A. G. (1998). Individual and group psychotherapy approaches for persons with mental retardation and developmental disabilities. *Journal of Developmental and Physical Disabilities, 10*(4), 365–386. https://doi.org/10.1023/A:1021806605662

Hwang, Y. S., & Kearney, P. (2013). A systematic review of mindfulness intervention for individuals with developmental disabilities: Long-term practice and long lasting effects. *Research in Developmental Disabilities, 34*, 314–326.

Idusohan-Moizer, H., Sawicka, A., Dendle, J., & Albany, M. (2015). Mindfulness-based cognitive therapy for adults with intellectual disabilities: An evaluation of the effectiveness of mindfulness in reducing symptoms of depression and anxiety. *Journal of Intellectual Disability Research, 59*(2), 93–104.

James, J. S. (2017). Cognitive-behavioral therapy for depression in individuals with intellectual disabilities: A review. *Journal of Mental Health Research in Intellectual Disabilities, 10*(1), 17–29. https://doi.org/10.1080/19315864.2016.1271485

Jones, R. S. P., Miller, B., Williams, H., & Goldthorpe, J. (1997). A radical behavioural perspective. In B. Stenfert Kroese, D. Dagnan, & K. Loumidis (Eds.), *Cognitive behaviour therapy for people with learning disabilities* (pp. 16–32). London: Routledge.

Kendall, P. C., & Hedtke, K. A. (2006). *Cognitive-behavioral therapy for anxious children: Therapist manual*. Ardmore, PA: Workbook Publishing.

Kirkland, J. (2005). Cognitive-behaviour formulation for three men with learning disability who experience psychosis: How do we make it make sense? *British Journal of Learning Disability, 33*, 160–165.

Kroese, B. S., Willott, S., Taylor, F., Smith, P., Graham, R., Rutter, T., … Wilner, P. (2016). Trauma-focused cognitive-behaviour therapy for people with mild intellectual disabilities: Outcomes of a pilot study. *Advances in Mental Health and Intellectual Disabilities, 10*(5), 299–310. https://doi.org/10.1108/AMHID-05-2016-0008

Leoni, M., Corti, S., & Cavagnola, R. (2015). Third generation behavioural therapy for neurodevelopmental disorders: Review and trajectories. *Advances in Mental Health and Intellectual Disabilities, 9*(5), 265–274. https://doi.org/10.1108/AMHID-06-2015-0031

Lindsay, W. R. (1999). Cognitive therapy. *The Psychologist, 12*, 238–241.

Lindsay, W. R., Neilson, C., & Lawrenson, H. (1997). Cognitive-behaviour therapy for anxiety for people with learning disabilities. In B. Stenfert Kroese, D. Dagnan, & K. Loumidis (Eds.), *Cognitive behaviour therapy for people with learning disabilities* (pp. 124–140). London: Routledge.

Lindsay, W. R., Tinsley, S., Beail, N., Hastings, R. P., Jahoda, A., Taylor, J. L., & Hatton, C. (2015). A preliminary controlled trial of a transdiagnostic programme for cognitive behaviour therapy with adults with intellectual disability. *Journal of Intellectual Disability Research, 59*(4), 360–369. https://doi.org/10.1111/jir.12145

Lunksky, Y., Lin, E., Balogh, R., Klein-Geltink, J., Wilton, A. S., & Kurdyak, P. (2012). Emergency department visits and use of outpatient physician services by adults with developmental disability and psychiatric disorder. *Canadian Journal of Psychiatry, 57*, 601–607.

Luscre, D. M., & Center, D. B. (1996). Procedures for reducing dental fear in children with autism. *Journal of Autism and Developmental Disorders, 26*(5), 547–556.

Lynch, C. (2004). Psychotherapy for persons with mental retardation. *Mental Retardation, 42*, 399–405.

Marwood, H., & Hewitt, O. (2012). Evaluating an anxiety group for people with ID using mixed methodology. *British Journal of Learning Disabilities, 41*(2), 150–158. https://doi.org/10.1111/j.1468-3156.2012.00749.x

McCabe, M. P., McGillivray, J. A., & Newton, D. C. (2006). Effectiveness of treatment programmes for depression among adults with mild/moderate intellectual disability. *Journal of Intellectual Disability Research, 50*, 239–247.

McGillivray, J. A., & Kershaw, M. (2015). Do we need both cognitive and behavioural components in interventions for depressed mood in people with mild intellectual disability? *Journal of Intellectual Disability Research, 59*(2), 105–115. https://doi.org/10.1111/jir.12110

McGillivray, J. A., McCabe, M. P., & Kershaw, M. M. (2008). Depression in people with intellectual disability: An evaluation of a staff-administered treatment program. *Research in Developmental Disabilities, 29*, 524–536.

Morin, D., Cobigo, V., Rivard, M., & Lepine, M. (2010). Intellectual disabilities and depression: How to adapt psychological assessment and intervention. *Canadian Psychology, 51*(3), 185–193.

Moskowitz, L. J., Walsh, C. E., Mulder, E., McLaughlin, D. M., Hajcak, G., Carr, E. G., & Zarcone, J. R.

(2017). Intervention for anxiety and problem behavior in children with autism spectrum disorder and intellectual disability. *Journal of Autism and Developmental Disorders, 47*(12), 3930–3948.

Mullins, J. L., & Christian, L. (2001). The effects of progressive relaxation training on the disruptive behavior of a boy with autism. *Research in Developmental Disabilities, 22*(6), 449–462.

Nicoll, M., Beail, N., & Saxon, D. (2013). Cognitive behavioural treatment for anger in adults with intellectual disabilities: A systematic review and meta-analysis. *Journal of Applied Research in Intellectual Disabilities, 26*, 47–62.

Obler, M., & Terwilliger, R. F. (1970). Pilot study on the effectiveness of systematic desensitization with neurologically impaired children with phobic disorders. *Journal of Consulting and Clinical Psychology, 34*(3), 314.

Öst, L. G. (1989). One-session treatment for specific phobias. *Behaviour Research and Therapy, 27*(1), 1–7.

Parent, V., Birtwell, K. B., Lambright, N., & DuBard, M. (2016). Combining CBT and behavior-analytic approaches to target severe emotion dysregulation in verbal youth with ASD and ID. *Journal of Mental Health Research in Intellectual Disabilities, 9*(1-2), 60–82.

Prout, H. T., Chard, K. M., Nowak-Drabik, K. M., & Johnson, D. M. (2000). Determining the effectiveness of psychotherapy with persons with intellectual disabilities: The need to move towards empirically based research. *NADD Bulletin, 6*, 83–86.

Prout, H. T., & Nowak-Drabik, K. M. (2003). Psychotherapy with persons who have mental retardation: An evaluation of effectiveness. *American Journal of Mental Retardation, 108*(2), 82–93. https://doi.org/10.1352/0895-8017(2003)108<0082:PWPWHM>2.0.CO;2

Reddy, V. (2015). Inpatient group therapeutic interventions for patients with intellectual disabilities. *Journal of Intellectual Disabilities, 19*(1), 51–57.

Ricciardi, J. N., Luiselli, J. K., & Camare, M. (2006). Shaping approach responses as intervention for specific phobia in a child with autism. *Journal of Applied Behavior Analysis, 39*(4), 445–448.

Richardson, C., Killeen, S., Jahoda, A., Christopher, R., Rose, J., & Willner, P. (2016). Assessment of anger-related cognitions of people with intellectual disabilities. *Behavioural and Cognitive Psychotherapy, 44*(5), 1–21. https://doi.org/10.1017/s1352465816000059

Singh, A., Satheesan, S., Nagpal, A., Nitturkar, A., Harsha Shetty, P., Nanjundaswamy, M., … Bada Math, S. (2013). Behavioural therapy in persons with intellectual disability and OCD: An effective augmentation strategy. *Australian and New Zealand Journal of Psychiatry, 47*(9), 883–884. https://doi.org/10.1177/0004867413479067

Singh, N. N., Lancioni, G. E., Myers, R. E., Karazsia, B. T., Winton, A. S. W., & Singh, J. (2014). A randomized controlled trial of a mindfulness-based smoking cessation program for individuals with mild intellectual disability. *International Journal of Mental Health and Addiction, 12*(2), 153–168. https://doi.org/10.1007/s11469-013-9471-0

Singh, N. N., Lancioni, G. E., Singh, A. N., Winton, A. S., Singh, J., McAleavey, K. M., & Adkins, A. D. (2008). A mindfulness-based health wellness program for an adolescent with Prader-Willi syndrome. *Behavior Modification, 32*(2), 167–181.

Smiley, E. (2005). Epidemiology of mental health problems in adults with learning disability: An update. *Advances in Psychiatric Treatment, 11*, 214–222.

Stavrakaki, C., & Klein, J. (1986). Psychotherapies with the mentally retarded. *Psychiatric Perspectives on Intellectual Disabilities, 9*, 733–743.

Stenfert Kroese, B. (1998). Cognitive-behavioural therapy for people with learning disabilities. *Behavioural and Cognitive Psychotherapy, 26*, 315–322.

Stenfert Kroese, B., Dagnan, D., & Loumidis, K. (Eds.). (1998). *Cognitive behaviour therapy for people with learning disabilities*. London: Routledge.

Sturmey, P. (2006). On some recent claims for the efficacy of cognitive therapy for people with intellectual disabilities. *Journal of Applied Research in Intellectual Disabilities, 19*(1), 109–117.

Summers, S. J., & Witts, P. (2003). Psychological intervention for people with learning disabilities who have experienced bereavement: A case study illustration. *British Journal of Learning Disabilities, 31*(1), 37–41. https://doi.org/10.1046/j.1468-3156.2003.00196.x

Suveg, C., Comer, J. S., Furr, J. M., & Kendall, P. C. (2006). Adapting manualized CBT for a cognitively delayed child with multiple anxiety disorders. *Clinical Case Studies, 5*(6), 488–510.

Taylor, J. L., Novaco, R. W., Gillmer, B. T., Robertson, A., & Thorne, I. (2005). Individual cognitive-behavioural anger treatment for people with mild-borderline intellectual disabilities and histories of aggression: A controlled trial. *British Journal of Clinical Psychology, 44*, 367–382.

Unwin, G., Tsimopoulou, J., Stenfert Kroese, B., & Azmi, S. (2016). Effectiveness of cognitive behavioural therapy (CBT) programmes for anxiety or depression in adults with intellectual disabilities: A review of the literature. *Research in Developmental Disabilities, 51-52*, 60–75.

Vereenooghe, L., Gega, L., Reynolds, S., & Langdon, P. E. (2016). Using computers to teach people with intellectual disabilities to perform some of the tasks used within cognitive behavioural therapy: A randomised experiment. *Behaviour Research and Therapy, 76*, 13–23. https://doi.org/10.1016/j.brat.2015.11.002

Waitman, A., & Reynolds, F. (1992). Demystifying traditional approaches to counselling and psychotherapy. In A. Waitman & S. Conboy-Hill (Eds.), *Psychotherapy and mental handicap* (pp. 202–220). London: Sage Publications.

White, S. W., Albano, A. M., Johnson, C. R., Kasari, C., Ollendick, T., Klin, A., … Scahill, L. (2010). Development of a cognitive-behavioral intervention program to treat anxiety and social deficits in teens with high-functioning autism. *Clinical Child and Family Psychology Review, 13*(1), 77–90.

Whitehouse, R. M., Tudway, J. A., Look, R., & Kroese, B. S. (2006). Adapting individual psychotherapy for adults with intellectual disabilities: A comparative review of the cognitive–behavioural and psychodynamic literature. *Journal of Applied Research in Intellectual Disabilities, 19*(1), 55–65. https://doi.org/10.1111/j.1468-3148.2005.00281.x

Willner, P. (2005). The effectiveness of psychotherapeutic interventions for people with learning disabilities: A critical overview. *Journal of Intellectual Disability Research, 49*.(Pt 1, 73–85. https://doi.org/10.1111/j.1365-2788.2005.00633.x

Willner, P. (2006). Readiness for cognitive therapy in people with intellectual disabilities. *Journal of Applied Research in Intellectual Disabilities, 19*(1), 5–16. https://doi.org/10.1111/j.1468-3148.2005.00280.x

Willner, P. (2009). Psychotherapeutic interventions in learning disability: Focus on cognitive behavioural therapy and mental health. *Psychiatry, 8*(10), 416–419. https://doi.org/10.1016/j.mppsy.2009.06.018

Willner, P., & Goodey, R. (2006). Interaction of cognitive distortions and cognitive deficits in the formulation and treatment of obsessive–compulsive behaviours in a woman with an intellectual disability. *Journal of Applied Research in Intellectual Disabilities, 19*(1), 67–73. https://doi.org/10.1111/j.1468-3148.2005.00279.x

Willner, P., Rose, J., Jahoda, A., Kroese, B. S., Felce, D., Cohen, D., … Hood, K. (2013). Group-based cognitive-behavioral anger management for people with mild to moderate intellectual disabilities: Cluster randomized controlled trial. *British Journal of Psychiatry, 203*(3), 288–296. https://doi.org/10.1192/bjp.bp.112.124529

Early Intervention

39

Michael J. Guralnick and Mary Beth Bruder

Contemporary systems of early intervention are designed to provide a comprehensive and integrated array of resources and supports to families whose children are experiencing or are at risk for a wide range of delays in development during the early childhood period. The overarching objective of these systems is to help create an environment that fosters children's development as optimally as possible and to establish a trajectory that will ultimately enable them to carry out their goals within family, community, and cultural contexts. Ideally this would be accomplished in conjunction with necessary supports. As expected, such community-based early intervention systems are complex and diverse. Although often including preventive intervention programs for children at risk for delays, early intervention systems focus primarily on a heterogeneous group of children for whom a range of communication, motor, socio-emotional, sensory-perceptual, or adaptive concerns and behaviors are evident. But also central to early intervention systems are children experiencing substantial delays in cognitive development, although these children demonstrate delays in many of the domains noted above as well. Children with a diagnosed condition that is highly likely to result in cognitive delays are also eligible to receive early intervention services. The vast majority of young children with substantial cognitive delays will meet criteria for intellectual disability by the time they reach school age, and it is these children and their families that are the focus of this chapter.

During the early childhood period in particular, promoting children's development is realized through systems consisting of a network of early intervention professionals who develop collaborative relationships with families to generate as optimal a developmentally supportive environment for the child as possible. Among other features, this family-centered approach is designed to assist families to become advocates for their child, to ensure that family priorities are respected and supported, and to provide sufficient information to enable families to understand the complex influences on and features of their child's development. Together, it is anticipated that families participating in this process will be able to confidently and competently adjust to their child's ever-changing characteristics and develop relationships that will support their child's development as optimally as possible over time (Bailey Jr. et al., 2006; Bruder, 2010; Dunst, 2017; Guralnick, 2011, 2019a).

The original version of this chapter was revised. The correction to this chapter is available at https://doi.org/10.1007/978-3-030-20843-1_58

M. J. Guralnick (✉)
University of Washington, Seattle, WA, USA
e-mail: mjgural@uw.edu

M. B. Bruder
University of Connecticut, Storrs, CT, USA

J. L. Matson (ed.), *Handbook of Intellectual Disabilities*, Autism and Child Psychopathology Series, https://doi.org/10.1007/978-3-030-20843-1_39

Principles

Centering early intervention on families constitutes an important principle for the field that has achieved international consensus (Guralnick, 2008). A corollary to that principle is the role developmental science plays in informing specific family-centered early intervention practices. For example, abundant evidence reveals that early intervention's focus on supporting parent-child relationships in numerous contexts, particularly relationships that constitute a discourse framework, an instructional partnership, and socioemotional connectedness, is a fundamental developmental mechanism that promotes a child's social and cognitive competence (Guralnick, 2011, 2019a). Other influential developmental processes consist of child experiences that are orchestrated by families. These include participation in community activities, diverse social networks, child care, preschool, and individual or group therapies, all of which constitute developmental pathways of considerable importance with respect to children's emerging competencies (Bruder, 2010; Dunst, Hamby, Trivette, Raab, & Bruder, 2000). Of course, family efforts to ensure a child's health and safety constitute mechanisms that can produce widespread effects on child development as well. In the context of developmental science, guidance to early intervention systems derived from these mechanisms of influence is provided with respect to assessment, intervention goals, corresponding intervention strategies, and their implementation and evaluation. In so doing comprehensive and integrated early intervention systems take advantage of the unusual levels of plasticity and sensitivity to environmental input characteristic of children during this developmental period (Bick & Nelson, 2017; Fox, Levitt, & Nelson III, 2010).

Early intervention principles that have also achieved international consensus address structural issues and values that can guide the system itself as well as related practices. Structural principles especially relevant include designing systems to ensure that early detection, surveillance, monitoring, and transition processes are in place; that evaluation procedures are embedded in all critical intervention goals, objectives, and activities; that the system can accommodate and attend to the complex and highly individualized needs of children and families; and that the system contains mechanisms for leadership capable of carrying out the overarching early intervention vision. Essential leadership functions must include the ability to achieve integration and coordination among components and service sectors at all levels to enable an early intervention system to function effectively. Principles related to values focus on the importance of providing intervention within a framework that maximizes full inclusion of children and families in community activities and programs and ensures that a meaningful partnership with families is established that includes an understanding of cultural differences and a recognition of their developmental implications and the importance of recommending intervention practices and strategies that have a firm evidence base embedded in a conceptually sound framework (Guralnick, 2008).

Vulnerable Children

These principles are incorporated to varying degrees into systems of early intervention in many countries throughout the world. In the United States, the systems of services for vulnerable children and their families are organized in the context of the Individuals with Disabilities Education Improvement Act (IDEA, 2004). This legislation, carried out through Part B of IDEA (Sect. 619), mandates services for preschool-age children in each state emphasizing educational interventions and is the responsibility of local school districts. In contrast, services for infants and toddlers (Part C of IDEA) center more directly on enhancing the capacity of families and providing individualized help-giving practices to meet children's needs (Bruder, 2005; Dunst, 2017; Hebbler, Greer, & Hutton, 2011). This program is administered by state designated agencies, and, although Part C programs are optional, all states continue to participate.

Eligibility to participate in the early intervention system for vulnerable children established by IDEA for Part C consists of three general categories. As suggested earlier, the first focuses on children demonstrating a substantial delay in one or more fundamental developmental domains (including cognitive development). Establishing the precise extent of the delay or delays required for eligibility varies from state-to-state and generally involves formal assessments, clinical opinion, and parental input. As expected, state eligibility definitions and assessment strategies have a major impact on the number and characteristics of the population that is served (Elbaum & Celimi-Aksoy, 2017; Rosenberg, Ellison, Fast, Robinson, & Lazar, 2013). Second, children are eligible if they have an established condition which indicates a high probability of exhibiting significant developmental delays during the early childhood period. This allows early intervention to commence prior to the appearance of those delays. Diagnosed conditions such as Down syndrome or fragile X syndrome are primary examples of children eligible in this category. Finally, although optional, states allow children to be eligible on the basis of biological risk factors. Extreme preterm birth, problems occurring during the perinatal period, or infectious diseases are among the most prominent biological risk factors that allow children with these risks to receive what is best conceptualized as preventive intervention; as they are provided services prior to the appearance of substantial delays should they emerge (see Guralnick, 2012). Children at risk due primarily to environmental factors, especially poverty and related concerns (see Evans & Kim, 2013), may also be included, but only a few states utilize this option as part of their eligibility criteria. Of course, many children in other delay categories are also at high risk due to environmental factors (Scarborough et al., 2004). Indeed, categories of environmental risk and delay overlap extensively (Halfon, Houtrow, Larson, & Newacheck, 2012). For children reaching preschool age (3–5 years), eligibility to participate in Part B can continue to be based on criteria related to developmental delay. Later-identified children, in particular, can also qualify for services by meeting categorical criteria including intellectual disability. Accordingly, early intervention systems, even those with relatively narrow eligibility requirements, must be prepared to address the complex and heterogeneous needs of children and families.

In this chapter, we focus on young children with substantial delays in cognitive development. For the most part, comparable delays in other developmental domains will also be apparent producing an overall pattern of general developmental delay despite characteristic unevenness of development (Shapiro & Batshaw, 2013; Shevell, Majnemer, Rosenbaum, & Abrahamowicz, 2001). Autism spectrum disorder, sensory and motor disorders, and especially clinically significant behavior problems (see Cheng, Palta, Kotelchuck, Poehlmann, & Witt, 2014; Crnic, Hoffman, Gaze, & Edelbrock, 2004) are among the conditions associated with substantial cognitive delay but will not be addressed directly in this chapter. We fully recognize the dynamic and changing nature of early development and corresponding difficulties achieving valid assessments and classifications specific to cognitive development at such young ages (Lobo, Paul, Mackley, Maher, & Galloway, 2014; Yang, Jong, Hsu, & Lung, 2011). Nevertheless, longitudinal studies indicate that, particularly during the preschool period, cognitive delays are highly likely to continue during the school years, qualifying children for services that meet criteria for intellectual disability (e.g., Keogh, Bernheimer, & Guthrie, 1997; Mangin, Horwood, & Woodward, 2017).

Information is provided in the next section with respect to the adjustments required to address the challenges encountered by families in their efforts to promote the development of their young child with a cognitive delay. As will be seen, challenges to development-promoting family patterns of interaction are considerable, and adjustments that are needed constitute an important framework to guide the design, implementation, and evaluation of early intervention systems.

Family Challenges and Adaptations

The extraordinary abilities of families with a child with a cognitive delay to modify their daily routines and to organize their home and community activities to accommodate to the developmental and behavioral patterns of their child, and to do so in a manner that supports their child's development, have been well documented (e.g., Bernheimer & Weisner, 2007). Many families, especially those with sufficient resources in terms of their own personal characteristics (e.g., mental and physical health, problem-solving abilities, coping style) and material resources (financial, social support network), have been able to display well-organized and integrated levels of sensitive responsiveness, affective warmth, and engagement in numerous situations with their child similar to that which occurs when parents interact with children without cognitive delays. For example, in the domain of parent-child interactions, compared to matched groups of typically developing children, studies have shown that parents of children with delays are able to appropriately scaffold and support their child in instructional and social play contexts, closely attune and adjust interaction strategies to children's changing ability levels over time, refrain from being highly directive, and find ways to support the autonomy of their child (de Falco, Venuti, Esposito, & Bornstein, 2009; Gilmore, Cuskelly, Jobling, & Hayes, 2009; Guralnick, Neville, Hammond, & Connor, 2008; Sterling, Barnum, Skinner, Warren, & Fleming, 2012; Venuti, de Falco, Esposito, & Bornstein, 2009). As in the case for children developing typically as well as those with other delays or risk factors, these and other adaptations of families are associated with advances in children's development (Guralnick, 2019a). Consistent evidence supports associations for these parent-child interactions with respect to cognition, social development, and various aspects of language and communication for heterogeneous groups of children with delays as well as those with specific etiologies (Feniger-Schaal & Joels, 2018; Fenning & Baker, 2012; Green, Caplan, & Baker, 2014; Hauser-Cram et al., 2001; Trivette, 2003; Warren & Brady, 2007; Warren, Brady, Sterling, Fleming, & Marquis, 2010; Zampini, Salvi, & D'Odorico, 2015). Moreover, these interaction patterns constitute the building blocks of parent-child and other relationships that are certain to support children's development throughout the early childhood period and beyond.

Child-Specific Challenges

The resilience of families of children with cognitive delays noted above can be appreciated further upon a close examination of the challenges posed to a family's ability to adjust to children's characteristics. To varying degrees, constraints imposed on children's developmental resources (cognitive, language, motor, social-emotional, and sensory-perceptual) and the processes that organize those resources (executive function, metacognition, social cognition, motivation, emotion regulation) result in a complex profile of social and cognitive competencies displayed in everyday goal-oriented activities (Guralnick, 2019a). General problems affecting a range of competencies, for example, with respect to seeking and organizing information about the physical and social world, or communicating needs and interests in an unambiguous manner, are common concerns (see Spiker, Boyce, & Boyce, 2002). Information obtained from cross-sectional and longitudinal studies of various etiologic-specific subgroups highlights many of the child characteristics that families must adjust to, find ways to organize the environment in a supportive and stimulating manner, and develop relationships that best facilitate their child's development. For example, for children with Williams syndrome, eye movement planning problems can create developmental consequences that adversely affect independent visual exploration and various aspects of attention (Brown et al., 2003; Landau, 2012). Similarly, developmental patterns affecting both developmental resources and organizational processes associated with children with Down syndrome have been well characterized and include special concerns with respect to executive function, task persistence and motivation, expressive language, and social cognition, among other developmental areas (e.g., Adamson, Bakeman, Deckner, & Romski, 2009; Cebula, Moore, & Wishart, 2010; Chapman & Bird, 2012; Daunhauer et al., 2014; Gilmore et al., 2009; Wishart, 1996). Constraints on competencies common to children with fragile X syndrome pose numerous problems for families due to their child's difficulties with respect to emotion regulation, social anxiety, working memory, gaze aversion, and effortful control; and many of these competencies are reflected in assessments of children's adaptive behavior (Abbeduto, Brady, & Kover, 2007; Kover, McCary, Ingram, Hatton, & Roberts, 2015; Roberts, Hatton, & Bailey, 2001; Robinson, Klusek, Poe, Hatton, & Roberts, 2018). Despite the higher probability that most of these child characteristics will be evident during the early childhood period, it is certainly the case that considerable variability (including minimal difficulties) in the developmental patterns of etiologic-specific and heterogeneous subgroups exists as a consequence of biological and environmental factors. Nevertheless, despite this variability in children's characteristics as well as a family's resources and their corresponding ability to adapt, considerable challenges to needed adjustments remain for many families. This circumstance may well require intensive and extensive involvement of an early intervention team.

Identifying Subgroups and Stressors

These constraints and others operate in the context of many child strengths, all contributing to the varying levels of children's social and cognitive competence evident at various points throughout the early childhood period and beyond (Burack, Russo, Flores, Iarocci, & Zigler, 2012). As a consequence, challenges to families to make adjustments to promote their child's development will differ in nature, scope, intensity, and timing as a result of current and changing child characteristics. Moreover, family resilience to meet these challenges varies as well, often linked to the availability of family resources (e.g., financial stability, social support, coping skills, or mental health status). These family resources are discussed later in a larger context. Taken together, as described below, subgroups of families emerge at various points in time in which challenges to adjust to specific child characteristics now constitute stressors that adversely affect the quality of various components of family patterns of interaction, particularly sensitive responsiveness. Similarly, child characteristics can create substantial challenges to a family's resources, with the resulting stressors also exerting an adverse effect on a family's pattern of interaction. The end result is a non-optimal environment that can potentially further constrain a child's development (Guralnick, 2001a, 2017a, 2017b).

Subgroups of families of children with cognitive delays susceptible to child-specific stressors can be identified through careful screening and assessment. As suggested, the consequences of these child-specific stressors take many forms. These include adverse effects on family patterns of interaction such as a lower quality of mother-child interactions, especially behaviors that are more negative and interfering, and providing language input not properly adjusted to children's ability levels (Blacher, Baker, & Kaladjian, 2013; Hauser-Cram et al., 2001; Thiemann-Bourque, Warren, Brady, Gilkerson, & Richards, 2014). More generally, these stressors can create circumstances that restrict relationships from forming between parents and children by adversely affecting processes related to establishing a framework in which extensive discourse can occur, by constraining the ability of children and parents to engage in mutual problem-solving actions (i.e., to form an instructional partnership), and by limiting activities that support the development of connectedness at the socioemotional level. Among other concerns, the limits imposed on socioemotional connectedness can affect the formation of a secure attachment and a cooperative pattern of relating (see Feniger-Schaal & Joels, 2018; Feniger-Schaal, Oppenheim, Koren-Karie, & Yirmiya, 2012). Addressing these relationship difficulties between parents and children (transactional processes) that result from a child's specific stressors that may emerge constitutes a critical task for the early intervention team.

Admittedly, it is not easy to predict or even identify those subgroups of families who are likely to have extensive difficulties successfully adjusting to their child's characteristics. Certain factors such as child behavior problems are predictive, but uncertainty remains, nevertheless, posing problems for an early intervention system from clinical, educational, and resource perspectives. Moreover, family resilience as reflected in the components of a family's resources also operates at a systems level, and it is often difficult to determine how the various components of a family's resources interact and work with one another in different situations to affect parent-child interactions as well as other family patterns of interaction. Consequently, a related but critical task for early intervention systems is to develop a process that is capable of identifying families most in need of support and then enabling them to access a family-centered intervention program that recognizes the interrelationships among children's characteristics, the various components of family patterns of interaction, and the resources available to families to support those interaction patterns. Ideally, such programs would operate in accord with the structural and values principles discussed earlier. Systems would also focus on the importance of relationships at all levels, ensure continuity of the program over time, and contain resources sufficient to comprehensively address the many stressors that may emerge capable of limiting optimal child development.

Community Involvement

In addition to child-specific effects on parent-child transactions noted above, it is also important to consider those community-based issues that often challenge a family's ability to maximize their child's development. Identifying high-quality and inclusive child care and school programs, assisting their child to form meaningful peer networks, finding a medical home with professionals knowledgeable and experienced in the care of children with cognitive delays, and engaging service providers with appropriate credentials and skills are among the community activities that parents must orchestrate to support their child's development and well-being (Dunst & Trivette, 2009). Combined with frequent negative societal attitudes and limited community resources, it is quite understandable how child characteristics can produce stressors restricting these development-enhancing community activities (Guralnick, 2019a).

Of considerable importance is the ability of families to maximize full participation in all community activities. This is especially the case for inclusive child care and preschool programs, as consistent evidence of developmental benefits has been found for those children participating in quality inclusive settings (Guralnick & Bruder, 2016; Justice, Logan, Lin, & Kaderavek, 2014). Once again, these issues highlight the importance of adopting a broader systems perspective in the context of early intervention and recognizing the importance of a comprehensive approach.

Family Resources

It is also the case that constant efforts to adapt to changing and challenging child characteristics create unusual problems that can affect many of the components of a family's resources throughout the early childhood period (Gallimore, Weisner, Bernheimer, Guthrie, & Nihira, 1993). For example, even at early stages, the emotional features of the diagnostic process can lead to many unresolved issues for parents which can adversely influence the quality of parent-child interactions over the long term (Barnett et al., 2006). Stress, often reaching clinical cutoff points, experienced by parents of children with delays directly related to child characteristics, is common and can be traced to child temperament, variations in mood, and especially behavior problems, with the latter elevating parent depressive symptoms (Gerstein, Crnic, Blacher, & Baker, 2009; Most, Fidler, Booth-LaForce, & Kelly, 2006; Neece, Green, & Baker, 2012; Woodman, Mawdsley, & Hauser-Cram, 2015; Zeedyk & Blacher, 2017). Periodic episodes of parent distress, including depression, can not only adversely affect parent-child interactions (Zeedyk & Blacher, 2017) but overall life satisfaction as well (see Nes et al., 2014).

Of importance, subgroup identification with respect to a family's resources for the purpose of early intervention remains critical, as most families demonstrate a remarkable degree of resilience. Indeed, ongoing adaptations rely extensively upon the personal characteristics of families, especially a positive coping style and the ability to engage their social support network (Minnes, Perry, & Weiss, 2015; Peer & Hillman, 2014). Social support can influence many components of a family's resources including parent mental health and the extent to which families feel competent and confident in their ability to parent a young child with a cognitive delay (Bailey, Nelson, Hebbeler, & Spiker, 2007; Guralnick, Hammond, Neville, & Connor, 2008; Kersh, Hedvat, Hauser-Cram, & Warfield, 2006).

Accordingly, a major task for early intervention systems with respect to supporting family adaptations is to accurately and sensitively monitor the various components of a family's resources, especially social support networks and levels of parent distress. The potential for these resources to be depleted at various points in time is considerable, especially for the many families with a child with a delay who also have extensive pre-existing environmental risk factors, particularly limited financial resources (Olsson & Hwang, 2008). These initial vulnerabilities in the form of higher environmental risk create a circumstance which can be exacerbated further such as by additional expenses related to child care responsibilities or lost income as a result of employment changes (Emerson & Hatton, 2009; Emerson, Hatton, Llewellyn, Blacher, & Graham, 2006). In view of the influence of these family resources on the quality of parent-child transactions, the extent of the child's involvement and participation in home, community, and educational activities as organized by parents, and on a child's health and safety, it is evident that the quality of the components of family resources has important consequences for a child's development (Guralnick, 2011, 2017a, 2017b).

As discussed more fully later, in view of the challenges and stressors to a family's resources and to a family's pattern of interacting with their child, effective systems of early intervention must be designed not only to address relationships but also to be organized in a manner that is comprehensive with an emphasis on continuity of services across the early childhood period. Taken together, the ability of the system to screen and then assess components relevant to children's characteristics, to family patterns of interaction that are linked to children's development, and to family resources that support those family interaction patterns are core elements of effective early intervention systems.

Systems and Effectiveness

Community-based systems of early intervention in the United States and many other countries have indeed been organized within a framework that reflects both a recognition of the diverse and complex needs of vulnerable children and the challenges facing families discussed above. Many of these community-based systems have also adopted the consensus principles governing the organization and operation of the system. This emergence and commitment to community-based early intervention systems as established by and modified through legislation and policy initiatives also reflect the expectation that both children and families will benefit substantially from a sophisticated array of well-organized and evidence-based interventions. Indeed, results from decades of intervention science involving diverse groups of families and children provided sufficient confidence to generate the political will to establish community-based early intervention systems and to continue to expand and refine the components of those systems. The relevant intervention science was summarized in 1997, during the period of rapid growth of early intervention programs just prior to and following the passage of P.L. 99–457 (Guralnick, 1997). This "first-generation" research included investigations involving children with cognitive delays and their families, demonstrating the ability of early intervention programs to reduce the decline in development that typically occurs over time in the absence of intervention and generate many other benefits (Guralnick, 2005, 2017a; Spiker & Hopmann, 1997). First-generation research also provided the foundation for subsequent "second-generation" studies that were designed to determine associations between well-defined family and child characteristics, emerging and innovative program features, and outcomes of relevance for both children and families. Findings from studies designed in this manner could form a database specifically organized to advance practice applications, with the potential for widespread implementation in community practice.

In the following section of this chapter, a brief history and description of the organization and practices of the early intervention system in the United States within the framework of the Individuals with Disabilities Education Act are discussed. This will be followed by a description of selected recent second-generation research designed to refine and enhance early intervention programs for children with cognitive delays and their families. In the final section, future directions intended to improve early intervention program quality and program effectiveness are discussed within a systems framework that is applicable to all young vulnerable children and their families.

Early Intervention: History, Organization, and Practices

In the United States, children under the age of 5 were not included in the federal mandate for special education under the Education of the Handicapped Act (EHA), passed in 1975 ("Education for All Handicapped Children Act of 1975, P.L. 94-142, 1977"). This law mandated that all school-age children with disabilities receive a free appropriate public education in the least restrictive environment under the direction of an Individualized Education Plan (IEP). States did receive incentive funds to enroll preschool-age children in public school programs and to provide training to preschool teachers under this Act. It was not until 1986, however, that Congress added several significant components to the EHA that specifically addressed the needs of eligible children under the age of 5. First, services for young children (ages 3–5) eligible for special education were mandated under the provisions of free, appropriate public education (Part B of P.L. 94–142). Second, these amendments created incentives for states to develop an early intervention program for children ages birth to three. This birth to three program was designed to establish a statewide system of interagency, multidisciplinary services available to eligible infants and toddlers and their families (as defined by each state through an agency also determined by each state). The name of the EHA was changed to the Individuals with Disabilities Education Act (IDEA) by the EHA amendments of 1991 (P.L. 102–119), and services for eligible infants and toddlers were made available in all states through IDEA amendments in 1997 (PL 105–17) under Part C of IDEA. This allowed states to receive funding for this age group if they implemented all assurances required under the subtitle. Part B, Section 619, addressed the preschool programmatic requirements of IDEA. Further, the IDEA amendments of 2004 required that services for infants and toddlers as well as preschool children be based on scientifically sound research findings, research that involves the application of rigorous, systematic, and objective procedures to obtain reliable and valid knowledge relevant to educational activities and programs. An added emphasis on the transition process between Part C and Part B (619) was also included in these amendments.

IDEA provides the guidance and structure for statewide systems of early childhood intervention for eligible infants, toddlers, and preschoolers and their families. While these two programs differ as a function of the age of the child and have different statutory requirements, both focus on structural and values components of early intervention to support children's development and family well-being. Some key elements of these two programs are described next.

Family Participation

Both Part C and Part B (619) address the civil rights of children and their parents through a system of procedural safeguards that are in place throughout the early intervention process. Part C, however, strengthens the role of the family by identifying it as a target of early intervention services. The preamble of the statute explicitly states a need for early intervention to enhance the capacity of families to meet the special needs of their infants and toddlers with disabilities. As a consequence, early intervention services for infants and toddlers are delivered in accordance with an Individual Family Service Plan (IFSP). Additionally, the IFSP outcomes must be based on a multidisciplinary assessment of the unique strengths and needs of the infant or toddler and the identification of services appropriate to meet such needs. Also required is a family-directed assessment of the resources, priorities, and concerns of the family and the identification of the supports and services necessary to enhance the family's capacity to meet the developmental needs of their infant or toddler.

Part B of IDEA also addresses parents in the preamble of the preschool and school age provisions of the statute by stating that IDEA Part B includes the protection of child and parent rights through the special education process and the assurance that teachers and parents have the necessary tools to improve educational results for children. Though not as explicit as Part C, the law does allow services that target parents under the related service of parent counseling and training. Likewise, parents may participate in the eligibility assessment for their child by providing information about their child in the context of assessment tools and strategies used to gather relevant functional, developmental, and educational information to determine whether the child has a delay or disability and to develop the content of a child's Individualized Education Plan (IEP).

Screening and Identification for Further Assessment

Both Part C and Part B (619) require a Child Find system that includes public awareness of the special needs of infants, toddlers, and preschoolers who may qualify for services under IDEA and the training of primary referral sources (e.g., medical and community program personnel) in screening and referral procedures. Specifically, IDEA requires that states ensure that all children with delays or disabilities are identified, located, and evaluated, and referral sources must be kept informed about early intervention supports, service models, and data on effectiveness.

There are multiple models for developmental and medical screening programs available (Gilliam, Meisels, & Mayes, 2005; McConnell & Rahn, 2016; McLean, Wolery, & Bailey, 2003; Yockelson, Linder, & Asman, 2015). One method used in many states is a developmental screening questionnaire completed by a parent or healthcare provider (Bricker, Macy, Squires, & Marks, 2013). A questionnaire serves several functions, ranging from identifying children who may not be meeting developmental milestones (and therefore need additional evaluation) to providing child development information and education to parents.

Assessment

An infant, toddler, or preschooler who is identified as needing more documentation to qualify for IDEA services undergoes a multidisciplinary assessment to generate a profile of their unique strengths and needs. In Part C the assessment is to be used for eligibility purposes and to identify services appropriate to meet an infant's or toddler's needs. The law states that the assessment must be a timely, comprehensive, and multidisciplinary evaluation of the development of each infant or toddler. To qualify for early intervention services, most children will require an assessment involving various disciplines. This assessment can serve a diagnostic function and create a valuable portrayal of the child's needs from medical, educational, and social systems perspectives. Part B (619) requires that assessments for children over 3 do not discriminate on race or have a cultural basis, are administered in the child's primary language, are used for purposes for which the assessments or measures are valid and reliable, and are administered by trained and knowledgeable personnel. As with screening, assessment tools should be standardized and result in an accurate representation of a child's skills (Gilliam et al., 2005; McConnell & Rahn, 2016; McLean et al., 2003).

After a child is determined to be eligible for Part C or Part B (619) services, further assessment may be carried out in conjunction with intervention (IFSP or IEP) planning. Assessment protocols are intended to focus on a child's application of their social and cognitive competencies in the context of home, community, and classroom-based activities and routines. It is important that these assessments are linked to intervention (Bagnato, McLean, Macy, & Neisworth, 2011), include the family (Macy, Thorndike-Christ, & Lin, 2010), and focus on the child's ability to participate in and use skills related to their adaptive functioning (communicating, walking, self-care, etc.) (Campbell & Sawyer, 2007; Fleming, Sawyer, & Campbell, 2011). Additionally, assessments for eligibility, program planning, and evaluation should share the same conceptual perspective and be linked for a seamless process.

Curriculum

Curricula for infants, toddlers, and preschoolers under IDEA are described as part of the IFSP or IEP process. As proposed by Dunst (1981), a curriculum consists of a series of carefully planned and designed activities, events, and experiences intentionally organized and implemented to reach specified objectives and goals and align with a particular philosophical and theoretical position. Both IFSPs and IEPs document a child's developmental and learning status, outline the day-to-day provision of services and supports designed to promote their development, identify those who will work with the family to deliver the services and interventions, and specify the evaluation procedures to monitor the effectiveness of the program. The IFSP, in particular, requires the inclusion of measurable outcomes expected to be achieved for the infant or toddler and the family, the assignment of a service coordinator to aid the family, and a formal transition process to preschool services when the eligible child turns 3 years of age.

The curricular content used in early childhood intervention derives primarily from developmental theory and the developmental trajectories common to most children. Most curricula in use in Part C and Part B (619) emphasize the unique patterns of individual children: their age, their strengths, their needs, their family preferences, and their outcomes and goals as developed by the intervention team, including the family. For children with cognitive delays in particular, a focus has been on the enhancement and facilitation of a child's competencies needed to participate in everyday activities and routines (Campbell & Sawyer, 2007; Dunst et al., 2000, 2001; McWilliam, 2010a, 2016; Woods, Kashinath, & Goldstein, 2004). This focus emphasizes accommodations, modifications, and adaptations needed by young children to learn within the context of routines and activities in the home, community, and classroom (Bruder, 2010). Examples of approaches, curricula, or combined strategies include embedded instruction, routine-based intervention, milieu teaching, naturalistic teaching, and activity-based instruction (see Snyder, Hemmeter, McLean, Sandall, & McLaughlin, 2013; Snyder, Rakap, Hemmeter, & McLaughlin, 2015). Of note, many of these interventions have their roots in curriculum development using incidental teaching (Hart & Risley, 1975) and teaching matrixes for children with cognitive delays (Williams & Gotts, 1977). These interventions were designed to be functional and efficient and to lead to generalized learning.

Many of the instructional methods that guide the orchestration of such development-enhancing learning opportunities across routines and activities continue to be based on applied behavioral analysis techniques. For almost 50 years, this instructional paradigm has been utilized in early childhood intervention (see Wolery, 2000) and has evolved from the isolated delivery of interventions to include a variety of expanded and naturalistic antecedents and logical consequences (e.g., environmental arrangements, group settings). Expanding and building new skills based on a child's demonstrated abilities or assets seem particularly effective (Raab, Dunst, & Hamby, 2016, 2017).

A more recent approach to the identification of instructional targets for young children with delays or disabilities is response to intervention or multi-tiered systems of instruction (Buysse & Peisner-Feinberg, 2013; Carta, McElhattan, & Guerrero, 2016). The features of this framework include ongoing assessment to provide information about how a child is responding to a general curriculum that is provided to all children (i.e., in inclusive settings). Interventions are then provided in progressive levels of intensity depending on the child's progress in the curriculum and instructional targets.

Services

Part C is designed to provide a coordinated array of services, all designed in a manner intended to center on families. These services include special instruction, coordinated family training, counseling, home visits, speech-language pathology and audiology, sign language and cued language, occupational therapy, physical therapy, psychological services, medical services only for diagnostic or evaluation purposes, health services necessary to enable the infant or toddler to benefit from the other early intervention services, social work services, vision services, assistive technology devices and assistive technology services, and transportation and related costs that are necessary to enable an infant or toddler and the infant's or toddler's family to receive other services, and service coordination. In contrast, Part B (619) provides special education and related services with instruction provided in the classroom and the home as well as in hospitals and institutions if needed. Services are also provided in other settings such as community early childhood or child care programs. A wide range of related services similar to Part C is also provided in these contexts.

Inclusion

IDEA also requires that services be delivered in environments with typical children. Preschool-age children receiving Part B (619) services must receive services in the least restrictive educational environment (LRE) to the maximum extent possible. LRE further mandates that special classes, separate schooling, or other removal of children with disabilities from the regular educational environment could occur only if the severity of their disability prohibited education in regular classes, when using supplementary aids and services. This principle of inclusion has not been easy to accomplish in practice, although IDEA data suggest that as many as two-thirds of eligible children participate in regular preschool settings for some amount of time (US Department of Education, 2017). As most of these children receive Part B (619) services in preschools, the challenge has been to combine well-integrated service delivery within typically occurring classroom routines with the general population of children. Inclusive practices are often constrained by schools where policy is to provide related services by multiple personnel from different disciplines, many of whom provide their services in one-to-one sessions outside of the classroom. While a team approach is still regarded as the preferred method to provide Part B (619) services, time constraints and high caseloads for related service personnel impact the time available to design and implement integrated service delivery for individual children and classrooms (Guralnick & Bruder, 2016).

Early intervention services under Part C of IDEA are to be delivered in natural environments in accordance with the IFSP. Natural environments include the home or places in which same age children who do not have disabilities participate (e.g., child care, community programs). In 2015, nearly 90 percent of infants and toddlers served under Part C received early intervention services primarily in the home (US Department of Education, 2017). Less frequently used were community-based settings as the primary early intervention environment for those served under Part C (Bruder, 2001; Bruder & Dunst, 2011; Guralnick & Bruder, 2016). It has been reported that home visits are delivered once a week on average, and data collected on the content and processes of early intervention home visits suggests that the majority of time is spent by the service provider teaching the infant or toddler, with less time collaborating and working with caregivers (Campbell & Sawyer, 2007; Kemp & Turnbull, 2014; Peterson, Luze, Eshbaugh, Jeon, & Ross Kantz, 2007).

A primary provider service model is common in these circumstances (Vail, Lieberman-Betz, & McCorkle, 2018). In this approach, one home visitor delivers services to families using a coaching method (Shelden & Rush, 2010). This service delivery model is not new and has its roots in the transdisciplinary model of teaming in early intervention which was designed to minimize the number of adults who visited a family and child while maximizing the efficiency of intervention (United Cerebral Palsy National Collaborative Infant Project, 1976). A central feature of the transdisciplinary model is the availability of a full complement of team members representing different disciplines and services and who meet and consult with the home visitor (usually representing the discipline and service most relevant to the needs and priorities of the child and family) to provide input on the delivery and integration of interventions (Bruder & Bologna, 1993). It is clearly essential that service providers understand and support their role in connection with the other service providers and to the family (King et al., 2009).

Guidance and reviews of successful practices for inclusive early childhood service delivery have been published over the years (see Barton & Smith, 2015; Bruder, 1993; Guralnick, 1978, 2001b; Odom, Buysse, & Soukakou, 2011; Sandall, Schwartz, & Gauvreau, 2016; Winton, 2016). Most provide examples of adaptations, supports, and modifications to facilitate a child's inclusion and learning within a general early childhood classroom in the context of activities and routines incorporating adapted materials and instructional practices (Campbell, Milbourne, & Kennedy, 2012). Universal design for learning (UDL) is also a framework being applied to modify and adapt classroom activities for young children with delays and disabilities (Butera, Horn, & Palmer, 2016). UDL has most often been used with older children, though the concepts of providing multiple means of representation of materials and learning activities, multiple means of engagement for children to be involved in learning or teaching routines, and multiple forms of expression for children with varying communication modes and abilities can enhance the development of all young children attending an inclusive classroom. Lastly, assistive technology devices and services may be especially valuable for children with substantial cognitive delays (see Campbell, Milbourne, Dugan, & Wilcox, 2006). These range from low-tech materials such as special seating or utensils to enhance a child's participation in everyday routines and activities to the use of high-tech communication devices with voice synthesizers to enable a child to communicate in the home, community, and classroom environments.

Summary

Taken together it is evident that, in the United States, the importance and benefits of a system of early intervention has been recognized through the implementation of IDEA. The main components of this system have been outlined above, all intended to enhance the development of children with cognitive and other delays and to support families. As is the case for any complex system, it must continue to develop by incorporating new information and adapting and refining its methods, conceptual framework, and structural features to achieve further progress. Recent studies advancing our knowledge base in critical areas focusing on children with cognitive delays are discussed next. In the final section of this chapter, some general suggestions for future directions are presented.

Current Research Directions

With early intervention systems firmly in place in the United States and elsewhere, smaller scale or focused studies involving children with cognitive delays have been conducted with the expectation that advances that result can be incorporated into the existing system of early intervention practices. For example, some of the more recent early intervention research studies focusing on children with cognitive delays have continued to organize intervention activities within a structured, behavioral framework. These techniques, primarily based on applied behavior analysis interventions that have demonstrated their effectiveness in increasing the cognitive development of children with autism spectrum disorder (Eldevik et al., 2010), have been drawn upon to provide comprehensive programs for children with cognitive delays. Preliminary findings suggest that increases in cognitive development occur for a heterogeneous group of children with delays receiving behavioral intervention while attending inclusive preschool programs (e.g., Eldevik, Jahr, Eikeseth, Hastings, & Hughes, 2010). These techniques have also been successfully applied in more specific developmental areas such as motor behaviors, verbal imitation, and requesting strategies for children with delays (Bauer & Jones, 2014, 2015). Clearly, as demonstrated in these and previous studies, structure to promote specific child skills at various points in time can be of considerable benefit. Further work to determine how these skills are integrated and utilized over time in the service of children's goals will provide valuable information for subsequent intervention approaches especially if organized within a broader developmental context.

Parent-Child Interactions

Indeed, given the numerous challenges posed by children with cognitive delays to a family's pattern of interactions throughout the early childhood period discussed earlier, it may be best for early intervention programs to adopt a long-term developmental perspective. This can be accomplished by recognizing at the earliest possible stages the components and significance of a family's pattern of interactions as the primary experiential pathways of influence on a child's development (Guralnick, 2011, 2019a). To be sure, numerous intervention strategies, including focused and structured approaches, can be of value in this regard, many of which can be embedded within a more comprehensive framework that addresses developmental pathways associated with children's social and cognitive competence. Critical to this more developmentally oriented comprehensive approach are strategies that promote relationship formation between parents and children. This often occurs in the context of a discourse framework, an instructional partnership, or socioemotional connectedness discussed earlier, thereby providing the longer-term developmental context essential for establishing high-quality parent-child transactions. Efforts to support these types of relationships can be readily incorporated into well-established family routines (see McWilliam, 2010b).

The building blocks of parent-child relationships noted earlier in this chapter consist of sensitive responsiveness, affective warmth, and engagement. Sensitive responsiveness in particular, in all its various forms, has continued to demonstrate its value with respect to promoting the social and cognitive competence of children with delays (Trivette, 2003). Sensitive responsiveness in the context of a discourse framework has been a special interest to investigators emphasizing, among other strategies, contingent responding to child initiatives, ensuring balanced exchanges, minimizing directives, focusing on the child's attention and interests, and narrating activities (Karaaslan, Diken, & Mahoney, 2013; Kim & Mahoney, 2005; Mahoney, Perales, Wiggers, & Herman, 2006). Through demonstrations, coaching, feedback, reflection, and practice in various contexts and family routines, parents participating in these investigations were able to become proficient in organizing and applying strategies that promote parent-child relationships. Although further evidence with respect to child-specific outcomes is needed, from a developmental and evidence-based perspective, promoting relationships in this manner can be seen as an essential element of contemporary early intervention programs.

Highly focused applications that incorporated sensitive responsiveness strategies in early intervention programs can also be found in studies designed to promote the language and social communication abilities of prelinguistic children with cognitive delays. A detailed review of findings for this extensive group of studies can be found elsewhere (Guralnick, 2019a). However, a common objective of these studies was to promote children's intentional communication and to expand their expressive language. The ultimate goal was intended to further a discourse relationship between parents and children. Interestingly, evidence suggests that even clinician-implemented interventions can produce the most benefits for children specifically from that subgroup of parents who demonstrate a higher initial level of responsiveness to their child at home (Yoder & Warren, 2001). Moreover, maternal responsiveness at home may also increase due to increased intentional communications by children participating in clinician-implemented interventions (Yoder & Warren, 1999). Subsequent work involving alternative and augmentative communication strategies for prelinguistic children has also been carried out, engaging both parents and clinicians to varying degrees in the intervention process. Results have been promising in this very complex area suggesting the value of evidence-based refinements to early intervention practices for prelinguistic children (Romski et al., 2010; Wright, Kaiser, Reikowsky, & Roberts, 2013). However, the existence of inconsistent findings or small effects for a range of parent and clinician strategies must be recognized as well, with an ongoing concern with respect to effects fading over time following the end of the intervention. To be most effective, early intervention systems must be designed to provide a sufficient degree of continuity to ensure optimum outcomes. Perhaps cost-effective techniques utilizing distance learning approaches (video teleconferencing) that are showing promise with regard to supporting a discourse framework will be of value as they are further refined (McDuffie et al., 2016).

From a larger perspective, this pattern further suggests the importance of adopting a systems approach to early intervention. This is especially the case when considering how family-orchestrated child experiences, such as clinician intervention and parent sensitive responsiveness, interact to produce a cumulative effect. Indeed, evidence for heterogeneous groups of preschool children with varying language abilities has indicated that strategies used as part of an enhanced milieu intervention that included both parents and clinicians produced greater benefits than a clinician only group (Kaiser & Roberts, 2013). Benefits included increases in parents' use of sensitive responsive strategies in a naturalistic manner and children's expressive vocabulary. Clearly, early intervention is most effectively accomplished by following a coordinated, comprehensive approach to strengthen children's social and cognitive competence.

It is also important to recognize that progress has been achieved in these and other investigations with respect to identifying subgroups of children and families most likely to be responsive to the types of interventions described above. Moreover, by adjusting to child-specific characteristics, increasingly informed by etiologic-specific studies, the early intervention team may be in a position to more effectively problem-solve over an extensive time period to establish a much needed discourse framework for children with limited communication skills. Ongoing work utilizing adult learning techniques, such as incorporating the sequence of teach-model-coach-review, may be of particular value to the early intervention team in this context (Wright & Kaiser, 2017).

Inclusive Preschools

It is clearly recognized that inclusion is a value well-grounded in philosophical, legal, legislative, as well as international human rights principles. Yet the implementation of inclusive practices in group settings such as preschool programs brings with it the obligation to ensure that children's developmental progress reflected in the many aspects of their social and cognitive competence will, at minimum, be equivalent to progress that would occur within highly resourced yet specialized programs (Guralnick, 2001b). Numerous smaller-scale studies, often evaluated within a resource-rich context implemented by well-trained staff, have indicated that through thoughtful program accommodations to children, this is indeed the case (Buysse, 2011; Guralnick, 2001c). Clearly, these earlier studies provided our field with a level of confidence that inclusive practices, especially in preschool settings, can be both feasible and effective for a wide range of children with delays and disabilities.

As the movement toward universal preschool education continues to gain momentum and the promise that more communities will actually promote full inclusion programs in the future, a key question concerns the progress of children with cognitive delays occurring in inclusive preschool environments organized within large-scale public systems. This is especially critical as preschool programs today are educationally oriented with a substantial number of goals designed to prepare children to be ready to achieve academically. Two studies focusing on high-quality public school programs have addressed this issue (Phillips & Meloy, 2012; Weiland, 2016). Employing regression discontinuity designs that take advantage of cutoff dates for children's enrollment in public preschool programs, evidence from this work revealed that greater progress over the course of 1 year was achieved by children with developmental delays on most academically oriented measures in comparison to children who participated in other programs or community activities. Measures of early literacy, receptive language, and early numeracy were central to these studies. Despite some inconsistency in terms of program effectiveness or rates of change between children who participated in the preschool program and comparison children across outcomes, these studies nevertheless provided strong evidence of the ability of high-quality large-scale public inclusive educational programs to meaningfully and effectively promote the pre-academic skills of children with delays.

Of interest, in the Weiland (2016) study in the Boston public preschool program, the rate of progress of children with special needs, including the subsample of children with developmental delays, even exceeded those of typically developing children on some measures. Certainly curriculum factors, teachers well trained to meet the educational needs of a developmentally diverse child population, and the organizational skills required to create a universal publicly funded pre-kindergarten program are among the factors that contributed to these positive outcomes. Also likely contributing were the daily interactions of children with delays with their typically developing peers. Experiencing more complex language, observing advanced social and problem-solving skills, and receiving informative feedback by more advanced peers can have a substantial developmental impact (Justice et al., 2014). Future research will help identify critical program elements and confirm the ability of these larger-scale publicly funded inclusive programs to meet the needs of children with substantial cognitive delays. Implementation science will likely play a prominent role as well in moving comprehensive early intervention programs to scale and incorporating mechanisms capable of refining and enhancing programs further to accommodate new findings (Halle, Metz, & Martinez-Beck, 2013).

Peer Relationships

Early work also demonstrated that social interactions between young children with delays and their peers occurred at a far higher level in inclusive settings in comparison to specialized settings (Buysse & Bailey, 1993; Guralnick, Connor, Hammond, Gottman, & Kinnish, 1996). However, related work revealed that more in-depth peer relationships such as those characterized as friendships seemed unaffected (Guralnick, Gottman, & Hammond, 1996). This suggested that, despite the social interaction benefits associated with inclusive settings, a closer look into the development and characteristics of the peer relationships of children with delays was in order. Of special interest was an examination of the fundamental features of children's peer-related social competence that underlie children's peer relationships. When research in this area was carried out, numerous problems became apparent (Guralnick, 1999, 2010). In general, compared to appropriately matched groups, preschool age children with cognitive delays displayed peer-related social competence abilities that were not commensurate with their level of cognitive development, likely adversely affecting friendships as well as other important aspects of their peer relationships. Critical aspects of peer-related social competence including the social tasks of peer group entry, maintaining play under rapidly changing circumstances, and resolving conflicts were clearly affected. Problems were also evident with respect to both the appropriateness and effectiveness of social strategies when engaging in social tasks with peers.

To address this issue, a 2-year comprehensive intervention based on a developmental framework was carried out to improve the peer-related social competence of children with delays (Guralnick, Connor, Neville, & Hammond, 2006). Both teachers and parents were involved in the intervention which was individualized to the characteristics of children with delays discussed earlier in this chapter. For example, strategies were devised to address problems related to executive function, social cognition, and emotion regulation in the context of social tasks. Results based on measures derived from observations of interactions in inclusive playgroups revealed that, compared to children randomized to usual community services, important features of the peer interactions of children in the intervention group benefitted, especially for those children with IQs below 70. Minimizing negative exchanges with peers as a consequence of the intervention was perhaps most notable. Yet other measures that reflected more in-depth changes in peer-related social competence, such as social strategies likely to foster friendships and promote core aspects of social competence, were unaffected.

Since the quality of interpersonal relationships becomes more closely linked to an individual's overall well-being over time, it is clear that future work is essential in order to alter this peer-related social competence trajectory. Fortunately, we now have a much better understanding of the underlying developmental processes likely to influence young children's social competence with their peers that can serve as a guide to intervention approaches. The effectiveness and appropriateness of children's relationships with their peers certainly depend upon their developmental resources (e.g., cognition, language) and organizational processes (e.g., executive function, social cognition) as is the case for all competencies displayed as children carry out their interpersonal goals. Designing innovative interventions for children with cognitive delays that can influence these child characteristics in the context of social tasks encountered during peer-peer interactions is essential whether they occur in home, school, or other settings. Of importance, peer relationships are best conceptualized as being embedded within the larger construct of social competence. As such, social competence, including competence displayed in peer contexts by children with delays, is associated with the way in which family members and children interact with one another and model the more balanced and responsive social interactions common to peer relationships (Guralnick, Connor, Neville, & Hammond, 2008; Guralnick, Neville, Hammond, & Connor, 2007). Accordingly, this highlights once again the importance of adopting a comprehensive developmental approach when considering interventions involving complex behavior patterns, including those related to peer interactions.

Future Directions

The consensus principles described earlier provide important guidance for the ongoing development and refinement of early intervention systems. Developmental science provides a well-established framework governing family-centered practices and contributes to our understanding of the mechanisms influencing a child's development. Intervention science consistent with developmental science continues to inform practice, seeking to ensure that only evidence-based practices are selected when IFSPs and IEPs are developed and implemented. Recent refinements, based on intervention science in the early intervention field discussed in this chapter focusing on children with cognitive delays, emphasized strategies to enhance parent-child interactions, studies of the effectiveness of community-based inclusive preschools, and outcomes of comprehensive curricula to promote children's peer-related social competence.

Close examination of current early intervention systems also reveals that numerous opportunities exist to substantially improve virtually all system components. Limits on the comprehensiveness of services are perhaps most evident. As discussed, we now have a firm understanding of the mechanisms that influence child development, including the various components of the quality of parent-child interactions, family-orchestrated child experiences, and a child's health and safety. Yet, many of these components are not incorporated and integrated within current systems or are carried out with such limited intensity to suggest that only minimal effects can be expected. Early intervention problem-solving that incorporates children's characteristics and a family's resources is not as well developed as our knowledge base. In addition to concerns about the comprehensiveness of the system are concerns about its continuity. The evolution of Part B (619) and Part C, with its differing administrative structures and emphases, does not easily allow for continuity of services that maintain a developmental perspective. Unquestionably, a more expansive transition plan would be of value. An emphasis on promoting the well-being of families and enhancing all aspects of a family's resources is especially critical for families of children with cognitive delays (Crnic, Neece, McIntyre, Blacher, & Baker, 2017).

Our dramatically increasing knowledge of the developmental trajectories and corresponding risk and protective factors for children with cognitive delays with confirmed genetic etiologies is certain to generate new intervention strategies responsive to these profiles (Dykens, Hodapp, & Finucane, 2000; Fidler, Daunhauer, Will, Gerlach-McDonald, & Schworer, 2016). The era of personalized interventions will allow early intervention teams to adapt strategies to known developmental mechanisms of influence as reflected in children's developmental profiles in order to maximize the impact of those intervention strategies. A major challenge for early intervention inclusive community-based systems is their ability to utilize this child-specific developmental information as well as second-generation research findings and incorporate this knowledge into practice. Close associations with the early intervention research community and a knowledgeable workforce are critical. Fortunately, an implementation science is also emerging with components that bring a conceptual framework and intervention science into better alignment as programs become established or add new approaches to their service system (Fixsen, Blase, Metz, & Van Dyke, 2013; Guralnick, 2019b; Metz, Halle, Bartley, & Blasberg, 2013).

Perhaps the most crucial element in this regard, and one critical for successful implementation of all conceptually sound and empirically supported interventions, is to ensure that the early intervention workforce operates within the well-established principles that guide early intervention described earlier. Unfortunately, recent examinations of the status of the early childhood intervention workforce have identified a number of issues of concern with respect to the quality and effectiveness of Part C and Part B (619) practices, services, and programs (Bruder, 2010, 2016; Bruder, Mogro-Wilson, Stayton, & Dietrich, 2009; Vail et al., 2018; Woods & Snyder, 2009; Zaslow, 2009). These issues include shortages of personnel; inequities in wages and compensation for personnel across programs; shortages of preservice programs of study, coursework, and practicum opportunities; limited funding for continuing education; the absence of integrated and comprehensive personnel development systems that meet national personnel standards and adult learning guidelines; and limited experimental evidence about the effects of preservice and continuing education on child and family learning. Clearly, outcomes for early intervention systems will be constrained unless this crucial issue is addressed.

These and other issues may ultimately be best addressed within the context of large-scale, comprehensive systems of early childhood development, one inclusive of all children. Although we are a long way from such inclusive, community-based early childhood systems, guidelines and specific recommendations suggest the feasibility and benefits of this framework from both administrative and conceptual perspectives (Bruder, 2010). Should such a system emerge, the successful implementation of agreed-upon early intervention principles and corresponding services and supports for young children with cognitive and other delays becomes more likely, especially in view of the increasing compatibility of systems with a developmental framework relevant to all children (Guralnick, 2019a).

Acknowledgments *Springer Publishing*: The writing of this chapter was supported by grants from the Administration on Intellectual and Developmental Disabilities (ACF-AIDD-90DD0680) and (ACF-AIDD-90DD0015), the National Institute of Child Health and Human Development (U54 HD083091), and the US Department of Education, Office of Special Education Programs (H325B120004). The content is solely the responsibility of the authors and does not necessarily represent the official views of the funding agencies.

Conflict of Interest: The authors declare no conflict of interest.

References

Abbeduto, L., Brady, N., & Kover, S. T. (2007). Language development and Fragile X syndrome: Profiles, syndrome-specificity, and within-syndrome differences. *Mental Retardation and Developmental Disabilities Research Reviews, 13*, 36–46. https://doi.org/10.1002/mrdd.20142

Adamson, L. B., Bakeman, R., Deckner, D. F., & Romski, M. (2009). Joint engagement and the emergence of language in children with autism and Down syndrome. *Journal of Autism and Developmental Disorders, 39*, 84–96. https://doi.org/10.1007/s10803-008-0601-7

Bagnato, S. J., McLean, M., Macy, M., & Neisworth, J. T. (2011). Identifying instructional targets for early childhood via authentic assessment: Alignment of professional standards and practice-based evidence. *Journal of Early Intervention, 33*, 243–253. https://doi.org/10.1177/1053815111427565

Bailey, D. B., Jr., Nelson, L., Hebbeler, K., & Spiker, D. (2007). Modeling the impact of formal and informal supports for young children with disabilities and their families. *Pediatrics, 120*, e992–e1001. https://doi.org/10.1542/peds.2006-2775

Bailey, D. B., Jr., Bruder, M. B., Hebbeler, K., Carta, J., Defosset, M., Greenwood, C., … Barton, L. (2006). Recommended outcomes for families of young children with disabilities. *Journal of Early Intervention, 28*, 227–251. https://doi.org/10.1177/105381510602800401

Barnett, D., Clements, M., Kaplan-Estrin, M., McCaskill, J. W., Hunt, K. H., Butler, C. M., . . . Janisse, H. C. (2006). Maternal resolution of child diagnosis: Stability and relations with child attachment across the toddler to preschooler transition. *Journal of Family Psychology, 20,* 100–107. https://doi.org/10.1037/0893-3200.20.1.100

Barton, E. E., & Smith, B. J. (2015). Advancing high-quality preschool inclusion: A discussion and recommendations for the field. *Topics in Early Childhood Special Education, 35*, 69–78. https://doi.org/10.1177/0271121415583048

Bauer, S. M., & Jones, E. A. (2014). A behavior analytic approach to exploratory motor behavior. *Infants & Young Children, 27*, 162–173. https://doi.org/10.1097/IYC.0000000000000004

Bauer, S. M., & Jones, E. A. (2015). Requesting and verbal imitation intervention for infants with Down syndrome: Generalization, intelligibility, and problem solving. *Journal of Developmental and Physical Disabilities, 27*, 37–66. https://doi.org/10.1007/s10882-014-9400-6

Bernheimer, L. P., & Weisner, T. S. (2007). Let me just tell you what I do all day. *Infants & Young Children, 20*, 192–201.

Bick, J., & Nelson, C. A., III. (2017). Early experience and brain development. *WIREs Cognitive Science, 8*, e1387. https://doi.org/10.1002/wcs.1387

Blacher, J., Baker, B. L., & Kaladjian, A. (2013). Syndrome specificity and mother-child interactions: Examining positive and negative parenting across contexts and time. *Journal of Autism and Developmental Disorders, 43*, 761–774. https://doi.org/10.1007/s10803-012-1605-x

Bricker, D., Macy, M., Squires, J., & Marks, K. (2013). *Developmental screening in your community: An integrated approach for connecting children with services*. Baltimore, MD: Brookes Publishing Co.

Brown, J., Johnson, M. H., Paterson, S. J., Gilmore, R., Longhi, E., & Karmiloff-Smith, A. (2003). Spatial representation and attention in toddlers with Williams syndrome and Down syndrome. *Neuropsychologia, 41*, 1037–1046. https://doi.org/10.1016/S0028-3932(02)00299-3

Bruder, M. B. (1993). The provision of early intervention and early childhood special education within community early childhood programs: Characteristics of effective service delivery. *Topics in Early Childhood Special Education, 13*, 19–37.

Bruder, M. B. (2001). Infants and toddlers: Outcomes and ecology. In M. J. Guralnick (Ed.), *Early childhood inclusion: Focus on change* (pp. 203–228). Baltimore, MD: Brookes Publishing Co.

Bruder, M. B. (2005). Service coordination and integration in a developmental systems approach to early intervention. In M. J. Guralnick (Ed.), *The developmental systems approach to early intervention* (pp. 29–58). Baltimore, MD: Brookes Publisher.

Bruder, M. B. (2010). Early childhood intervention: A promise to children and families for their future. *Exceptional Children, 76*, 339–355. https://doi.org/10.1177/001440291007600306

Bruder, M. B. (2016). Personnel development practices in early childhood intervention. In B. Reichow, B. A.

Boyd, E. E. Barton, & S. L. Odom (Eds.), *Handbook of early childhood special education* (pp. 289–334). Switzerland: Springer.

Bruder, M. B., & Bologna, T. M. (1993). Collaboration and service coordination for effective early intervention. In W. Brown, S. K. Thurman, & L. Pearl (Eds.), *Family-centered early intervention with infants and toddlers: Innovative cross-disciplinary approaches* (pp. 103–127). Baltimore, MD: Brookes.

Bruder, M. B., & Dunst, C. J. (2011). Infant, toddler and preschool inclusion in community activities. *Revista Educacion Inclusiva, 4*, 21–34.

Bruder, M. B., Mogro-Wilson, C. M., Stayton, V. D., & Dietrich, S. J. (2009). The national status of in-service professional development systems for early intervention and early children special education practitioners. *Infants & Young Children, 22*, 13–20. https://doi.org/10.1097/01.IYC.0000343333.49775.f8

Burack, J. A., Russo, N., Flores, H., Iarocci, G., & Zigler, E. (2012). The more you know the less you know, but that's OK: Developments in the developmental approach to intellectual disability. In J. A. Burack, R. M. Hodapp, G. Iarocci, & E. Zigler (Eds.), *The Oxford handbook of intellectual disability and development* (pp. 3–12). New York, NY: Oxford University Press.

Butera, G., Horn, E. M., & Palmer, S. B. (2016). Understanding science, technology, engineering, arts, and mathematics (STEAM). In B. Reichow, B. Boyd, E. E. Barton, & S. L. Odom (Eds.), *Handbook of early childhood special education* (pp. 143–161). New York, NY: Springer.

Buysse, V. (2011). Access, participation, and supports: The defining features of high-quality inclusion. *Zero to Three, 31*, 24–31.

Buysse, V., & Bailey, D. B., Jr. (1993). Behavioral and developmental outcomes in young children with disabilities in integrated and segregated settings: A review of comparative studies. *The Journal of Special Education, 26*, 434–461. https://doi.org/10.1177/002246699302600407

Buysse, V., & Peisner-Feinberg, E. S. (Eds.). (2013). *Handbook of response to intervention in early childhood*. Baltimore, MD: Brookes Publishing.

Campbell, P. H., Milbourne, S. A., Dugan, L. M., & Wilcox, M. J. (2006). A review of the evidence on practices for teaching young children to use assistive technology devices. *Topics in Early Childhood Special Education, 26*, 3–13. https://doi.org/10.1177/02711214060260010101

Campbell, P. H., Milbourne, S. A., & Kennedy, A. A. (2012). *Cara's kit for toddlers: Creating adpations for routines and activities*. Baltimore, MD: Brookes Publishing Co.

Campbell, P. H., & Sawyer, L. B. (2007). Supporting learning opportunities in natural settings through participation-based services. *Journal of Early Intervention, 29*, 287–305.

Carta, J., McElhattan, T. E., & Guerrero, G. (2016). The application of response to intervention to young children with identified disabilities. In B. Reichow, B. Boyd, E. E. Barton, & S. L. Odom (Eds.), *Handbook of early childhood special education* (pp. 163–178). New York, NY: Springer.

Cebula, K. R., Moore, D. G., & Wishart, J. G. (2010). Social cognition in children with Down's syndrome: Challenges to research and theory building. *Journal of Intellectual Developmental Disability Research, 54*, 113–134. https://doi.org/10.1111/j.1365-2788.2009.01215.x

Chapman, R. S., & Bird, E. K.-R. (2012). Language development in childhood, adolescence, and young adulthood in persons with Down syndrome. In J. A. Burack, R. M. Hodapp, G. Iarocci, & E. Zigler (Eds.), *The Oxford handbook of intellectual disability and development* (pp. 167–183). New York, NY: Oxford University Press.

Cheng, E. R., Palta, M., Kotelchuck, M., Poehlmann, J., & Witt, W. P. (2014). Cognitive delay and behavior problems prior to school age. *Pediatrics, 134*, e749–e757. https://doi.org/10.1542/peds.2014-0259

Crnic, K. A., Hoffman, C., Gaze, C., & Edelbrock, C. (2004). Understanding the emergence of behavior problems in young children with developmental delays. *Infants and Young Children, 17*, 223–235.

Crnic, K. A., Neece, C. L., McIntyre, L. L., Blacher, J., & Baker, B. L. (2017). Intellectual disability and developmental risk: Promoting intervention to improve child and family well-being. *Child Development, 88*, 436–445. https://doi.org/10.1111/cdev.12740

Daunhauer, L., Fidler, D. J., Hahn, L., Will, E., Lee, N. R., & Hepburn, S. (2014). Profiles of everyday executive functioning in young children with Down syndrome. *American Journal on Intellectual and Developmental Disabilities, 119*, 303–318. https://doi.org/10.1352/1944-7558-119.4.303

de Falco, S., Venuti, P., Esposito, G., & Bornstein, M. H. (2009). Mother-child and father-child emotional availabiity in families of children with Down syndrome. *Parenting, Science and Practice, 9*, 198–215. https://doi.org/10.1080/15295190902844381

Dunst, C. J. (1981). *Infant learning: A cognitive-linguistic intervention strategy* (Vol. 4). Allen, TX: DLM Teaching Resources.

Dunst, C. J. (2017). Family systems early childhood intervention. In H. Sukkar, C. J. Dunst, & J. Kirkby (Eds.), *Early childhood intervention: Working with families of young children with special needs* (pp. 36–58). Oxon, UK: Routledge Press.

Dunst, C. J., Bruder, M. B., Trivette, C., Hamby, D. W., Raab, M., & McLean, M. (2001). Characteristics and consequences of everyday natural learning opportunities. *Topics in Early Childhood Special Education, 21*, 68–92. https://doi.org/10.1177/027112140102100202

Dunst, C. J., Hamby, D., Trivette, C. M., Raab, M., & Bruder, M. B. (2000). Everyday family and community life and children's naturally occurring learning opportunities. *Journal of Early Intervention, 23*, 151–164. https://doi.org/10.1177/10538151000230030501

Dunst, C. J., & Trivette, C. M. (2009). Capacity-building family-systems intervention practices. *Journal of Family Social Work, 12*, 119–143. https://doi.org/10.1080/10522150802713322

Dykens, E. M., Hodapp, R. M., & Finucane, B. M. (2000). Toward etiology-based work. In E. M. Dykens, R. M. Hodapp, & B. M. Finucane (Eds.), *Genetics and mental retardation syndromes: A new look at behavior and interventions* (pp. 3–22). Baltimore, MD: Brookes Publishing Co.

Education for All Handicapped Children Act of 1975, P.L. 94–142, Title 20, U. S. C. 1401 et seq U.S. Statutes at Large, 89, 773–796, Pub. L. No. 94–142 (1977).

Elbaum, B., & Celimi-Aksoy, S. (2017). Empirically identified subgroups of children served in part C early intervention programs. *Journal of Developmental & Behavioral Pediatrics, 38*, 510–520. https://doi.org/10.1097/DBP.0000000000000475

Eldevik, S., Hastings, R. P., Hughes, J. C., Jahr, E., Eikeseth, S., & Cross, S. (2010). Using participant data to extend the evidence base for intensive behavioral intervention for children with autism. *American Journal on Intellectual and Developmental Disabilities, 115*, 381–405. https://doi.org/10.1352/1944-7558-115.5.381

Eldevik, S., Jahr, E., Eikeseth, S., Hastings, R. P., & Hughes, C. J. (2010). Cognitive and adaptive behavior outcomes of behavioral intervention for young children with intellectual disability. *Behavior Modification, 34*, 16–34. https://doi.org/10.1177/0145445509351961

Emerson, E., & Hatton, C. (2009). Socioeconomic position, poverty,and family research. In L. M. Glidden & M. M. Seltzer (Eds.), *International review of research in mental retardation* (Vol. 37, pp. 95–130). St. Mary's City, MD: Elsevier.

Emerson, E., Hatton, C., Llewellyn, G., Blacher, J., & Graham, H. (2006). Socio-economic position, household composition, health status and indicators of the well-being of mothers of children with and without intellectual disabilities. *Journal of Intellectual Disability Research, 50*, 862–873. https://doi.org/10.1111/j.1365-2788.2006.00900.x

Evans, G. W., & Kim, P. (2013). Childhood poverty, chronic stress, self-regulation, and coping. *Child Development Perspectives, 7*, 43–48. https://doi.org/10.1111/cdep.12013

Feniger-Schaal, R., & Joels, T. (2018). Attachment quality of children with ID and its link to maternal sensitivity and structuring. *Reseach in Developmental Disabilities, 76*, 56–64. https://doi.org/10.1016/j.ridd.2018.03.004

Feniger-Schaal, R., Oppenheim, D., Koren-Karie, N., & Yirmiya, N. (2012). Parenting and intellectual disability: An attachment perspective. In J. A. Burack, R. Hodapp, G. Iarocci, & E. Zigler (Eds.), *The Oxford handbook of intellectual disability and development* (pp. 334–348). New York, NY: Oxford University Press.

Fenning, R. M., & Baker, J. K. (2012). Mother-child interaction and resilience in children with early developmental risk. *Journal of Family Psychology, 26*, 411–420. https://doi.org/10.1037/aa0028287

Fidler, D. J., Daunhauer, L., Will, E., Gerlach-McDonald, B., & Schworer, E. (2016). The central role of etiology in science and practice in intellectual disability. In R. M. Hodapp & D. J. Fidler (Eds.), *International Review of Research in Developmental Disabilities* (Vol. 50, pp. 33–70). Cambridge, MA: Academic Press/Elsevier.

Fixsen, D., Blase, K., Metz, A., & Van Dyke, M. (2013). Statewide implementation of evidence-based programs. *Exceptional Children, 79*, 213–230. https://doi.org/10.1177/001440291307900206

Fleming, J. L., Sawyer, L. B., & Campbell, P. H. (2011). Early intervention providers' perspectives about implementing participation-based practices. *Topics in Early Childhood Special Education, 30*, 233–244. https://doi.org/10.1177/0271121410371986

Fox, S. E., Levitt, P., & Nelson, C. A., III. (2010). How the timing and quality of early experiences influences the development of brain architecture. *Child Development, 81*, 28–40. Retrieved from http://www.jstor.org/stable/40598964

Gallimore, R., Weisner, T. S., Bernheimer, L. P., Guthrie, D., & Nihira, K. (1993). Family responses to young children with developmental delays: Accommodation activity in ecological and cultural context. *American Journal on Mental Retardation, 98*, 185–206.

Gerstein, E. D., Crnic, K. A., Blacher, J., & Baker, B. L. (2009). Resilience and the course of daily parenting stress in families of young children with intellectual disabilities. *Journal of Intellectual Disability Research, 53*, 981–997. PMCID: PMC279238. https://doi.org/10.1111/j.1365-2788.2009.01220.x

Gilliam, W. S., Meisels, S., & Mayes, L. (2005). Screening and surveillance in early intervention systems. In M. J. Guralnick (Ed.), *A developmental systems approach to early intervention: National and international perspectives* (pp. 73–98). Baltimore, MD: Brookes Publishing Co.

Gilmore, L., Cuskelly, M., Jobling, A., & Hayes, A. (2009). Maternal support for autonomy: Relationships with persistence for children with Down syndrome and typically developing children. *Research in Developmental Disabilities, 30*, 1023–1033. https://doi.org/10.1016/j.ridd.2009.02.005

Green, S. A., Caplan, B., & Baker, B. (2014). Maternal supportive and interfering control as predictors of adaptive and social development in children with and without developmental delays. *Journal of Intellectual Disability Research, 58*, 691–703. https://doi.org/10.1111/jir.12064

Guralnick, M. J. (Ed.). (1978). *Early intervention and the integration of handicapped and nonhandicapped preschool children*. Baltimore, MD: University Park Press.

Guralnick, M. J. (Ed.). (1997). *The effectiveness of early intervention*. Baltimore, MD: Brookes Publishing Co.

Guralnick, M. J. (1999). Family and child influences on the peer-related social competence of young children with developmental delays. *Mental Retardation and Developmental Disabilities Research Reviews, 5*, 21–29. https://doi.org/10.1002/(SICI)1098-2779(1999)5:1<21::AID-MRDD3>3.0.CO;2-O

Guralnick, M. J. (2001a). A developmental systems model for early intervention. *Infants & Young Children, 14*, 1–18.

Guralnick, M. J. (2001b). A framework for change in early childhood inclusion. In M. J. Guralnick (Ed.), *Early childhood inclusion: Focus on change* (pp. 3–35). Baltimore, MD: Brookes Publishing.

Guralnick, M. J. (Ed.). (2001c). *Early childhood inclusion: Focus on change*. Baltimore, MD: Brookes Publishing.

Guralnick, M. J. (2005). Early intervention for children with intellectual disabilities: Current knowledge and future prospects. *Journal of Applied Research in Intellectual Disabilities, 18*, 313–324. https://doi.org/10.1111/j.1468-3148.2005.00270.x

Guralnick, M. J. (2008). International perspectives on early intervention: A search for common ground. *Journal of Early Intervention, 30*, 90–101. https://doi.org/10.1177/1053815107313483

Guralnick, M. J. (2010). Early intervention approaches to enhance the peer-related social competence of young children with developmental delays: A historical perspective. *Infants & Young Children, 23*, 73–83. PMCID: PMC2880512. https://doi.org/10.1097/IYC.0b013e3181d22e14

Guralnick, M. J. (2011). Why early intervention works: A systems perspective. *Infants & Young Children, 24*, 6–28. https://doi.org/10.1097/IYC.0b013e3182002cfe

Guralnick, M. J. (2012). Preventive interventions for preterm children: Effectiveness and developmental mechanisms. *Journal of Developmental and Behavioral Pediatrics, 33*, 352–364. https://doi.org/10.1097/DBP.0b013e31824eaa3c

Guralnick, M. J. (2017a). Early intervention for children with intellectual disabilities: An update. *Journal of Applied Research in Intellectual Disabilities, 30*, 211–229. https://doi.org/10.1111/jar.12233

Guralnick, M. J. (2017b). Early intervention for young children with developmental delays: Contributions of the Developmental Systems Approach. In H. Sukkar, J. Kirby, & C. Dunst (Eds.), *Early childhood intervention: Working with families of young children with special needs* (pp. 17–35). Oxon, UK: Routledge.

Guralnick, M. J. (2019a). *Effective Early Intervention: The Developmental Systems Approach*. Baltimore, MD: Brookes Publishing Co.

Guralnick, M. J. (2019b). Implementation science, the Developmental Systems Approach, and family-centered practices. In C. Escorcia & L. Rodriguez (Eds.), *Family-centered practices in early intervention* (pp. 51–71). UNED: Madrid, Spain.

Guralnick, M. J., & Bruder, M. B. (2016). Early childhood inclusion in the United States: Goals, current status, and future directions. *Infants & Young Children, 29*, 166–177. https://doi.org/10.1097/IYC.0000000000000071

Guralnick, M. J., Connor, R. T., Hammond, M., Gottman, J. M., & Kinnish, K. (1996). Immediate effects of mainstreamed settings on the social interactions and social integration of preschool children. *American Journal on Mental Retardation, 100*, 359–377. Retrieved from http://www.ncbi.nlm.nih.gov/entrez/query.fcgi?cmd=Retrieve&db=PubMed&dopt=Citation&list_uids=8718991

Guralnick, M. J., Connor, R. T., Neville, B., & Hammond, M. A. (2006). Promoting the peer-related social development of young children with mild developmental delays: Effectiveness of a comprehensive intervention. *American Journal on Mental Retardation, 111*, 336–356. https://doi.org/10.1352/0895-8017(2006)111[336:PTPSDO]2.0.CO;2

Guralnick, M. J., Connor, R. T., Neville, B., & Hammond, M. A. (2008). Developmentally delayed children's influence attempts with mothers predict interactions with peers over time. *Journal of Applied Developmental Psychology, 29*, 238–248. https://doi.org/10.1016/j.appdev.2008.02.002

Guralnick, M. J., Gottman, J. M., & Hammond, M. A. (1996). Effects of social setting on the friendship formation of young children differing in developmental status. *Journal of Applied Developmental Psychology, 17*, 625–651.

Guralnick, M. J., Hammond, M. A., Neville, B., & Connor, R. T. (2008). The relationship between sources and functions of social support and dimensions of child- and parent-related stress. *Journal of Intellectual Disability Research, 52*, 1138–1154. PMCID: PMC2585608. https://doi.org/10.1111/j.1365-2788.2008.01073.x

Guralnick, M. J., Neville, B., Hammond, M. A., & Connor, R. T. (2007). Linkages between delayed children's social interactions with mothers and peers. *Child Development, 78*, 459–473. https://doi.org/10.1111/j.1467-8624.2007.01009x

Guralnick, M. J., Neville, B., Hammond, M. A., & Connor, R. T. (2008). Mothers' social communicative adjustments to young children with mild developmental delays. *American Journal on Mental Retardation, 113*, 1–18. https://doi.org/10.1352/0895-8017(2008)113[1:MSCATY]2.0.CO;2

Halfon, N., Houtrow, A., Larson, K., & Newacheck, P. W. (2012). The changing landscape of disability in childhood. *The Future of Children, 22*, 13–42. http://www.jstor.org/stable/41475645

Halle, T., Metz, A., & Martinez-Beck, I. (Eds.). (2013). *Applying implementation science in early childhood programs and systems*. Baltimore, MD: Brookes Publishing.

Hart, B., & Risley, T. (1975). Incidental teaching of language in the preschool. *Journal of Applied Behavior Analysis, 8*, 411–420.

Hauser-Cram, P., Warfield, M. E., Shonkoff, J. P., Krauss, M. W., Sayer, A., & Upshur, C. C. (2001). Children with disabilities: A longitudinal study of child development and parent well-being. *Monographs of the*

Society for Research in Child Development, 66, 1–114. https://doi.org/10.1111/1540-5834.00154

Hebbler, K., Greer, M., & Hutton, B. (2011). From then to now: The evolution of Part C. *Zero to Three, 31*, 4–10.

Individuals with Disabilities Education Improvement Act. (2004). *Pub. L. No., 108*, 446.

Justice, L. M., Logan, J. A. R., Lin, T.-J., & Kaderavek, J. N. (2014). Peer effects in early childhood education: Testing the assumptions of special-education inclusion. *Psychological Science, 25*, 1722–1729. https://doi.org/10.1177/0956797614538978

Kaiser, A. P., & Roberts, M. Y. (2013). Parent-implemented enhanced milieu teaching with preschool children who have intellectual disabilities. *Journal of Speech, Language, and Hearing Research, 56*, 295–309. https://doi.org/10.1044/1092-4388(2012/11-0243)

Karaaslan, O., Diken, I. H., & Mahoney, G. (2013). A randomized control study of responsive teaching with young Turkish children and their mothers. *Topics in Early Childhood and Special Education, 33*, 18–27. https://doi.org/10.1177/0271121411429749

Kemp, P., & Turnbull, A. P. (2014). Coaching with parents in early intervention. *Infants & Young Children, 27*, 305–324. https://doi.org/10.1097/IYC.0000000000000018

Keogh, B. K., Bernheimer, L. P., & Guthrie, D. (1997). Stability and change over time in cognitive level of children with delays. *American Journal on Mental Retardation, 101*, 365–373. Retrieved from http://www.ncbi.nlm.nih.gov/entrez/query.fcgi?cmd=Retrieve&db=PubMed&dopt=Citation&list_uids=9017083

Kersh, J., Hedvat, T. T., Hauser-Cram, P., & Warfield, M. E. (2006). The contribution of marital quality to the well-being of parents of children with developmental disabilities. *Journal of Intellectual Disability Research, 50*, 883–893. https://doi.org/10.1111/j.1365-2788.2006.00906.x

Kim, J.-M., & Mahoney, G. (2005). The effects of relationship focused intervention on Korean parents and their young children with disabilities. *Research in Developmental Disabilities, 26*, 117–130. https://doi.org/10.1016/j.ridd.2004.08.001

King, G., Strachan, D., Tucker, M., Duwyn, B., Desserud, S., & Shillington, M. (2009). The application of a transdisciplinary model for early intervention services. *Infants & Young Children, 22*, 211–223. https://doi.org/10.1097/IYC.0b013e3181abe1c3

Kover, S. T., McCary, L. M., Ingram, A. M., Hatton, D. D., & Roberts, J. E. (2015). Language development in infants and toddlers with fragile X syndrome: Change over time and the role of attention. *American Journal on Intellectual and Developmental Disabilities, 120*, 125–144. https://doi.org/10.1352/1944-7558-120.2.125

Landau, B. (2012). The organization and development of spatial representation: Insights from Williams syndrome. In J. A. Burack, R. M. Hodapp, G. Iarocci, & E. Zigler (Eds.), *The Oxford handbook of intellectual disability and development* (pp. 61–88). New York, NY: Oxford University Press.

Lobo, M. A., Paul, D. A., Mackley, A., Maher, J., & Galloway, J. C. (2014). Instability of delay classification and determination of early intervention eligibility in the first two years of life. *Research in Developmental Disabilities, 35*, 117–126. https://doi.org/10.1016/j.ridd.2013.10.017

Macy, M., Thorndike-Christ, T., & Lin, Y. (2010). Parental reports of perceived assessment utility: A comparison of authentic and conventional approaches. *Infants & Young Children, 23*, 286–302. https://doi.org/10.1097/IYC.0b013e3181f1ec1b

Mahoney, G., Perales, F., Wiggers, B., & Herman, B. (2006). Responsive teaching: Early intervention for children with Down syndrome and other disabilities. *Down Syndrome*. *Research and Practice, 11*, 18–28. https://doi.org/10.3104/perspectives.311

Mangin, K. S., Horwood, L. J., & Woodward, L. J. (2017). Cognitive development trajectories of very preterm and typically developing children. *Child Development, 88*, 282–298. https://doi.org/10.1111/cdev.12585

McConnell, S., & Rahn, N. L. (2016). Assessment in early childhood special education. In B. Reichow, B. Boyd, E. E. Barton, & S. L. Odom (Eds.), *Handbook of early childhood special education*. New York, NY: Springer. https://doi.org/10.1007/978-3-319-28492-7_3pp.89-106

McDuffie, A., Oakes, A., Machalicek, W., Ma, M., Bullard, L., Nelson, S., & Abbeduto, L. (2016). Early language intervention using distance videoteleconferencing: A pilot study of young boys with Fragile X syndrome and their mothers. *American Journal of Speech-Language Pathology, 25*, 46–66. https://doi.org/10.1044/2015_AJSLP-14-0137

McLean, M., Wolery, M., & Bailey, D. B. (2003). *Assessing infants and preschoolers with special needs* (3rd ed.). Columbus, OH: Merrill/Prentice Hall.

McWilliam, R. A. (2010a). Assessing families' needs with the Routines-Based Interview. In R. A. McWilliam (Ed.), *Working with families of young children with special needs* (pp. 27–59). New York, NY: Guilford Press.

McWilliam, R. A. (Ed.). (2010b). *Working with families of young children with special needs*. New York, NY: Guilford Press.

McWilliam, R. A. (2016). Birth to three: Early intervention. In B. Reichow, B. A. Boyd, E. E. Barton, & S. L. Odom (Eds.), *Handbook of early childhood special education* (pp. 75–88). Switzerland: Springer.

Metz, A., Halle, T., Bartley, L., & Blasberg, A. (2013). The key components of successful implementation. In T. Halle, A. Metz, & I. Martinez-Beck (Eds.), *Applying implementation science in early childhood programs and systems* (pp. 21–42). Baltimore, MD: Brookes.

Minnes, P., Perry, A., & Weiss, J. A. (2015). Predictors of distress and well-being in parents of young children with developmental delays and disabilities: The importance of parent perceptions. *Journal of Intellectual Disability Research, 59*, 551–560. https://doi.org/10.1111/jir.12160

Most, D. E., Fidler, D. J., Booth-LaForce, C., & Kelly, J. (2006). Stress trajectories in mothers of young children with Down syndrome. *Journal of Intellectual Disability Research, 50*, 501–514. https://doi.org/10.1111/j.1365-2788.2006.00796.x

Neece, C. L., Green, S. A., & Baker, B. L. (2012). Parenting stress and child behavior problems: A transactional relationship across time. *American Journal on Intellectual and Developmental Disabilities, 117*, 48–66. https://doi.org/10.1352/1944-7558-117.1.48

Nes, R. B., Røysamb, E., Hauge, L. J., Kornstad, T., Landolt, M. A., Irgens, L. M., . . . Vollrath, M. E. (2014). Adaptation to the birth of a child with a congenital anomaly: A prospective longitudinal study of maternal well-being and psychological distress. *Developmental Psychology, 50*, 1827–1839. https://doi.org/10.1037/a0035996

Odom, S. L., Buysse, V., & Soukakou, E. P. (2011). Inclusion for young children with disabilities: A quarter century of research perspectives. *Journal of Early Intervention, 33*, 344–356. https://doi.org/10.1177/1053815111430094

Olsson, M. B., & Hwang, C. P. (2008). Socioeconomic and psychological variables as risk and protective factors for parental well-being in families of children with intellectual disabilities. *Journal of Intellectual Disability Research, 52*, 1102–1113. https://doi.org/10.1111/j.1365-2788.2008.01081.x

Peer, J. W., & Hillman, S. B. (2014). Stress and resilience for parents of children with intellectual and developmental disabilities. *Journal of Policy and Practice in Intellectual Disabilities, 11*, 92–98. https://doi.org/10.1111/jppi.12072

Peterson, C. A., Luze, G. J., Eshbaugh, E. M., Jeon, H. J., & Ross Kantz, K. (2007). Enhancing parent-child interactions through home visiting: Promising practice or unfulfilled promise? *Journal of Early Intervention, 29*, 119–140. https://doi.org/10.1177/105381510702900205

Phillips, D. A., & Meloy, M. E. (2012). High-quality school-based pre-K can boost early learning for children with special needs. *Exceptional Children, 78*, 471–490. https://doi.org/10.1177/001440291207800405

Raab, M., Dunst, C. J., & Hamby, D. W. (2016). Effectiveness of contrasting approaches to response-contingent learning among children with significant developmental delays and disabilities. *Research and Practice for Persons with Severe Disabilities, 41*, 36–51. https://doi.org/10.1177/1540796915621189

Raab, M., Dunst, C. J., & Hamby, D. W. (2017). Efficacy trial of contrasting approaches to the response-contingent learning of young children with significant developmental delays and multiple disabilities. *Journal of Educational and Developmental Psychology, 7*, 12–28. https://doi.org/10.5539/jedp.v7n1p12

Roberts, J. E., Hatton, D. D., & Bailey, D. B. (2001). Development and behavior of male toddlers with Fragile X syndrome. *Journal of Early Intervention, 24*, 207–223. https://doi.org/10.1177/10538151010240030601

Robinson, M., Klusek, J., Poe, M. D., Hatton, D. D., & Roberts, J. E. (2018). The emergence of effortful control in young boys with fragile X syndrome. *American Journal on Intellectual and Developmental Disabilities, 123*, 89–102. https://doi.org/10.1352/1944-7558-123.2.89

Romski, M. A., Sevcik, R. A., Adamson, L. B., Cheslock, M., Smith, A. L., Barker, R. M., & Bakeman, R. (2010). Randomized comparison of augmented and nonaugmented language interventions for toddlers with developmental delays and their parents. *Journal of Speech, Language, and Hearing Research, 53*, 350–364. https://doi.org/10.1044/1092-4388(2009/08-0156)

Rosenberg, S. A., Ellison, M. C., Fast, B., Robinson, C. C., & Lazar, R. (2013). Computing theoretical rates of Part C eligibility based on developmental delays. *Maternal and Child Health Journal, 17*, 384–390. https://doi.org/10.1007/s10995-012-0982-2

Sandall, S. R., Schwartz, I. S., & Gauvreau, A. G. (2016). Using modifications and accommodations to enhance learning of young children with disabilities: Little changes that yield big impacts. In B. Reichow, B. Boyd, E. E. Barton, & S. L. Odom (Eds.), *Handbook of early childhood special education* (pp. 349–361). New York, NY: Springer.

Scarborough, A. A., Spiker, D., Mallik, S., Hebbeler, K. M., Bailey, D. B., & Simeonsson, R. J. (2004). A national look at children and families entering early intervention. *Exceptional Children, 70*, 469–483. https://doi.org/10.1177/001440290407000406

Shapiro, B. K., & Batshaw, M. L. (2013). Developmental delay and intellectual disability. In M. L. Batshaw, N. J. Roizen, & G. R. Lotrecchiano (Eds.), *Children with Disabilities* (7th ed., pp. 291–306). Baltimore, MD: Brookes Publishing Co.

Shelden, M. L., & Rush, D. D. (2010). In R. A. McWilliam (Ed.),. *Working with families of young children with special needs A primary-coach approach to teaming and supports in early childhood intervention* (pp. 175–202). New York, NY: Guilford Press.

Shevell, M. I., Majnemer, A., Rosenbaum, P., & Abrahamowicz, M. (2001). Profile of referrals for early childhood developmental delay to ambulatory subspecialty clinics. *Journal of Child Neurology, 16*, 645–650. https://doi.org/10.1177/088307380101600904

Snyder, P., Hemmeter, M. L., McLean, M., Sandall, S. R., & McLaughlin, T. (2013). Embedded instruction to support early learning in response-to-intervention frameworks. In V. Buysse & E. Peisner-Feinberg (Eds.), *Handbook of response-to-intervention in early childhood* (pp. 283–298). Baltimore, MD: Brookes Publishing Co.

Snyder, P., Rakap, S., Hemmeter, M. L., & McLaughlin, T. W. (2015). Naturalistic instructional approaches in early learning: A systematic review. *Journal of Early Intervention, 37*, 69–97. https://doi.org/10.1177/1053815115595461

Spiker, D., Boyce, G. C., & Boyce, L. K. (2002). Parent-child interactions when young children have disabilities. In L. M. Glidden (Ed.), *International Review of Research in Mental Retardation* (pp. 35–70). San Diego, CA: Academic Press.

Spiker, D., & Hopmann, M. R. (1997). The effectiveness of early intervention for children with Down syndrome. In M. J. Guralnick (Ed.), *The effectiveness of early intervention* (pp. 271–305). Baltimore, MD: Brookes Publishing Co.

Sterling, A. M., Barnum, L., Skinner, D., Warren, S. F., & Fleming, K. (2012). Parenting young children with and without Fragile X syndrome. *American Journal on Intellectual and Developmental Disabilities, 117*, 194–206. https://doi.org/10.1352/1944-7558-117.3.194

Thiemann-Bourque, K. S., Warren, S. F., Brady, N., Gilkerson, J., & Richards, J. A. (2014). Vocal interaction between children with Down syndrome and their parents. *Journal of Speech-Language Pathology, 23*, 474–485. PMCID: 4047176. https://doi.org/10.1044/2014_AJSLP-12-0010

Trivette, C. M. (2003). Influence of caregiver responsiveness on the development of young children with or at risk for developmental disabilities. *Winterberry Research Syntheses, 1*, 1–15. Retrieved from http://www.wbpress.com/shop/influence-of-caregiver-responsiveness-on-the-development-of-young-children-with-or-at-risk-for-developmental-disabilities/

U.S. Department of Education. (2017). *39th Annual Report to Congress on the Implementation of the Individuals with Disabilities Education Act, 2017*: p. 330.

United Cerebral Palsy National Collaborative Infant Project. (1976). *Staff development handbook: A resource for the transdisciplinary process*. New York, NY: Author.

Vail, C. O., Lieberman-Betz, R. G., & McCorkle, L. S. (2018). The impact of funding on Part C systems: Is the tail wagging the dog? *Journal of Early Intervention, 40*, 229–245. https://doi.org/10.1177/1053815118771388

Venuti, P., de Falco, S., Esposito, G., & Bornstein, M. H. (2009). Mother-child play: Children with Down syndrome and typical development. *American Journal on Intellectual and Developmental Disabilities, 114*, 274–288. https://doi.org/10.1352/1944-7558-114.4

Warren, S. F., Brady, N., Sterling, A., Fleming, K., & Marquis, J. (2010). Maternal responsivity predicts language development in young children with Fragile X syndrome. *American Journal on Intellectual and Developmental Disabilities, 115*, 54–75. https://doi.org/10.1352/1944-7558-115.1.54

Warren, S. F., & Brady, N. C. (2007). The role of maternal responsivity in the development of children with intellectual disabilities. *Mental Retardation and Developmental Disabilities Research Reviews, 13*, 330–338. https://doi.org/10.1002/mrdd.20177

Weiland, C. (2016). Impacts of the Boston prekindergarten program on the school readiness of young children with special needs. *Developmental Psychology, 52*, 1763–1776. https://doi.org/10.1037/dev0000168

Williams, W., & Gotts, E. A. (1977). Selected considerations on developing curriculum for severely handicapped students. In E. Sontag, J. Smith, & N. Certo (Eds.), *Educational programming for the severely and profoundly handicapped* (pp. 221–236). Reston, VA: Division on Mental Retardation, The Council for Exceptional Children.

Winton, P. J. (2016). Taking stock and moving forward: Implementing quality early childhood inclusive practices. In B. Reichow, B. Boyd, E. E. Barton, & S. L. Odom (Eds.), *Handbook of early childhood special education* (pp. 57–74). New York, NY: Springer.

Wishart, J. G. (1996). Avoidant learning styles and cognitive development in young children. In B. Stratford & P. Gunn (Eds.), *New approaches to Down syndrome* (pp. 173–205). London, UK: Cassell.

Wolery, M. (2000). Recommended practices in child-focused interventions. In S. Sandall, M. E. McLean, & B. J. Smith (Eds.), *DEC recommended practices in early intervention/early special education* (pp. 29–37). Longmont, CO: Sopris West.

Woodman, A. C., Mawdsley, H. P., & Hauser-Cram, P. (2015). Parenting stress and child behavior problems within families of children with developmental disabiities: Transactional relations across 15 years. *Research in Developmental Disabilities, 36*, 264–276. https://doi.org/10.1016/j.ridd.2014.10.011

Woods, J. J., Kashinath, S., & Goldstein, H. (2004). Effects of embedding caregiver-implemented teaching strategies in daily routines on children's communication outcomes. *Journal of Early Intervention, 26*, 175–193.

Woods, J. J., & Snyder, P. (2009). Interdisciplinary doctoral leadership training in early intervention: Considerations for research and practice in the 21st century. *Infants & Young Children, 22*, 32–43. https://doi.org/10.1097/01.IYC.000034335.26904.db

Wright, C. A., & Kaiser, A. P. (2017). Teaching parents enhanced milieu teaching with words and signs using the Teach-Model-Coach-Review model. *Topics in Early Childhood and Special Education, 36*, 192–204. https://doi.org/10.1177/0271121415621027

Wright, C. A., Kaiser, A. P., Reikowsky, D. I., & Roberts, M. Y. (2013). Effects of a naturalistic sign intervention on expressive language of toddlers with Down syndrome. *Journal of Speech, Language, and Hearing Research, 56*, 994–1008. https://doi.org/10.1044/1092-4388(2012/12-0060)

Yang, P., Jong, Y.-J., Hsu, H.-Y., & Lung, F.-W. (2011). Role of assessment tests in the stability of intelligence scoring of pre-school children with uneven/delayed cognitive profile. *Journal of Intellectual Disability Research, 55*, 453–461. https://doi.org/10.1111/j.1365-2788.2011.01396.x

Yockelson, S., Linder, C., & Asman, R. (2015). A collaborative approach to early identification and referral of children who are in family childcare settings, birth to five, born to teenage mothers. *Journal of Intellectual Disability Diagnosis and Treatment, 3*, 205–212. https://doi.org/10.6000/2292-2598.2015.03.04.6

Yoder, P., & Warren, S. F. (1999). Maternal responsivity mediates the relationship between prelinguistic intentional communication and later language. *Journal* *of Early Intervention, 22*, 126–136. https://doi.org/10.1177/105381519902200205

Yoder, P., & Warren, S. F. (2001). Relative treatment effects of two prelinguistic communication interventions on language development in toddlers with developmental delays vary by maternal characteristics. *Journal of Speech, Language, and Hearing Research, 44*, 224–237. https://doi.org/10.1044/1092-4388(2001/019)

Zampini, L., Salvi, A., & D'Odorico, L. (2015). Joint attention behaviours and vocabulary development in children with Down syndrome. *Journal of Intellectual Disability Research, 59*, 891–901. https://doi.org/10.1111/jir.12191

Zaslow, M. (2009). Strengthening the conceptualization of early childhood professional development initiatives and evaluations. *Early Education & Development, 20*, 1–10. https://doi.org/10.1080/10409280902908833

Zeedyk, S. M., & Blacher, J. (2017). Longitudinal correlates of maternal depression among mothers of children with or without intellectual disability. *American Journal on Intellectual and Developmental Disabilities, 122*, 374–394. https://doi.org/10.1352/1944-7558-122.5.374

40

Parenting Support

Trevor G. Mazzucchelli, Lienke Wilker,
and Matthew R. Sanders

Of all the potentially modifiable environmental risk and protective factors that can meaningfully influence children's development, none is more important than the quality of parenting children receive. The influence of parenting on children's development is pervasive, affecting aspects as diverse as the child's cognition, language, emotion regulation, social skills, peer relationships, and physical and mental health (Sanders & Mazzucchelli, 2018b). This chapter explores the challenges faced by families raising a child with an intellectual disability to provide the context and a rationale for the provision of parenting support to parents and carers of children with an intellectual disability. A brief history of parent support in the disability field is provided before a range of contemporary evidence-based parenting support programs are introduced. There is considerable evidence supporting the efficacy and effectiveness of parenting interventions based on social learning theory principles; however, these interventions reach too few families. We present an argument for a population-based approach to parenting support for families with a child who has a disability. We present a model for what such an approach might look like, evidence to show such an approach is both feasible and effective, and describe challenges and future directions for the field.

Challenges Faced by Families Raising a Child with an Intellectual Disability

Parents report both benefits and burdens from raising a child with an intellectual disability. For instance, parents can report personal benefits such as becoming a better person—more compassionate, less selfish, more tolerant—as well as a sense of satisfaction and accomplishment at having done one's best (Hastings & Taunt, 2002). On the other hand, parents also report additional burdens. Raising a child can be a demanding and emotional task for any parent; however, parents raising a child with an intellectual disability are often faced with a range of additional unique and

---

T. G. Mazzucchelli
Child and Family Research Group, and Brain, Behaviour and Mental Health Research Group, Curtin University, Perth, WA, Australia

Parenting and Family Support Centre, School of Psychology, The University of Queensland, Brisbane, QLD, Australia
e-mail: trevor.mazzucchelli@curtin.edu.au

L. Wilker
Child and Family Research Group, and Brain, Behaviour and Mental Health Research Group, Curtin University, Perth, WA, Australia
e-mail: lienke.wilker@postgrad.curtin.edu.au

M. R. Sanders (✉)
Parenting and Family Support Centre, School of Psychology, The University of Queensland, Brisbane, QLD, Australia
e-mail: m.sanders@psy.uq.edu.au

J. L. Matson (ed.), *Handbook of Intellectual Disabilities*, Autism and Child Psychopathology Series, https://doi.org/10.1007/978-3-030-20843-1_40

emotionally demanding challenges (Mazzucchelli & Studman, 2018).

Children with an intellectual disability typically require care and support beyond that provided to typically developing children. By definition, children with intellectual disability present with high support needs in adaptive behavior and functional life activities (Harries, Guscia, Nettelbeck, & Kirby, 2009; Schalock et al., 2010). These children are at greater risk of having other comorbid disabilities such as physical and sensory disabilities and chronic health issues such as seizure disorders which can further complicate parenting (Kapell et al., 1998; Orelove, Sobsey, & Silberman, 2004). Additionally, children with an intellectual disability are three to four times more likely than their typically developing counterparts to develop significant emotional and behavioral problems, including severe and sometimes life-threatening behavior problems such as aggression and self-injurious behavior (Einfeld, Ellis, & Emerson, 2011; Lang et al., 2013).

Severe behavior problems can have serious consequences for the child, their parents, and other family members. They can threaten the physical health of children (Borthwick-Duffy, 1994; Nissen & Haveman, 1997), negatively impact the child's learning at school and peer relationships (Taanila, Ebeling, Heikura, & Jarvelin, 2003), restrict children's access to recreational and educational programs (Parmenter, Einfeld, Tonge, & Dempster, 1998), lead to exclusion from community settings (Borthwick-Duffy & Eyman, 1990; Borthwick-Duffy, Eyman, & White, 1987), and reduce occupational opportunities in the post-school period (Anderson, Lakin, Hill, & Chen, 1992). Behavior problems are also one of the main predictors of parental distress (McIntyre, Blacher, & Baker, 2006; Plant & Sanders, 2007), use of respite services (Chan & Sigafoos, 2001; Sloper, Knussen, Turner, & Cunningham, 1991), and parents relinquishing the care of their child (Nankervis, Rosewarne, & Vassos, 2011).

Even in the absence of severe behavior problems, daily hassles related to providing care can adversely affect a family's ability to cope (Crnic & Greenberg, 1990; Plant & Sanders, 2007). Parents of children with intellectual disability are at greater risk compared to parents of typically developing children of reporting high levels of stress, anxiety, and depression (Herring et al., 2006; Singer, 2006) as well as poorer psychological well-being (Barlow, Cullen-Powell, & Cheshire, 2006; Eisenhower, Baker, & Blacher, 2005). Parents often experience grief when they learn that their child has a disability, and the process of adapting to disability can be prolonged in nature (Eakes, Burke, & Hainsworth, 1998; Rentinck, Ketelaar, Jongmans, & Gorter, 2007). Parents often report difficulty finding respite and specialized childcare for their child (McConkey, Kelly, & Craig, 2011) and experience financial stress due to their reduced capacity to maintain full-time employment due to childcare responsibilities as well as the cost of therapy and medical expenses (e.g., Lin et al., 2013; Parish, Seltzer, Greenberg, & Floyd, 2004). Research also suggests that parents of children with intellectual disability are at increased risk for relationship distress (Kersh, Hedvat, Hauser-Cram, & Warfield, 2006; Risdal & Singer, 2004) and divorce (Hartley et al., 2010).

While individual biological and psychological factors may place children with an intellectual disability at increased risk for developing behavioral and emotional problems, there is strong evidence to suggest that these factors interact with social and environmental factors to precipitate and perpetuate problem behavior (Oliver, Demetriades, & Hall, 2002; Tonge & Einfeld, 2003). In particular, coercive parenting practices, emotional adjustment difficulties of carers, and relationship conflict between caregivers have been implicated in the development and maintenance of the emotional and behavioral problems of children with developmental disabilities (Chadwick, Kusel, & Cuddy, 2008; Koskentausta et al., 2007). Studies indicate that children with developmental disabilities are more likely to be exposed to such risk factors than their nondisabled peers (Green & Baker, 2011; Stalker & McArthur, 2012). Further, a bidirectional relationship has been found between parenting stress and child behavior problems: high parenting stress contributing to a worsening in child behavior problems over time and child behavior problems exacerbating parental stress (Neece, Green, & Baker, 2012; Woodman et al., 2015).

In summary, parents of children with intellectual disability are often faced with challenges greater than those faced by parents of typically developing children but have fewer supports and resources available to address those challenges. These parents are at greater risk of experiencing high levels of stress, and a reciprocal relationship can occur between parenting stress and child behavior problems.

Rationale for Parenting Interventions

Because of the unique needs of children with intellectual disability, their increased risk of developing behavioral and emotional problems, and the severe negative impact of such problems, there is widespread recognition of the importance of prevention and early intervention (McIntyre et al., 2006; Sanders & Mazzucchelli, 2018b). In tandem with this is the recognition that parents of these children have an increased need for support and specifically request information on how to interact with their children to promote their children's development and manage problem behavior (Bailey Jr. et al., 1999; Hamilton, Mazzucchelli, & Sanders, 2015).

Parents have a powerful influence on children's development through the day-to-day, moment-to-moment interactions that take place between themselves and their children (Dunst et al., 2001; Sanders & Mazzucchelli, 2018b). Through their interactions, parents can teach, prompt, and encourage their child's behavior to promote their acquisition of adaptive skills. They can also arrange the situational context and respond to their child's behavior in ways that decrease stressful parent-child interactions and the development of behavior problems (e.g., Buschbacher, Fox, & Clarke, 2004; Clarke, Dunlap, & Vaughn, 1999). Assisting parents to acquire these parenting skills is also likely to enhance parents' self-efficacy, an important element of self-regulation. Parental self-efficacy refers to a parent's belief about their capacity to successfully undertake their parenting responsibilities. When a parent has high levels of self-efficacy, they are more likely to persist with a task and achieve their parenting goals. They are also more likely to report less stress and higher levels of well-being (Sanders & Mazzucchelli, 2013).

Parenting practices, parental adjustment, and family relationships are obvious foci for supporting parents and families. Parenting support has the potential to enhance these important functions thereby addressing a range of factors that may positively impact children's development and family adjustment.

History of Parenting Support in Disability

Early Social History Surrounding Parenting Support

The emergence of parent training has been closely associated with societal change. Until the twentieth century, children with intellectual disabilities were separated from their families and were forced to live in asylums (Braddock & Parish, 2002). This institutionalization was enforced by government mental healthcare acts. Punitive methods of behavioral management were used by custodians, and no serious efforts were made to advance the development of these children (Nehring & Lindsey, 2016). Harsh attitudes found within institutions were reflective of prevailing societal attitudes. Disparaging views of intellectual disability peaked in the 1940s as competition for resources increased during the Second World War. Government-sanctioned massacres were completed in European countries, and an estimated 250,000 adults and children deemed to be "useless eaters" were killed, which included those with intellectual disabilities (Cocks, 1998).

Following the atrocities of the Second World War, global attitudes shifted to having increased tolerance toward others. Progress was made in providing support for people with intellectual disabilities (Braddock & Parish, 2002). In the 1940s and 1950s, there was rapid growth of charitable organizations consisting primarily of parents and family members of children with disabilities (Cocks, 1998). These groups advocated against the removal and maltreatment of their children as well as for better standards of service. This was known as the parent movement, and these associations created alternative residential and educational opportunities for children with intellectual disabilities (Davis, 2016).

The promotion of the civil rights movement in the 1950s and 1960s highlighted the difficulties experienced by minority groups, which contributed to the betterment of social practices concerning people with disabilities, prompting the disability rights movement (Braddock & Parish, 2002). Another considerable influence was concept of normalization proffered in the 1960s by Bengt Nirje, which was widely disseminated by Wolf Wolfensberger in the 1970s (Wolfensberger, Nirje, Olshansky, Perske, & Roos, 1972). The idea of normalization proposed that individuals with a disability should have access to opportunities akin to that of persons without a disability. The notion of normalization in combination with the other movements prompted global systemic change through enabling legislative reforms (Davis, 2016). These advancements in disability rights led to the complete deinstitutionalization of children with an intellectual disability (Nehring & Lindsey, 2016). Alongside this came a great need for interventions for building parental skills (Kazdin 2008).

Theoretical Principles Underlying Parenting Support

A direct influence on the formation of parenting support was the research on operant conditioning (Kazdin, 2008). Skinner suggested that the frequency that a behavior is emitted can be influenced by ensuing environmental consequences. After investigating behavioral modification with animals, Skinner described operant processes based on the type of stimulus change operation that immediately follows a behavior and the effect that operation has on the future frequency of that behavior. These processes are broadly categorized as either reinforcement or punishment (Skinner, 1938). Reinforcement occurs when a stimulus change (e.g., praise and touch) immediately follows a behavior (e.g., following an instruction) and increases the future frequency of that behavior in similar conditions. Punishment occurs when a stimulus change (e.g., a reprimand) follows a behavior (e.g., hitting) and decreases the future frequency of that behavior. Reinforcement is critical for increasing desirable behavior and promoting skill development (Kazdin, 2013; Mazzucchelli, 2018; Skinner, 1953). However, reinforcement alone may not be sufficient for managing severe behavior problems. In these cases, mild, effective, disciplinary consequences may be warranted to produce change (Morawska & Sanders, 2011).

A significant theoretical extension of operant conditioning was Albert Bandura's social learning theory. Bandura (1977) argued behavioral theories did not fully explain the development of behavior and posited that learning also occurs through observation. This theory was incorporated into parent training programs through techniques encouraging parents to model desired behaviors, so that children might learn to enact those behaviors through observation (McIntyre & Neece, 2016; Sanders & Mazzucchelli, 2018a). Later strategies derived from behavior modification principles gave increased attention to the antecedents of behavior, for example, how instructions are delivered as well as the settings in which they are given. The three fundamental constituents of forming behavioral responses are the antecedent stimulus (A), the behavior (B), and the consequence (C), also known as the ABCs of behavior (Kazdin, 2013; Skinner, 1953). The impact of consequences is influenced by motivating operations (Michael, 1982). Motivating operations refer to antecedent events or changes in the environment that alter the effects of consequences on behavior. For example, having recently engaged in exercise, a drink may be strongly reinforcing (Simó-Pinatella et al., 2012).

Initial Research on Parenting Support

Beginning in the 1960s, research on parent training strategies was a response to the lack of effective interventions for children who presented with behavior problems. Psychodynamic approaches had been commonly used but did not result in reliable and generalized reductions in problematic behavior (Eyberg, 1988). A number of psychologists began conducting single group designs and descriptive case studies researching new models of parent training, which promoted parents as agents of change, equipping them with knowledge of behavioral principles. For instance, Jensen and Womack (1967) taught operant conditioning techniques to a mother with a 6-year-old child with autism spectrum disorder. This intervention resulted in substantial decreases in the number of tantrums and disruptive behaviors emitted by the child. Findings from such studies indicated that using operant principles was effective in reducing problematic behaviors. These insights contributed to the founding of the first structured behavioral parent training interventions (Shaffer, Kotchick, Dorsey, & Forehand, 2001).

Parent training programs targeted deficits in parenting skills which were maintaining challenging behaviors. Improvements in parenting practices aimed to strengthen prosocial behaviors of their children, such as following instructions, and to weaken antisocial behaviors such as tantrums (Forehand, Kotchick, Shaffer, & McKee, 2010). An influential model of parenting support was developed by Constance Hanf at the University of Oregon in the 1960s and early 1970s. Hanf's work occurred contemporaneously to that of Gerald Patterson and his colleagues (also in Oregon) whose research program led to what is now known as Parent Management Training - Oregon Model (PMTO). In comparison to PMTO, Hanf's model placed greater importance on the parent-child relationship and was designed as an early intervention for use with younger children (aged 3–8 years). Hanf's work was initially undertaken with children with a developmental disability and their families, and the resulting model formed the basis of many contemporary parenting support programs (Reitman & McMahon, 2013).

In the 1970s and 1980s, the primary objective of parent training research was to investigate the generalization of treatment effects and long-term outcomes (Forehand et al., 2010). Sanders and Plant (1989) investigated the generalization of treatment of effects of parent training with children who had developmental disabilities. Their findings indicated that parents were able to successfully implement behavioral management strategies across environments other than the training setting when provided instruction in antecedent/preemptive parenting techniques. It is now recognized that the effects of parent training often generalize across settings, over time, to other siblings who are not the focus of treatment and to other behaviors which were not targeted (Shaffer et al., 2001).

Contemporary Parenting Support Programs

This section will provide a description of contemporary, empirically supported, parenting support programs for families with children who have a disability.

The Portage Project

The Portage Project is an early intervention program which aims to provide parents with education on behavioral principles via home visits to support the development of children with an intellectual disability. The program was originally designed for children with disabilities from birth to 6 years of age as well as their family (Shearer & Shearer, 1976). The New Portage Guide, which is the current version of the program, begins with a skill assessment of parents' ability to manage difficult behaviors and the child's level of functioning. Parents are then trained to teach their children new skills, which are progressively completed based on the milestones achieved by children up to the age of six. Each week has a different goal, and specific instructions are provided to parents in terms of how to teach their child the new skill and how to record the progress of the child. Home visits by health professionals are intermittently incorporated to assist parents in appropriately delivering the behavioral strategies (Baker, Marquis, & Feinfield, 2016).

The Portage program has been widely disseminated and applied seemingly effectively across diverse populations and cultures, as it is used in over 60 different countries with successful modification of behaviors of children with a disability being reported (Baker et al., 2016). An evaluation of anecdotal and empirical results from studies assessing the effectiveness of the Portage Project program found positive results being reported by the professionals and parents involved in delivering the intervention (Brue & Oakland, 2001). However, it must be noted that there are only a small number of quantitative studies with high methodological rigor which assess for the Portage Project's effectiveness, meaning that further research is needed (Brue & Oakland, 2001).

Parent-Child Interaction Therapy

Parent-child interaction therapy (PCIT) is a behavioral approach which was developed to address challenging behaviors of children aged 2–7 years (Eyberg, 1988). The objective of the treatment is to establish a warm relationship between a parent and a child as well as to change problematic parent-child interactional sequences through the medium of play. Disruptive behaviors are decreased, and prosocial behaviors are increased through the improvements made in the quality of the relationship and by resolving coercive interactional styles that serve to maintain these behaviors (Eyberg, 1988). The therapist provides psychoeducation, role-plays warm interactions, and coaches the parent by giving immediate and specific feedback on the parent-child interaction occurring during play with the child (McNeil & Hembree-Kigin, 2010). Parents are encouraged to avoid using any commands, instructions, and criticisms, which would most likely threaten the warm relationship. Parents are taught skills to deliver verbal directions calmly and clearly and to apply appropriate consequences to target behaviors in ways which the child can understand (Eyberg, 1988).

Bagner and Eyberg (2007) conducted the first randomized controlled trial which specifically examined the efficacy of PCIT with 30 families of children aged 3–6 years with an intellectual disability. The parenting intervention significantly reduced child disruptive behavior and parental stress compared to those in the waitlist condition. Furthermore, it was reported that children in the intervention condition were more compliant post-intervention and that improvements were also observed in parent-child relationships (Bagner & Eyberg, 2007).

A recent case study documented the use of PCIT combined with visual supports with a 5-year-old girl who had an intellectual disability and comorbid epilepsy and autism spectrum disorder (Armstrong, DeLoatche, Preece, & Agazzi, 2014). Improvements in behavioral functioning were reported across home, school, and community settings providing further evidence that the treatment effects of PCIT can generalize across settings. Although there is a significant body of research on PCIT with typically developing children, few studies with high methodological rigor have specifically examined PCIT for use with children with developmental disabilities, and further investigation is required (Armstrong et al., 2014).

The Incredible Years Program

The Incredible Years program is an early intervention parent training program which aims to reduce problematic behaviors of children. The Incredible Years parenting program consists of a suite of programs developed for different age groups, including babies, toddlers, pre-primary schoolers, and primary school aged. The program is delivered in a group format with parents in 12–20 weekly sessions. Trained group facilitators use techniques such as role-play, didactic instruction, video modeling, and discussion. These techniques are used to cover topics such as rewards, strengthening the parent-child relationship, reducing punitive punishments, play, boundary setting, and managing disruptive behaviors. Problematic behaviors of children are reduced through changing negative, coercive, parenting styles to more positive, noncoercive, parenting styles. Adjustments are made to the Incredible Years program when being delivered to parents of children with disabilities, such as including discussions on the unique blessings and challenges faced by families that have children with developmental delays (McIntyre & Neece, 2016).

Adapted versions of Incredible Years have been consistently found to yield significant reductions in disruptive behaviors emitted by children with an intellectual or developmental disability (Kleve et al., 2011; McIntyre, 2008a, 2008b; Phaneuf & McIntyre, 2011). This is consistent with the effectiveness of Incredible Years when used with parents of typically developing children (Menting, de Castro, & Matthys, 2013).

Signposts for Building Better Behavior

Signposts is a multilevel parent training intervention which has been developed specifically for parents or caregivers of youth, aged 3 to 16 years of age, with a developmental delay or intellectual disability (Hudson et al., 2003). The primary aims of the program are to assist parents to implement strategies to manage difficult child behavior, to promote prosocial behavior, and to teach these children new coping skills (Hudson et al., 2003). The program is prevention orientated, with a stated goal being to prevent the escalation of disruptive behavior before the severity rises to a clinical level and requires expensive, specialized interventions (Hudson, Cameron, & Matthews, 2008).

The standard program delivery of Signposts comprises six fortnightly sessions with a 4-week break between session five and six (McIntyre & Neece, 2016). Signposts can be delivered in a group format, individual face-to-face, or in a self-directed fashion with telephone support (Hudson et al., 2008). Each session has a duration of 2 h and 30 min and is delivered by a trained health professional (McIntyre & Neece, 2016). Parents are trained in using strategies such as recording behaviors, modifying interactional patterns, planning to avoid cueing disruptive behaviors, and other behavioral approaches to reduce problem behaviors. The strategies are based on common scenarios which occur within family settings of children with disabilities (McIntyre & Neece, 2016). A small number of studies have found support for the effectiveness of Signposts. In the foundational evaluation, the parents of 115 families who participated in the program reported lower levels of stress, increased efficacy at managing disruptive behavior, and increases in desired behaviors in their children (Hudson et al., 2003). Findings have also indicated that the effectiveness of the intervention is enhanced through fathers' involvement in the program (May et al., 2013). Furthermore, factors such as age, gender, and associated disability have not been found to moderate the treatment outcomes for children with intellectual disabilities or their families (Hudson, Reece, Cameron, & Matthews, 2009).

A Population Approach

Research findings from numerous studies have demonstrated that parenting interventions based on social learning theory, functional analysis, and cognitive behavioral principles result in increased parental competence and confidence in raising their child with a developmental disability, reductions in child emotional and behavioral problems, and significant reductions in parent stress (Hudson et al., 2009; Ruane & Carr, 2018; Tellegen & Sanders, 2013). Unfortunately, despite the strength of the evidence, still too few parents who might benefit from evidence-based parenting support (EBPS) actually participate. Traditional clinical models of service delivery typically reach few families, and the small proportion (13%) of parents that do access parenting programs do not access EBPS (Einfeld et al., 2018a; Sanders et al., 1999). Despite the fact that parenting groups are widely advocated in the parent training field, parents of children with a disability believe that a range of delivery modalities are likely to be useful (e.g., group, individual, seminar, web-based, over the telephone), and parents differ in opinion as to which modalities are likely to be most useful (Mazzucchelli & Moran, 2017). These findings confirm that contemporary parents are looking for alternative, flexible ways of accessing parenting advice (Sanders et al., 1999). The consequence of low program availability, limited program options, and nonoptimal participation rates is inadequate program reach.

Limited program reach means that most families who could benefit from parenting programs do not access them. When relatively few families derive the benefits of EBPS, the potential of these programs to reduce the prevalence of problematic outcomes for children in the entire population is markedly weakened. Effective dissemination of EBPS is critical for programs to have any significant community impact. Of the parent training programs that have been disseminated to date, many have been delivered late in the developmental trajectory as interventions for children already showing signs of challenging behavior rather than as prevention programs (Mazzucchelli & Sanders, 2011).

A population approach to parenting support involves making EBPS available to all who choose to access such support. This usually requires the program to be offered across multiple sites and by many different practitioners (e.g., in disability, education, and health sectors and nongovernment organizations). It also requires that the parenting program can be delivered in different intensities and through different modalities. The benefits of a population approach rather than an approach which targets families who are identified as experiencing difficulties are many. If programs are offered to every family, then participating is normalized, and any stigma that might occur is eliminated. When programs can be accessed through multiple agencies and in a variety of modalities and intensities, then barriers to attendance are reduced, and parents can choose to access as much or as little support as they require at a time (Mazzucchelli & Sanders, 2011).

We suggest that a population approach that addresses the broader ecological context of parenting is required to ensure that more parents can access parenting programs and to achieve a population-level reduction in the prevalence of social, emotional, and behavioral problems in children with developmental disability (Mazzucchelli & Sanders, 2011).

The Stepping Stones Triple P System

Stepping Stones Triple P (SSTP; Mazzucchelli & Studman, 2018; Sanders, Mazzucchelli, & Studman, 2004) is one of the few examples of a population-based approach to parenting for families with a child who has a developmental disability. Parallel to the core Triple P—Positive Parenting Program (see Sanders & Prinz, 2018), SSTP was developed as a comprehensive system of parenting and family support to improve the quality of parenting advice available to parents of children with a disability. The system aims to promote children's development and prevent severe social and emotional problems and child maltreatment, by enhancing the knowledge skills and confidence of parents. It incorporates five levels of intervention on a tiered continuum of increasing strength for parents of children from birth to age 12. The multilevel suite of programs is designed to create a "family friendly" environment that supports parents in the task of raising their children (see Table 40.1). It specifically targets the social contexts that influence parents on a day-to-day basis. These contexts include primary healthcare services, childcare and school systems, disability support organizations, and mental health services. The multilevel strategy is designed to maximize efficiency, to avoid waste and overservicing, and to ensure that the program has wide reach in the community (see Fig. 40.1). It also allows for choice in the intensity and delivery modality to match programs to families' needs and preferences.

Table 40.1 The Stepping Stones Triple P system of parenting and family support

Level of intervention	Target population	Intervention methods	Facilitators
Level 1 Communications strategy • *Universal Triple P* • *Stay Positive*	All parents interested in information about parenting and promoting their child's development	Coordinated communications strategy raising awareness of parent issues and encouraging participation in parenting programs. May involve electronic and print media (e.g., brochures, posters, websites, television, talk-back radio, newspaper and magazine editorials)	Typically coordinated by communications, health, or welfare staff members
Level 2 Health promotion strategy/brief selective intervention • *Selected Stepping Stones Triple P*	Parents of children with a disability who are interested in parenting education or with specific concerns about their child's development or behavior	Health promotion information or specific advice for a discrete developmental issue or minor child behavior problem. May involve a group seminar format or brief (up to 20 min) telephone or face-to-face clinician contact	Practitioners who provide parent support during routine well-child healthcare (e.g., health, education, allied health, and childcare staff) or disability early intervention services (e.g., therapy or psychology staff)
Level 3 Narrow-focus parent training • Primary Care Stepping Stones Triple P • Stepping Stones Triple P Discussion Groups	Parents of children with disability who have specific concerns, as above, who require consultations or active skills training	Brief program (about 80 min over 4 sessions, or 2 h discussion groups) combining advice, rehearsal, and self-evaluation to teach parents to manage a discrete child problem behavior. May involve telephone or face-to-face clinician contact or group sessions	Same as for Level 2
Level 4 Broad-focus parent training • Standard Stepping Stones Triple P • Group Stepping Stones Triple P • Self-Directed Stepping Stones Triple P	Parents of children with a disability who want intensive training in positive parenting skills. Typically, parents of children who want to promote their children's development or with behavior problems such as aggressive or oppositional behavior	Broad-focus program (about 9–16 h over 9–10 sessions) focusing on parent-child interaction and the application of parenting skills to a broad range of target behaviors seen in a disability context. Includes generalization enhancement strategies. May be self-directed or involve telephone or face-to-face clinician contact or group sessions	Intensive parenting interventions (e.g., disability, mental health, and welfare staff and other allied health and education professionals who regularly consult with parents about child behavior)
Level 5 Intensive family intervention modules • Enhanced Triple P	Parents of children with behavior problems and concurrent family dysfunction (e.g., parental depression or stress) or conflict between partners	Intensive individually tailored program with modules (sessions last 60–90 min) including practice sessions to enhance parenting skills, mood management and stress coping skills, and partner support skills	Intensive family intervention work (e.g., disability, mental health, and welfare staff)
• Pathways Triple P	Parents at risk of maltreating their children. Program targets anger management problems and other factors associated with abuse	Intensive individually tailored or group program with modules (60–120 min sessions) depending on delivery model including attribution retraining and anger management	Same as above
• Group Lifestyle Triple P	Parents of overweight or obese children. Targets healthy eating and increasing activity levels, as well as general child behavior	Intensive 14-session group program (including telephone consultations) focusing on nutrition, healthy lifestyle, and general parenting strategies. Includes generalization enhancement strategies	As above, plus dieticians /nutritionists with experience in delivering parenting interventions
• Family Transitions Triple P	Parents going through separation or divorce	Intensive 12-session group program (including telephone consultations) focusing on coping skills, conflict management, general parenting strategies, and developing a healthy co-parenting relationship	Intensive family intervention work (e.g., disability, mental health, and welfare staff)

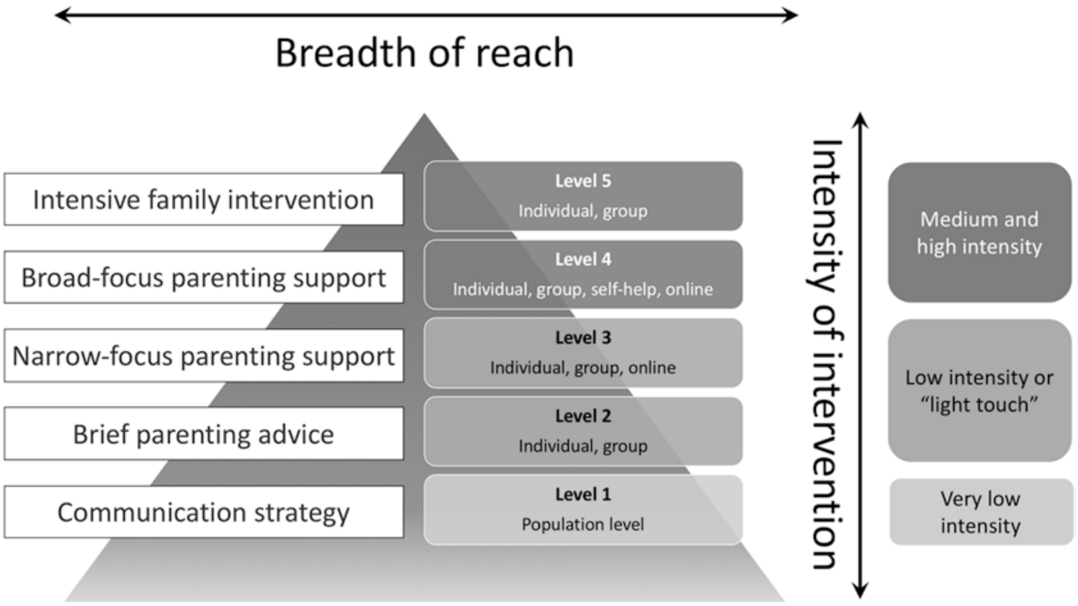


Fig. 40.1 The Triple P model of graded reach and intensity of parenting and family support services

The seven core positive parenting principles that form the basis of the program were selected to address specific risk and protective factors known to predict positive developmental and mental health outcomes in children with a disability (see Table 40.2). Table 40.3 shows how the principles are operationalized into a range of specific parenting skills; see Mazzucchelli and Sanders (2012) and Mazzucchelli and Studman (2018) for a more complete overview and description of the different interventions that make up the system.

The Stepping Stones Triple P system has been featured in two meta-analyses that have drawn from 16 RCTs and 3 uncontrolled trials that have evaluated individual interventions that make up the SSTP system (Ruane & Carr, 2018; Tellegen & Sanders, 2013). Both meta-analyses concluded that the interventions are effective in improving outcomes for children and parents. Statistically and clinically significant effects for all delivery format and program variants have been found for children's emotional and behavioral outcomes; parenting practices; parenting satisfaction and efficacy; parental adjustment; the parental couple relationship; and observed child and parent behavior (Ruane & Carr, 2018; Tellegen & Sanders, 2013). The positive outcomes demonstrated for each of the Stepping Stones Triple P levels provide support for a multilevel, multidisciplinary system of parenting programs, including prevention and treatment options, to increase timely access to cost-effective services promoting child, parent, and family well-being.

A population-level trial of the SSTP system involving the simultaneous delivery of all levels of the SSTP system has recently been undertaken across three states of Australia (Sofronoff, Gray, Einfeld, & Tonge, 2018). Preliminary results from this trial indicate that this approach increases access to parenting support for parents of children with a disability with 38% of the target population participating in a program. Furthermore, it results in significant improvements in a range of indices of family functioning including reductions in parental stress; reductions in negative parenting styles such as parental inconsistency, coercive parenting, and lack of positive encouragement; and decreases in child behavioral and emotional problems (Einfeld et al., 2018a; Gray et al., 2017; Sofronoff et al., 2018). In addition, the population trial was found to be cost-effective; when the cost of establishing and implementing SSTP was calculated and compared with the value of cost savings, the SSTP system

Table 40.2 Principles of positive parenting children with a disability

Principle	Description
A safe and engaging environment	Children of all ages need a safe, supervised, and therefore protective environment that provides opportunities for them to explore, experiment, and play. This principle is essential to promote healthy development and to prevent accidents and injuries in the home
A positive learning environment	This involves educating parents in their role as their child's first teacher and specifically teaching parents to respond positively and constructively to child-initiated interactions (e.g., requests for help, information, advice, and attention) through incidental teaching and other techniques to assist children to learn how to solve problems for themselves
Assertive and consistent discipline	Stepping Stones Triple P teaches parents specific child management and behavior change strategies that are alternatives to coercive and ineffective discipline practices (e.g., shouting, threatening, or using physical punishment). The aim is to develop predictable and consistent responses and avoid factors that may maintain problem behavior (e.g., accidental rewards)
Adapting to having a child with a disability	This involves parents coming to terms with their child's disability, finding a balance between the demands and stresses of parenting and the resources they have to cope, and staying optimistic about the future. Parents may need to acknowledge any sense of grief or sense of loss they feel. They may also find it helpful to decrease demands and stressors, increase their coping resources, and find personal meaning and a sense of control
Realistic expectations	This involves exploring with parents their expectations, assumptions, and beliefs about the causes of children's behavior and choosing goals that are developmentally appropriate for the child and realistic for the parents. Parents who are at risk of abusing their children are more likely to have unrealistic expectations of children's capabilities
Being part of the community	Every individual with a disability has a right to a full and rewarding life that includes employment or meaningful activities throughout the day, living independently in the community, a meaningful social life with friends and family members, and recreation, hobbies, and leisure time. Creating this life begins in childhood by families regularly using facilities and participating in activities in the community
Taking care of oneself as a parent	Parenting is affected by a range of factors that impact on a parent's self-esteem and sense of well-being. All levels of Stepping Stones Triple P specifically address this issue by encouraging parents to view parenting as part of a larger context of personal self-care, resourcefulness, and well-being and by teaching parents practical parenting skills

was found to save each family AUD$574 per year. This was primarily because of increased capacity of parents to return to work (Einfeld et al., 2018a). These results suggest that a population-level approach holds great promise for increasing the reach of EBPPs and having a positive impact on the well-being of all children with a disability and their families. In the next sections, we will outline some of the issues and challenges in implementing such an approach.

Issues and Challenges in Providing Parenting Support

Increasing Reach and Uptake

Despite parenting programs having been around for decades, only a minority of parents of children with disabilities participate in them, even though they have three to four times the level of behavioral and emotional problems of typically developing children. Part of the rationale behind developing a multilevel system of parenting support has been to ensure that parents are able to participate in programs more suited to their preferences, interest, and needs. Strategies from population health need to be employed to social market programs to create "pull" or consumer demand (e.g., Wilkinson, 2018). This will help reduce stigma and to challenge a view that parenting programs are for parents who are struggling.

Engaging Fathers

The vast majority of parents who participate in parenting intervention studies are mothers (Panter-Brick et al., 2014). The under-presentation of fathers in both research and clinical practice

Table 40.3 Core parenting skills introduced in Stepping Stones Triple P

Safe and engaging environment	Positive learning environment	Assertive discipline	Adapting to having a child with a disability	Realistic expectations	Being part of the community	Parental self-care
• Spending brief quality time • Communicating with children • Showing affection • Providing engaging activities	• Giving nonverbal attention • Giving descriptive praise • Giving other rewards • Setting a good example • Using physical guidance • Using incidental teaching • Using ask-say-do • Teaching backward • Using behavior charts	• Using diversion to another activity • Establishing ground rules • Using directed discussion • Using planned ignoring • Giving clear, calm instructions • Teaching children to communicate what they want • Using logical consequences • Using blocking • Using brief interruption • Using quiet time • Using time-out	• Improving personal communication habits • Giving and receiving constructive feedback • Having casual conversations • Supporting each other when problem behavior occurs • Problem-solving • Improving relationship happiness • Getting personal and professional support • Having a break and using respite services • Maintaining optimism	• Monitoring children's behavior • Setting developmentally appropriate goals • Setting practice tasks • Self-evaluating strengths and weaknesses • Setting personal goals for change	• Planning and advanced preparation • Discussing ground rules for specific situations • Selecting engaging activities • Providing incentives • Providing consequences • Holding follow-up discussions	• Catching unhelpful thoughts • Using relaxation and stress management • Developing personal coping statements • Challenging unhelpful thoughts • Developing coping plans for high-risk situations

can be addressed by ensuring program outreach emphasizes the importance of father participation, by training practitioners to identify the preferences, priorities, and needs of fathers and to tailor program delivery so that the needs of mothers and fathers are addressed. There is emerging evidence that programs that tailor delivering to the needs and interests of fathers can be very successful. For example, Frank, Keown, and Sanders (2015) used focus groups and surveys of fathers to identify parenting topics and issues that were concerning to fathers. They then incorporated these findings into program delivery through example selection, emphasis on the importance of the father's relationship with children and their paternal role, and individually tailored homework for fathers and mothers, not just couple homework. Following program completion, intervention group fathers and mothers reported significantly fewer child behavior problems, dysfunctional parenting practices, and interparental conflict about child-rearing than waitlist parents. Intervention group mothers also reported increased parenting confidence and rated their partners as showing significantly fewer dysfunctional parenting practices. Intervention effects were maintained at 6-month follow-up. Both mothers and fathers reported high levels of satisfaction with the program.

Addressing Behavioral Phenotypes

Children with disabilities are not a homogeneous group, and there are a wide range of behavioral phenotypes or syndromes (e.g., fragile X). These children may have unique features (e.g., touch aversion) that require parenting advice to be adapted to take into account the type of condition the child has. The MHYPEDD research team (Einfeld et al., 2018b) developed a range of parent and practitioner tip sheets to assist practitioner adapt principles of positive parenting to the child's unique presentation. Interestingly although these resources were valued by both parents and practitioners, they were rarely used in the rollout of the program.

Practitioner Attitudes and Beliefs

Practitioners across a range of disciplines have a special role in providing timely parenting support in the context of disability, but some practitioners have beliefs about parenting support that may act as a barrier to this. For instance, practitioners may think that parenting support is somehow incompatible with a "family-centered" practice orientation that emphasizes an equal and supportive parent-practitioner relationship. Practitioners might hold the belief that providing instruction/coaching to parents regarding their interactions with children cannot be collaborative (Mahoney et al., 1999). Practitioners may also believe that providing such training may cause parents additional stress, implicitly blame parents, overemphasize parents' role as teachers undermining the parent-child relationship, and be culturally insensitive. These beliefs are not consistent with feedback from parental surveys or the results from trials of EBPS. Good parenting training is not incompatible with family-centered practice; indeed, a supportive and collaborative relationship is a prerequisite to successful parent training (Mazzucchelli & Sanders, 2010).

Staff Turnover

The disability sector has a generally high rate of staff turnover (Hatton et al., 2001). This means that organizations have to continuously invest in training new staff in the delivery of parenting interventions. Low cost on the job training is rarely sufficient for staff to learn how to deliver a parenting intervention and to deliver it with fidelity. Training or purveyor organizations offering training need to develop cost-effective alternatives to intensive 3- to 5-day training programs which may not be offered when an organization needs new staff to be trained. While web-based training that can be accessed 24/7 seems an obvious alternative, there are major challenges in designing online training programs that provide coaching and feedback to practitioners based on their actual performance in a consultation or a simulated one. Until online training courses using artificial intelligence have been shown to be just as effective as in person training, organizations are likely to have to team up with other like organizations to arrange group-based training for new staff. They can manage the supervision process internally using models of supervision such as peer-assisted supervision and support (PASS, McPherson & Schroeter, 2018).

Public Policy

The quality of parenting services available to families of a child with a disability is influenced by the favorability of the broader policy environment and funding priorities of government. For example, in an Australian context although funding is available to support parents' access to individual parenting programs through the National Disability Insurance Agency, group-based programs delivered by organizations are not. This anomaly needs to be addressed as a matter of public policy so parents are able to access programs in a format they are seeking.

Another policy matter that has caused controversy in the disability sector is the use of restrictive practices such as time-out. In some jurisdictions the use of time-out is frowned upon and actually banned in some institutional settings (e.g., childcare, residential care). Misuse of time-out as a punitive measure in a negative environment is very different from parental use of time-out in a highly reinforcing home environment. Also, reports in major news outlets (Time Magazine) claiming time-out damages children's brains have been sensationalized and led to calls to abandon an extremely useful intervention for certain behavior problems (Siegel & Bryson, 2014).

These political considerations point to the ongoing importance of policy advocacy on the basis of evidence to ensure that parents can access programs that work.

Future Directions

The field of EBPS for parents of children with a disability continues to evolve as more studies demonstrate that both children and parents benefit from these interventions. In line with the parenting field generally, there is now a much more diverse range of delivery modalities that have been tested including the use of online parenting programs combined with social media, self-help programs, and low-intensity primary care interventions (Hinton, Sheffield, Sanders, & Sofronoff, 2017; Tellegen & Sanders, 2014).

Although there have been relative few cost-effective studies, a recent independent economic analysis of the Stepping Stone Triple P system showed that it was extremely cost-effective, saving participating families approximately $574 per year primarily because of less missed days of work per year (Einfeld et al., 2018a). Other economic analyses of parenting interventions based on social learning principles with typically developing children have consistently shown them to be cost-effective (Sampaio, Feldman, Richards-Jones, & Mihalopoulos, 2018).

Parenting intervention has primarily focused on challenging behavior in younger children; however, parents continue to influence the well-being of children in every subsequent phase of development. Recently, Stepping Stones Triple P has been extended to support parents of adolescents (Building Bridges Triple P). Pilot evaluation data shows promising positive impact on adolescent behavior problems, parenting practices, and parental confidence and emotional adjustment; however, future randomized trials are needed to establish a sufficient evidence base to justify dissemination (Mazzucchelli, Jenkins, & Sofronoff, 2018).

Grandparents play an important role in the lives of many children with a disability and where they have frequent childcare responsibilities, it makes sense to involve the grandparent in parenting programs. Several recent randomized trials involving both noncustodial and custodial grandparents showed when grandparents were offered Group Triple P (either the standard program or an adapted version specifically for grandparents), there were positive effects on grandparent well-being and parent-grandparent communication, and improvements in grandchildren's behavior (Smith et al., 2018; Kirby & Sanders, 2014). Similar studies are needed with grandparents of children with a disability.

Conclusion

Evidence-based parenting support has evolved from intensive home-based individual and group programs many of which were originally developed for typically developing children with conduct problems to a comprehensive population-based, trans-diagnostic, multilevel system of parenting support (Sanders & Mazzucchelli, 2018c). As with other parents, it is highly likely that offering parenting support to parents of children with a developmental disability as a "one size fits all" will be unlikely to meet the diverse needs of parents. The field should avoid an "inoculation" approach which provides programs in early childhood but ignores the parenting challenges at other stages of the life cycle (elementary school age, adolescent, young adulthood, and beyond). There is little work to date addressing the needs of parents of adult children with a disability. Substantially more research is needed to examine the effects of current program variants with more culturally diverse families. Research is also needed to find ways to engage socioeconomically disadvantaged parents and support them to successfully complete programs. Where parenting programs tailored to the needs of parents with special needs are made available as part of a population-based parenting support strategy for all parents, completing a parenting program will become free of stigma, routine, expected, not exceptional, and a normative rite of passage for all parents.

Take-Home Messages

- Parenting programs based on social learning theory, functional analysis, and cognitive behavioral principles result in increased parental competence and confidence in raising their child with a developmental disability, reductions in child emotional and behavioral problems, and significant reductions in parent stress.
- Evidence-based parenting support should be accessible to all parents of children with a disability.
- A population-level approach holds great promise for increasing the reach of evidence-based parenting programs and having a positive impact on the well-being of all children with a disability and their families.
- Parenting support should be accessible for parents at each phase of the child's life span.

Disclosure Statement The Parenting and Family Support Centre is partly funded by royalties stemming from published resources of the Triple P—Positive Parenting Program—which is developed and owned by The University of Queensland (UQ). Royalties from the program are also distributed to the Faculty of Health and Behavioural Sciences at UQ and contributory authors of Triple P programs. Triple P International (TPI) Pty Ltd. is a private company licensed by UniQuest, Pty Ltd., a commercialization company of UQ, to publish and disseminate Triple P worldwide. T.G.M. and M.R.S. are employees of, or hold honorary positions at, The University of Queensland. T.G.M. and M.R.S. have received, receive, or may in the future receive royalties and/or consultancy fees from TPI. L.W. has no conflicts to declare.

References

Anderson, D. J., Lakin, K. C., Hill, B. K., & Chen, T.-H. (1992). Social integration of older persons with mental retardation in residential facilities. *American Journal on Mental Retardation, 96*, 488–501.

Armstrong, K., DeLoatche, K. J., Preece, K. K., & Agazzi, H. (2014). Combining parent–child interaction therapy and visual supports for the treatment of challenging behavior in a child with autism and intellectual disabilities and comorbid epilepsy. *Clinical Case Studies, 14*(1), 3–14. https://doi.org/10.1177/1534650114531451.

Bagner, D. M., & Eyberg, S. M. (2007). Parent-child interaction therapy for disruptive behavior in children with mental retardation: A randomized controlled trial. *Journal of Clinical Child and Adolescent Psychology, 36*, 418–429. https://doi.org/10.1080/15374410701448448.

Bailey, D. B., Jr., Skinner, D., Correa, V., Arcia, E., Reyes-Blanes, M. E., Rodriguez, P., . . . Skinner, M. (1999). Needs and supports reported by Latino families of young children with developmental disabilities. American Journal on Mental Retardation, 104(5), 437–451. https://doi.org/10.1352/0895-8017%281999%29104%3C0437:NASRBL%3E2.0.CO;2.

Baker, B. L., Marquis, W. A., & Feinfield, K. A. (2016). Early intervention and parent education. In A. Carr, C. Linehan, G. O'Reilly, P. N. Walsh, & J. McEvoy (Eds.), *Handbook of intellectual disability and clinical psychology practice* (2nd ed., pp. 311–338). Abingdon, England: Routledge.

Bandura, A. (1977). *Social learning theory*. Englewood Cliffs, NJ: Prentice-Hall.

Barlow, J. H., Cullen-Powell, L. A., & Cheshire, A. (2006). Psychological well-being among mothers of children with cerebral palsy. *Early Child Development and Care, 176*, 421–428. https://doi.org/10.1080/0300443042000313403.

Borthwick-Duffy, S. A. (1994). Prevalence of destructive behaviors. In T. Thompson & D. B. Gray (Eds.), *Destructive behavior in developmental disabilities: Diagnosis and treatment* (pp. 3–23). Thousand Oaks, CA: Sage.

Borthwick-Duffy, S. A., & Eyman, R. K. (1990). Who are the dually diagnosed? *American Journal on Mental Retardation, 94*, 586–595.

Borthwick-Duffy, S. A., Eyman, R. K., & White, J. F. (1987). Client characteristics and residential placement patterns. *American Journal of Mental Deficiency, 92*(1), 24–30.

Braddock, D., & Parish, S. (2002). An institutional history of disability. In D. Braddock (Ed.), *Disability in the dawn of the 21st century and the state of the states* (pp. 11–68). Washington, DC: AAMR.

Brue, A. W., & Oakland, T. (2001). The Portage guide to early intervention: An evaluation of published evidence. *School Psychology International, 22*, 243–252. https://doi.org/10.1177/0143034301223001.

Buschbacher, P., Fox, L., & Clarke, S. (2004). Recapturing desired family routines: A parent-professional behavioral collaboration. *Research and Practice for Persons with Severe Disabilities, 29*, 25–39. https://doi.org/10.2511/rpsd.29.1.25.

Chadwick, O., Kusel, Y., & Cuddy, M. (2008). Factors associated with the risk of behaviour problems in adolescents with severe intellectual disabilities. *Journal of Intellectual Disability Research, 52*, 864–876. https://doi.org/10.1111/j.1365-2788.2008.01102.x.

Chan, J. B., & Sigafoos, J. (2001). Does respite care reduce parental stress in families with developmentally disabled children? *Child and Youth Care Forum, 30*, 253–263.

Clarke, S., Dunlap, G., & Vaughn, B. (1999). Family-centered, assessment-based intervention to improve behavior during an early morning routine. *Journal of Positive Behavior Interventions, 1*, 235–241. https://doi.org/10.1177/109830079900100406.

Cocks, E. (1998). *An introduction to intellectual disability in Australia* (3rd ed.). Fyshwick, Australia: Australian Institute on Intellectual Disability.

Crnic, K. A., & Greenberg, M. T. (1990). Minor parenting stresses with young children. *Child Development, 61*, 1628–1637. https://doi.org/10.2307/1130770.

Davis, R. W. (2016). Service and care in Australia. In I. L. Rubin, J. Merrick, D. E. Greydanus, & D. Patel (Eds.), *Health care for people with intellectual and developmental disabilities across the lifespan* (pp. 571–579). New York, NY: Springer.

Dunst, C. J., Bruder, M. B., Trivette, C. M., Hamby, D., Raab, M., & McLean, M. (2001). Characteristics and consequences of everyday natural learning opportunities. *Topics in Early Childhood Special Education, 21*, 68–92. https://doi.org/10.1177/027112140102100202.

Eakes, G. G., Burke, M. L., & Hainsworth, M. A. (1998). Middle-range theory of chronic sorrow. *Image Journal of Nursing Scholarship, 30*, 179–184.

Einfeld, S. L., Ellis, L. A., & Emerson, E. (2011). Comorbidity of intellectual disability and mental disorder in children and adolescents: A systematic review. *Journal of Intellectual and Developmental Disability, 36*, 137–143. https://doi.org/10.1080/13668250.2011.572548.

Einfeld, S. L., Sanders, M., Tonge, B., Gray, K. M., Sofronoff, K., & The MHYPEDD Team. (2018a). *Is statewide delivery of Stepping Stones Triple P effective?* Sydney, Australia: The University of Sydney.

Einfeld, S. L., Sanders, M., Tonge, B., Gray, K. M., Sofronoff, K., & The MHYPEDD Team. (2018b). *Supplementary parent and practitioners tipsheet resources for Stepping Stones practitioners*. Sydney, Australia: The University of Sydney. triplep-parenting.net/provider.

Eisenhower, A. S., Baker, B. L., & Blacher, J. (2005). Preschool children with intellectual disability: Syndrome specificity, behaviour problems, and maternal well-being. *Journal of Intellectual Disability Research, 49*, 657–671. https://doi.org/10.1111/j.1365-2788.2005.00699.x.

Eyberg, S. (1988). Parent-child interaction therapy: Integration of traditional and behavioral concerns. *Child & Family Behavior Therapy, 10*, 33–46. https://doi.org/10.1300/J019v10n01_04.

Forehand, R. A., Kotchick, B., Shaffer, A., & McKee, L. G. (2010). Parent management training. In I. B.

Weiner & W. E. Craighead (Eds.), *The Corsini encyclopedia of psychology*. New York, NY: Wiley.

Frank, T. J., Keown, L. J., & Sanders, M. R. (2015). Enhancing father engagement and interparental teamwork in an evidence-based parenting intervention: A randomized-controlled trial of outcomes and processes. *Behavior Therapy, 46*, 749–763. https://doi.org/10.1016/j.beth.2015.05.008.

Gray, K. M., Tonge, B. J., Taffe, J. R., Sanders, M., Sofronoff, K., & Einfeld, S. L. (2017). A community-based parenting intevention for parents of children with a disability: Outcomes in the Australian Stepping Stones Triple P project. *Mental Health Research in Intellectual Disabilities, 10*, 140–141.

Green, S., & Baker, B. (2011). Parents' emotion expression as a predictor of child's social competence: Children with or without intellectual disability. *Journal of Mental Deficiency Research, 55*, 324–338. https://doi.org/10.1111/j.1365-2788.2010.01363.x.

Hamilton, A., Mazzucchelli, T. G., & Sanders, M. R. (2015). Parental and practitioner perspectives on raising an adolescent with a disability: A focus group study. *Disability and Rehabilitation, 37*, 1664–1673. https://doi.org/10.3109/09638288.2014.973969.

Harries, J., Guscia, R., Nettelbeck, T., & Kirby, N. (2009). Impact of additional disabilities on adaptive behavior and support profiles for people with intellectual disabilities. *American Journal on Intellectual and Developmental Disabilities, 114*, 237–253. https://doi.org/10.1352/1944-7558-114.4.237-253.

Hartley, S. L., Barker, E. T., Seltzer, M. M., Floyd, F., Greenberg, J., Orsmond, G., & Bolt, D. (2010). The relative risk and timing of divorce in families of children with an autism spectrum disorder. *Journal of Family Psychology, 24*, 449–457. https://doi.org/10.1037/a0019847.

Hastings, R. P., & Taunt, H. M. (2002). Positive perceptions in families of children with developmental disabilities. *American Journal on Mental Retardation, 107*, 116–127.

Hatton, C., Emerson, E., Rivers, M., Mason, H., Swarbrick, R., Mason, L., et al. (2001). Factors associated with intended staff turnover and job search behaviour in services for people with intellectual disability. *Journal of Intellectual Disability Research, 45*, 258–270. https://doi.org/10.1046/j.1365-2788.2001.00321.x .

Herring, S., Gray, K., Taffe, J., Tonge, B., Sweeney, D., & Einfeld, S. (2006). Behaviour and emotional problems in toddlers with pervasive developmental disorders and developmental delay: Associations with parental mental health and family functioning. *Journal of Intellectual Disability Research, 50*, 874–882. https://doi.org/10.1111/j.1365-2788.2006.00904.x.

Hinton, S., Sheffield, J., Sanders, M. R., & Sofronoff, K. (2017). A randomized controlled trial of a telehealth parenting intervention: A mixed-disability trial. *Research in Developmental Disabilities, 65*, 74–85.

Hudson, A., Cameron, C., & Matthews, J. (2008). The wide-scale implementation of a support program for parents of children with an intellectual disability and difficult behaviour. *Journal of Intellectual and Developmental Disability, 33*, 117–126. https://doi.org/10.1080/13668250802065885.

Hudson, A. M., Matthews, J. M., Gavidia-Payne, S. T., Cameron, C. A., Mildon, R. L., Radler, G. A., & Nankervis, K. L. (2003). Evaluation of an intervention system for parents of children with intellectual disabilities and challenging behaviour. *Journal of Intellectual Disability Research, 47*, 238–239. https://doi.org/10.1046/j.1365-2788.2003.00486.x.

Hudson, A., Reece, J., Cameron, C., & Matthews, J. (2009). Effects of child characteristics on the outcomes of a parent support program. *Journal of Intellectual and Developmental Disability, 34*, 123–132. https://doi.org/10.1080/13668250902850426.

Jensen, G. D., & Womack, M. G. (1967). Operant conditioning techniques applied in the treatment of an autistic child. *American Journal of Orthopsychiatry, 37*, 30–34. https://doi.org/10.1111/j.1939-0025.1967.tb01064.x.

Kapell, D., Nightingale, B., Rodriguez, A., Lee, J. H., Zigman, W. B., & Schupf, N. (1998). Prevalence of chronic medical conditions in adults with mental retardation: Comparison with the general population. *Mental Retardation, 36*, 269–279. https://doi.org/10.1352/0047-6765(1998)036<0269:POCMCI>2.0.CO;2.

Kazdin, A. E. (2008). *Parent management training: Treatment for oppositional, aggressive, and antisocial behavior in children and adolescents*. New York, NY: Oxford University Press.

Kazdin, A. E. (2013). *Behavior modification in applied settings*. Long Grove, IL: Waveland Press.

Kersh, J., Hedvat, T. T., Hauser-Cram, P., & Warfield, M. E. (2006). The contribution of marital quality to the well-being of parents of children with developmental disabilities. *Journal of Intellectual Disability Research, 50*, 883–893. https://doi.org/10.1111/j.1365-2788.2006.00906.x.

Kirby, J. N., & Sanders, M. R. (2014). A randomized controlled trial evaluating a parenting program designed specifically for grandparents. *Behaviour Research and Therapy, 52*, 35–44. https://doi.org/10.1016/j.brat.2013.11.002.

Kleve, L., Crimlisk, S., Shoebridge, P., Greenwood, R., Baker, B., & Mead, B. (2011). Is the Incredible Years programme effective for children with neuro-developmental disorders and for families with social services involvement in the "real world" of community CAMHS? *Clinical Child Psychology and Psychiatry, 16*, 253–264. https://doi.org/10.1177/1359104510366280.

Koskentausta, T., Iivanainen, M., & Almqvist, F. (2007). Risk factors for psychiatric disturbance in children with intellectual disability. *Journal of Intellectual Disability Research, 51*, 43–53. https://doi.org/10.1111/j.1365-2788.2006.00871.x.

Lang, R., Sigafoos, J., van der Meer, L., O'Reilly, M. F., Lancioni, G. E., & Didden, R. (2013). Early signs and early behavioral intervention of challenging behavior. In R. Hastings & J. Rojahn (Eds.), International Review of Research in Developmental Disabilities (Vol. 44, pp. 1–35). Cambridge, MA: Academic Press.

Lin, L.-P., Kuan, C.-Y., Hsu, S.-W., Lee, T.-N., Lai, C.-I., Wu, J.-L., & Lin, J.-D. (2013). Outpatient visits and expenditures for children and adolescents diagnosed with autism spectrum disorders and co-occurring intellectual disability: An analysis of the national health insurance claims data. *Research in Autism Spectrum Disorders, 7*, 1625–1630. https://doi.org/10.1016/j.rasd.2013.09.003.

Mahoney, G., Kaiser, A., Girolametto, L., MacDonald, J., Robinson, C., Safford, P., & Spiker, D. (1999). Parent education in early intervention: A call for a renewed focus. *Topics in Early Childhood Special Education, 19*, 131–140. https://doi.org/10.1177/027112149901900301.

May, F. S., McLean, L. A., Anderson, A., Hudson, A., Camerson, C., & Matthews, J. (2013). Father participation with mothers in the Signposts program: An initial investigation. *Journal of Intellectual and Developmental Disability, 38*, 39–47. https://doi.org/10.3109/13668250.2012.748184.

Mazzucchelli, T. G. (2018). Social learning influences: Modelling, instructions, consequences. In M. R. Sanders & A. Morawska (Eds.), *Handbook of parenting and child development across the lifespan (chapter 4)*. New York, NY: Springer. https://doi.org/10.1007/978-3-319-94598-9_4 .

Mazzucchelli, T. G., Jenkins, M., & Sofronoff, K. (2018). Building Bridges Triple P: Pilot study of a behavioural family intervention for adolescents with autism spectrum disorder. *Research in Developmental Disabilities, 76*, 46–55. https://doi.org/10.1016/j.ridd.2018.02.018.

Mazzucchelli, T. G., & Moran, L. (2017). Carers of adolescents with a disability: Access to, and preferences for, parenting services. Manuscript submitted for publication.

Mazzucchelli, T. G., & Sanders, M. R. (2010). Facilitating practitioner flexibility within an empirically supported intervention: Lessons from a system of parenting support. *Clinical Psychology: Science and Practice, 17*, 238–252. https://doi.org/10.1111/j.1468-2850.2010.01215.x.

Mazzucchelli, T. G., & Sanders, M. R. (2011). Preventing behavioural and emotional problems in children who have a developmental disability: A public health approach. *Research in Developmental Disabilities, 32*, 2148–2156. https://doi.org/10.1016/j.ridd.2011.07.022.

Mazzucchelli, T. G., & Sanders, M. R. (2012). *Stepping Stones Triple P: A population approach to the promotion of competent parenting of children with disability* (Vol. 2). Brisbane, Australia: University of Queensland.

Mazzucchelli, T. G., & Studman, L. J. (2018). Parenting support for children with developmental disability. In M. R. Sanders & T. G. Mazzucchelli (Eds.), *The power of positive parenting: Transforming the lives of children, parents, and communities using the Triple P system* (pp. 121–134). New York, NY: Oxford University Press.

McConkey, R., Kelly, F., & Craig, S. (2011). Access to respite breaks for families who have a relative with intellectual disabilities: A national survey. *Journal of Advanced Nursing, 67*, 1349–1357. https://doi.org/10.1111/j.1365-2648.2010.05586.x.

McIntyre, L. L. (2008a). Adapting Webster-Stratton's Incredible Years parent training for children with developmental delay: Findings from a treatment group only study. *Journal of Intellectual Disability Research, 52*, 1176–1192. https://doi.org/10.1111/j.1365-2788.2008.01108.x.

McIntyre, L. L. (2008b). Parent training for young children with developmental disabilities: Randomized controlled trial. *American Journal on Mental Retardation, 113*, 356–368. https://doi.org/10.1352/2008.113:356-368.

McIntyre, L. L., Blacher, J., & Baker, B. L. (2006). The transition to school: Adaptation in young children with and without intellectual disability. *Journal of Intellectual Disability Research, 50*, 349–361.

McIntyre, L. L., & Neece, C. L. (2016). Parent training. In N. N. Singh (Ed.), *Handbook of evidence-based practices in intellectual and developmental disabilities* (pp. 467–492). New York, NY: Springer.

McNeil, C. B., & Hembree-Kigin, T. L. (2010). *Parent-child interaction therapy* (2nd ed.). New York, NY: Springer Science + Business Media.

McPherson, K. E., & Schroeter, B. (2018). Promoting program fidelity through peer-assisted supervision and support. In M. R. Sanders & T. G. Mazzucchelli (Eds.), *The power of positive parenting: Transforming the lives of children, parents, and communities using the Triple P system* (pp. 405–415). New York, NY: Oxford University Press.

Menting, A. T., de Castro, B. O., & Matthys, W. (2013). Effectiveness of the Incredible Years parent training to modify disruptive and prosocial child behavior: A meta-analytic review. *Clinical Psychology Review, 33*, 901–913. https://doi.org/10.1016/j.cpr.2013.07.006.

Michael, J. (1982). Distinguishing between discriminative and motivational functions of stimuli. *Journal of Experimental Analysis of Behavior, 37*, 149–155. https://doi.org/10.1901/jeab.1982.37-149.

Morawska, A., & Sanders, M. (2011). Parental use of time out revisited: A useful or harmful parenting strategy? *Journal of Child and Family Studies, 20*, 1–8. https://doi.org/10.1007/s10826-010-9371-x.

Nankervis, K. L., Rosewarne, A., & Vassos, M. (2011). Why do families relinquish care? An investigation of the factors that lead to relinquishment into out-of-home respite care. *Journal of Intellectual Disability Research, 55*, 422–433. https://doi.org/10.1111/j.1365-2788.2011.01389.x.

Neece, C. L., Green, S. A., & Baker, B. L. (2012). Parenting stress and child behavior problems: A transactional relationship across time. *American Journal of Intellectual and Developmental Disabilities, 117*(1). https://doi.org/10.1352/1944-7558-117.1.48.

Nehring, W. M., & Lindsey, B. (2016). History of health care for people with intellectual and developmental disability. In I. L. Rubin, J. Merrick, D. E. Greydanus, & D. Patel (Eds.), *Health care for people with intellectual and developmental disabilities across the lifespan* (pp. 33–46). New York, NY: Springer.

Nissen, J. M. J. F., & Haveman, M. J. (1997). Mortality and avoidable death in people with severe self-injurious behaviour: Results of a Dutch study. *Journal of Intellectual Disability Research, 41*, 252–257. https://doi.org/10.1111/j.1365-2788.1997.tb00704.x.

Oliver, C., Demetriades, L., & Hall, S. (2002). Effects of environmental events on smiling and laughing behavior in Angelman syndrome. *American Journal on Mental Retardation, 107*, 194–200. https://doi.org/10.1352/0895-8017(2002)107<0194:EOEEOS>2.0.CO;2.

Orelove, F. P., Sobsey, D., & Silberman, R. (2004). *Educating children with multiple disabilities: A transdisciplinary approach* (4th ed.). Baltimore, MD: Paul H. Brookes.

Panter-Brick, C., Burgess, A., Eggerman, M., McAllister, F., Pruett, K., & Leckman, J. F. (2014). Practitioner Review: Engaging fathers—recommendations for a game change in parenting interventions based on a systematic review of the global evidence. *Journal of Child Psychology and Psychiatry, 55*, 1187–1212. https://doi.org/10.1111/jcpp.12280.

Parish, S. L., Seltzer, M. M., Greenberg, J. S., & Floyd, F. (2004). Economic implications of caregiving at midlife: Comparing parents with and without children who have developmental disabilities. *Mental Retardation, 42*, 413–426. https://doi.org/10.1352/0047-6765%282004%2942%3C413:EIOCAM%3E2.0.CO;2.

Parmenter, T. R., Einfeld, S. L., Tonge, B. J., & Dempster, J. A. (1998). Behavioural and emotional problems in the classroom of children and adolescents with intellectual disability. *Australia and New Zealand Journal of Developmental Disabilities, 23*, 71–77. https://doi.org/10.1080/13668259800033591.

Phaneuf, L., & McIntyre, L. L. (2011). The application of a three-tier model of intervention to parent training. *Journal of Positive Behavior Interventions, 13*, 198–207. https://doi.org/10.1177/1098300711405337.

Plant, K. M., & Sanders, M. R. (2007). Predictors of care-giver stress in families of preschool-aged children with developmental disabilities. *Journal of Intellectual Disability Research, 51*, 109–124. https://doi.org/10.1111/j.1365-2788.2006.00829.x.

Reitman, D., & McMahon, R. J. (2013). Constance "Connie" Hanf (1917–2002): The mentor and the model. *Cognitive and Behavioral Practice, 20*, 106–116. https://doi.org/10.1016/j.cbpra.2012.02.005.

Rentinck, I. C., Ketelaar, M., Jongmans, M. J., & Gorter, J. W. (2007). Parents of children with cerebral palsy: A review of factors related to the process of adaptation. *Child: Care, Health and Development, 33*, 161–169. https://doi.org/10.1111/j.1365-2214.2006.00643.x.

Risdal, D., & Singer, G. H. S. (2004). Marital adjustment in parents of children with disabilities: A historical review and meta-analysis. *Research and Practice for Persons with Severe Disabilities, 29*, 95–103.

Ruane, A., & Carr, A. (2018). *Systematic review and meta-analysis of Stepping Stones Triple P for parents of children with disabilities. Family Process. Advance online publication*. https://doi.org/10.1111/famp.12352.

Sampaio, F., Feldman, I., Richards-Jones, S., & Mihalopoulos, C. (2018). The economic benefits of sustained investment in parenting. In M. R. Sanders & A. Morawska (Eds.), *Handbook of parenting and child development across the lifespan (chapter 35)*. New York, NY: Springer. https://doi.org/10.1007/978-3-319-94598-9_35 .

Sanders, M. R., & Mazzucchelli, T. G. (2013). The promotion of self-regulation through parenting interventions. *Clinical Child and Family Psychology Review, 16*, 1–17. https://doi.org/10.1007/s10567-013-0129-z.

Sanders, M. R., & Mazzucchelli, T. G. (2018a). Core principles and techniques of positive parenting. In M. R. Sanders & T. G. Mazzucchelli (Eds.), *The power of positive parenting: Transforming the lives of children, parents and communities using the Triple P system* (pp. 63–78). New York, NY: Oxford University Press.

Sanders, M. R., & Mazzucchelli, T. G. (2018b). How parenting influences the lives of children. In M. R. Sanders & T. G. Mazzucchelli (Eds.), *The power of positive parenting: Transforming the lives of children, parents and communities using the Triple P system* (pp. 5–31). New York, NY: Oxford University Press.

Sanders, M. R., & Mazzucchelli, T. G. (Eds.). (2018c). *The power of positive parenting: Transforming the lives of children, parents, and communities using the Triple P system*. New York, NY: Oxford University Press.

Sanders, M. R., Mazzucchelli, T. G., & Studman, L. J. (2004). Stepping Stones Triple P: The theoretical basis and development of an evidence-based positive parenting program for families with a child who has a disability. *Journal of Intellectual and Developmental Disability, 29*, 265–283. https://doi.org/10.1080/13668250412331285127.

Sanders, M. R., & Plant, K. (1989). Programming for generalization to high and low risk parenting situations in families with oppositional developmentally disabled preschoolers. *Behavior Modification, 13*, 283–300. https://doi.org/10.1177/01454455890133001.

Sanders, M. R., & Prinz, R. J. (2018). Emergence of a population approach to evidence-based parenting support. In M. R. Sanders & T. G. Mazzucchelli (Eds.), *The power of positive parenting: Transforming the lives of children, parents, and communities using the Triple P system* (pp. 32–62). New York, NY: Oxford University Press.

Sanders, M. R., Tully, L. A., Baade, P. D., Lynch, M. E., Heywood, A. H., Pollard, G. E., & Youlden, D. R. (1999). A survey of parenting practices in Queensland: Implications for mental health promotion. *Health Promotion Journal of Australia, 9*, 105–114.

Schalock, R. L., Borthwick-Duffy, S. A., Bradley, V. J., Buntinx, W. H. E., Coulter, D. L., Craig, E. M. P., et al. (2010). *Intellectual disability: Definition, classification, and systems of supports* (11th ed.). Washington, DC: AAIDD.

Shaffer, A., Kotchick, B. A., Dorsey, S., & Forehand, R. (2001). The past, present, and future of behavioral parent training: Interventions for child and adolescent problem behavior. *The Behavior Analyst Today, 2*, 91–105. https://doi.org/10.1037/h0099922.

Shearer, M. S., & Shearer, D. E. (1976). The Portage Project: A model for early childhood education. In D. D. Tjossem (Ed.), *Intervention strategies for high risk infants and young children*. Baltimore, MD: University Park Press.

Siegel, D. J., & Bryson, T. P. (2014, September). 'Time-outs' are hurting your child. *Time*. Retrieved from http://time.com/3404701/discipline-time-out-is-not-good/.

Simó-Pinatella, D., Font-Roura, J., Planella-Morató, J., McGill, P., Alomar-Kurz, E., & Giné, C. (2012). Types of motivating operations in interventions with problem behavior: A systematic review. *Behavior Modification, 37*, 3–38. https://doi.org/10.1177/0145445512448096 .

Singer, G. H. S. (2006). Meta-analysis of comparative studies of depression in mothers of children with and without developmental disabilities. *American Journal on Mental Retardation, 111*, 155–169. https://doi.org/10.1352/0895-8017(2006)111[155:MOCSOD]2.0.CO;2.

Skinner, B. F. (1938). *The behavior of organisms: An experimental analysis*. New York, NY: Appleton-Century-Crofts.

Skinner, B. F. (1953). *Science and human behavior*. New York, NY: Free Press.

Sloper, P., Knussen, C., Turner, S., & Cunningham, C. (1991). Factors related to stress and satisfaction with life in families of children with Down's syndrome. *Journal of Child Psychology and Psychiatry, 32*, 655–676. https://doi.org/10.1111/j.1469-7610.1991.tb00342.x.

Smith, G. C., Hayslip, B., Jr., Hancock, G. R., Strieder, F. H., & Montoro-Rodriguez, J. (2018). A randomized clinical trial of interventions for improving well-being in custodial grandfamilies. *Journal of Family Psychology, 32*, 816–827. https://doi.org/10.1037/fam0000457.

Sofronoff, K., Gray, K. M., Einfeld, S. L., & Tonge, B. J. (2018). Supporting families of children with a disability. In M. R. Sanders & T. G. Mazzucchelli (Eds.), *The power of positive parenting: Transforming the lives of children, parents, and communities using the Triple P system* (pp. 442–454). New York, NY: Oxford University Press.

Stalker, K., & McArthur, K. (2012). Child abuse, child protection and disabled children: A review of recent research. *Child Abuse Review, 21*, 24–40. https://doi.org/10.1002/car.1154.

Taanila, A., Ebeling, H., Heikura, U., & Jarvelin, M.-R. (2003). Behavioural problems of 8-year-old children with and without intellectual disability. *Journal of Pediatric Neurology, 1*, 15–24.

Tellegen, C. L., & Sanders, M. R. (2013). Stepping Stones Triple P-Positive Parenting Program for children with disability: A systematic review and meta-analysis. *Research in Developmental Disabilities, 34*, 1556–1571. https://doi.org/10.1016/j.ridd.2013.01.022.

Tellegen, C. L., & Sanders, M. R. (2014). A randomized controlled trial evaluating a brief parenting program with children with autism spectrum disorders. *Journal of Consulting and Clinical Psychology, 82*, 1193–1200. https://doi.org/10.1037/a0037246.

Tonge, B. J., & Einfeld, S. L. (2003). Psychopathology and intellectual disability: The Australian child to adult longitudinal study. In L. M. Glidden (Ed.), *International review of research in mental retardation* (Vol. 26, pp. 61–91). San Diego, CA: Academic Press.

Wilkinson, L. (2018). Using social marketing strategies to enhance program reach. In M. R. Sanders & T. G. Mazzucchelli (Eds.), *The power of postive parenting: Transforming the lives of children, parents, and communities using the Triple P system* (pp. 383–394). New York, NY: Oxford University Press.

Wolfensberger, W., Nirje, B., Olshansky, S., Perske, R., & Roos, P. (1972). *The principle of normalization in human services*. Toronto, Canada: National Institute on Mental Retardation.

Woodman, A. C., Mawdsley, H. P., & Hauser-Cram, P. (2015). Parenting stress and child behaviour problems within families of children with developmental disabilities: Transactional relations across 15 years. *Research in Developmental Disabilities, 36*, 264–276. https://doi.org/10.1016/j.ridd.2014.10.011.

41

Self-Help Skills

Johnny L. Matson and Esther Hong

Overview

A major area of research, historically, in the field of intellectual disabilities (ID; formerly "mental retardation" for those doing literature searches on the topic) has been self-help skills. These issues are particularly salient for children and persons with severe or profound cognitive deficits. However, this area of skill training also has implications for older persons with ID as well as for those individuals whose cognitive deficits are not as impairing.

Other factors are also at play regarding the focus on self-help skills. Obviously, these skills promote independence. Additionally, the move toward normalization, deinstitutionalization, and independent living has played major roles in the development of the area. These trends are exemplified by studies such as the one by O'Neill, Brown, Gordon, and Schonhorn (1985), which studied 27 adults with severe or profound IDs, who had been transitioned from institutional care to a post-institutional placement program in New York City. These new residences were apartments with two or three occupants and group homes with eight residents. The authors noted increased activity and increased communication skills. Additionally, the participants demonstrated improvements in self-care skills, such as eating, table manners, selecting clothing, dressing and undressing, showering, toileting, grooming, preparing meals, and house cleaning skills. The authors speculated that factors, such as more learning opportunities in community living arrangements, may have contributed to these improved outcomes; however, clear reasons for these gains were not directly tested.

Another theme is presented by Fox, Holtz, and Moist (2009). In their study, four adults with autism spectrum disorder (ASD) and severe or profound ID were treated for serious long-term behavior problems. They described their intervention as a community-based accommodation program. The stated focus was to design a highly predictable environment based on individual preference. The authors reported that one participant preferred to work in a rocking chair with cushions, at a table away from others. A second participant preferred to be seated so he could observe people walking down the hall near his work area. Reinforcer surveys were also used to further personalize training, and these data were used in combination with a contingency management program to enhance prosocial behaviors and to aid in the reduction of challenging behaviors.

These examples demonstrate important trends in promoting living in the community, being independent, having programs that are customized

---

J. L. Matson · E. Hong (✉)
Department of Psychology, Louisiana State University, Baton Rouge, LA, USA
e-mail: ehong1@lsu.edu

J. L. Matson (ed.), *Handbook of Intellectual Disabilities*, Autism and Child Psychopathology Series, https://doi.org/10.1007/978-3-030-20843-1_41

to the individual, and having the right to make choices. Millar (2008) underscored this point in her paper regarding issues that emerge when an individual reaches the age of majority and are impacted by Individuals with Disabilities Education Improvement Act (IDEIA; Rehabilitation Act Amendments of 1992 and 1998, US Congress). Based on each individual's needs and strengths, supports need to be developed to assist the person in reaching their maximum potential. These goals have in large part been achieved via the development of treatments that largely fit into a teaching, structured program, which are complemented by specific applied behavior analysis (ABA) treatment methods.

Assessment

A number of tests, such as the Vineland Social Maturity Scale, have been developed and are widely used in clinical practice. These tests are covered extensively elsewhere in this volume. As a result, the focus here is on a few methods that are used less often but are also valuable. Additionally, these measures are very specific to assisting in the development of treatments for self-help skills. Matson, Dempsey, and Fodstad (2009), for example, studied the Adaptive Behavior Task Analysis Checklist (ABTAC). As with the Murdock Center task analyses, this instrument provides information that can assist in targeting and breaking down skills into discrete tasks. These tasks can be prompted, modeled, and/or reinforced to develop self-help and independence skills. There were 234 adults with ID only and adults with both ID and ASD who participated. Items involved dressing and undressing, grooming, housekeeping, and meal preparation. The greatest skill deficits were evident for persons with ID and ASD. Further, as ASD symptoms became more severe, so did deficits in independent living skills.

Another assessment measure, this one specific to feeding problems, has also been described (Screening Tool of Feeding Problems [STEP]; Matson & Kuhn, 2001). They evaluated 57 people between 10 and 87 years of age, with ID levels of mild to profound. The STEP has 23 items, all of which address essential feeding problems such as the ability to chew, food stealing, eating small amounts of food, and selecting food by texture. The general conclusion was that the measure had good psychometric properties.

These are two studies that present measures to help identify behaviors for intervention and aid in measuring treatment progress. As noted, popular measures of adaptive behavior are covered elsewhere in this book. The reader may also wish to consult Kraijer (2000), who provides a nice review of adaptive behavior measures for persons with ID and ASD.

Overview of Treatment Methods

A variety of strategies have been used to teach independent living skills. Schoen and Sivil (1989) described the teaching of self-help skills to children with disabilities. A variety of methods were employed including chaining, prompting (both verbal and physical, which were gradually faded out), observational learning or modeling, and reinforcement. This group of methods has proven to be typical for many programs with the ID population and has routinely been effective.

Ferretti, Cavalier, Murphy, and Murphy (1993) provided another excellent example of frequently used treatment methods. They rightly pointed out that the transition to less restrictive living settings for persons with ID is important. These authors reviewed self-management strategies and broke them into three categories. Categories were characterized as self-instruction and verbal-mediated chains of behaviors (e.g., goal setting, strategy selection, self-monitoring, self-evaluation, and providing one's own consequences for behaviors). The third category involved self-management skills. Under the self-instruction skills category, trainees learned how to define the problem and learned the correct sequence in which to perform skills that aid in a resolution of the problem. Other skills focused on evaluating one's performance as well as dealing with mistakes and the frustration that can result following the mistakes. Finally, methods of providing consequences, whether correct or incorrect, were taught.

The second category was self-management of a chain of behaviors. Among the skills taught included self-monitoring, self-management, and learning to record target behaviors. Self-administering reinforcement and punishment were also employed in an effort to help the trainee focus on the components of a task that needed to be performed and whether these were being carried out correctly. When necessary, therapists provided prompts and feedback to aid in the learning process. The final category that Ferretti et al. (1993) described was multicomponent self-management studies. They noted that multiple procedures were used at once with these treatments. Among the methods employed were self-evaluation, self-monitoring, self-reinforcement, self-recording, tokens, performance goals, and visual cues.

Akhmetzyanova (2014) pointed out that structured, step-by-step instruction is important for teaching self-help skills to persons with ID. She also pointed out that individuals with severe and profound ID require special one-to-one training in their efforts to achieve independence. We would add that shorter but more frequent training sessions should be employed, with smaller behavioral steps or goals, more physical prompts, and the use of more tangible reinforcements.

Millar (2008), in discussing goals in treatment, underscored the need for self-determination. Certainly, this is a critical goal, and the development of self-help skills provides a vital tool in achieving this goal. These goals and skills within the self-help literature are therefore often referred to as independent living skills. Matson, Smalls, Hampff, Smiroldo, and Anderson (1998) described the use of staff training, feedback, edible reinforcers, modeling, and physical guidance to train these skills among 22 adults with severe and profound ID. Even with these most difficult-to-train adults, marked improvements were noted.

Browder, Hines, McCarthy, and Fees (1984) provided another good example of daily living skills training, which they suggest helped adults with moderate ID to integrate into the community. They used small group training with frequent prompting, using high-rate behaviors to reinforce low-rate behaviors, and time delay to teach sight words in a booklet (i.e., on food preparation, laundry, and telephone skills) to great effect. Falkenstine, Collins, Schuster, and Kleinert (2009) made a very good point with respect to instruction, particularly as applied to special education teachers. They stressed that teachers are constantly looking for strategies that can be employed in small group formats. Obviously, given the teacher-student ratio for children with ID makes these methods essential. They stressed the use of instructions, feedback, and observation of discrete targeted tasks. Falkenstine et al. (2009) stressed that small group instruction, when properly implemented, can be very effective. It was also stressed that the Individualized Education Plan (IEP) is extremely important and should serve as a major source for establishing treatment goals.

Another effective system for teaching and prompting adaptive behaviors has been described for adults with ID living in a group home (Anderson, Sherman, Sheldon, & McAdam, 1997). This study included two women (i.e., a 22-year-old with moderate ID, a 37-year-old with severe ID) and one man (i.e., 21-year-old with severe ID). The big focus was on activity schedules that employed photographs and line drawings. Tasks were performed independently when possible and with staff assistance when necessary. The types of tasks that were prompted included toothbrushing, handwashing, eating, housekeeping, vacuuming, preparing meals, watching TV, playing cards, and reading magazines. No formalized training of staff or residents occurred, yet positive effects were achieved.

Another study using activity schedules is described by Koyama and Wang (2011). They conducted a review and found 23 studies that used this approach for persons with ID. It was concluded that activity schedules could be very effective for teaching self-management and overall independence. Finally, Banda and Grimmett (2008) also promoted the use of activity schedules, in their case for persons with ASD. Their study was also a review which included 13 studies.

Again, as with the Koyama and Wang (2011) review, these authors reported good results. Of particular note was the increase in appropriate social interactions, on-task behavior, and transitions to new activities or settings. In the latter case, a marked decrease in tantrums and other challenging behaviors was noted.

Mancil (2009) stressed the value of milieu therapy. The author defined these treatments as some combination of modeling, manding, time delay, and/or incidental teaching. Time delay, manding, and modeling were the most commonly used methods. To enhance overall quality of life, there was also a focus on involvement of typically developing peers in social situations.

In sum, a range of different treatment procedures have been effectively employed with persons who have ID, which is estimated to occur in as many as 70% of persons with ASD. A combination of structure, schedules, and components of ABA appears to be the primary procedures used. Some of the skill areas that are most prominently featured in the self-help and enhance independence are further discussed.

Mealtime Behavior

Mealtime issues are a major concern for persons with ID. Among the common issues this group experiences are stunted growth, oral-motor deficits, and marked impairments in feeding one's self (Thommessen, Heiberg, Kase, Larsen, & Riis, 1991). A central theme in the self-help literature has been teaching persons with ID to eat independently or at least semi-independently. Typical of these efforts was a program by Song and Gandhi (1974), which taught four children with profound ID to eat with a spoon. They broke the tasks down into steps and used shaping, reinforcement, and, later, the fading of prompts to achieve this goal.

O'Brien, Bugle, and Azrin (1972) also taught a 6-year-old child with profound ID to eat with a spoon rather than with her hands. Manual guidance from the teacher was used for a six-step sequence of skills. These skills were trained in the following order: (1) hold the spoon in the right hand; (2) dip the spoon into the bowl and lift one inch above the bowl; (3) move the spoon to within 2 inches of her mouth; (4) the teacher places her hand on the child's chin and gently opens the child's mouth; (5) the spoon is guided into her mouth; and (6) the spoon is tilted upward, depositing the food into the girl's mouth, and then the spoon is removed. Independence was achieved, but when the treatment stopped, the child began using her hands to eat again; thus, treatment was reinstated and proven to be effective again.

Stimbert, Minor, and McCoy (1977) have also described a feeding program, which was implemented with six "low functioning" children with ID. The participants were served six meals a day, with a maximum time for eating of 15 minutes. Marked improvement in the use of a spoon to eat was noted for all the children treated. Physical guidance, using the least amount of contact possible, was paired with overcorrection when children made a mistake.

Another study using ABA to improve eating skills for persons with severe and profound ID was reported by Munk and Repp (1994). These authors noted that above 80% of persons with this level of cognitive impairment experience problems in eating. In their paper, five participants between 8 and 21 years of age sampled a variety of foods with different textures. Based on their findings, eating problems were grouped into one of four categories: (1) total refusal, (2) type of food, (3) texture of food, and (4) type and texture of food.

Teaching self-feeding skills to 14 boys aged 9–17 years who evinced profound ID was reported by Berkowitz, Sherry, and Davis (1971). Training was conducted on a one-on-one basis and was provided for all 21 meals served each week. A seven-step treatment model was followed. In step one, the trainer held the child's hand while the child grasped a spoon and scooped food from the plate, and the trainer guided the spoon to the child's mouth and then back to the plate. In step two, the child independently guided the spoon for the final 2–3 inches. In step three, the child independently guided the spoon for 6 inches. Step four involved independent eating from the plate. Step five involved independent spoon use, beginning with scooping food from the plate. Step six required the child to bring the spoon to the plate, scoop food, and lift the spoon and food from the plate, after the trainer had released the child's hand. In the seventh and final step, the child completed the entire sequence independently. Depending on the skill level of each child, training varied from 2 to 60 days before independent feeding occurred. The authors concluded that the study underscored that independent eating can be trained to persons with profound ID.

Children with multiple disabilities, including ID and emotional disorders, have also been treated. Sisson and Dixon (1986) worked with six boys who ranged in age from 4 to 9 years old, who had mild to profound ID. Comorbid disorders included attention-deficit hyperactivity disorder (ADHD), ASD, psychosis, or conduct disorder. Intervention targets included chewing with one's mouth closed, using a utensil appropriately, and using a napkin. Intervention procedures consisted of verbal praise and time-out for incorrectly carrying out target behaviors (e.g., pushing plate to the center of the table and refraining from verbal communication for 10 seconds). After the time-out, verbal instructions were provided, and praise for correct execution of target behaviors was given. The authors reported marked improvement of target behavior but modest generalization and maintenance.

Rehfeldt, Dahman, Young, Cherry, and Davis (2003) approached a related but different set of skills, meal preparation. They treated adults with severe or moderate ID. Training included observing a video of a person with ID making a peanut butter and jelly sandwich, during which a trainer praised the participant for each successful step in the sandwich-making process. This approach was sufficient in allowing the three trainees to develop this food preparation skill. Developing skills such as the ones mentioned above is critically important in developing a more independent lifestyle.

The importance of certain aspects of dining is further underscored by Angell, Bailey, and Larson (2008). Their study is of interest in that it focuses on social behaviors of five high school students (15–19 years old) with moderate ID. Behaviors taught included taking turns and asking partner-focused questions. Eye contact and tone of voice were also taught using role-playing and social reinforcement. Graduate students were paired with participants for lunch, twice a week. During lunch, the graduate student initiated scripted communication scenarios. This training model resulted in improved social interactions for the high school students during lunchtime.

A final example study on mealtime behavior reviewed here was conducted by Cannella-Malone et al. (2006). Their treatment included showing videos to five men and one woman, all of whom fell in the mild to moderate domains of ID. The tasks focused on putting away groceries and setting the table. Putting away groceries involved unloading a paper bag that contained 10 items: two apples in a plastic bag, a bag of frozen peas, two oranges in a plastic bag, a box of cereal, a bottle of salad dressing, a can of juice, a box of snacks, and a can of beans. The Murdock Center has an extensive number of skills that have been task analyzed and broken into steps. To promote uniformity, these tasks were adopted from the Murdock materials. Ten steps for unpacking groceries were used, including taking items from the paper bag to placing items in the refrigerator, putting cans in the cupboard, and putting boxes on top of the refrigerator. Setting the table also had 10 steps and included items such as putting down the placement, putting the forks to the left of the large plate, and sitting down at the place and eating a small meal. Prompts, modeling, social reinforcement, and practice of the steps proved to be an effective intervention package.

Video modeling has been a popular choice with respect to treatment options for teaching daily living skills. This method of training is typically paired with the use of task-analyzed steps, prompts, and social reinforcement (Shipley-Benamou, Lutzker, & Taubman, 2002). Note that one paper reviewed previously in this chapter involved teaching conversational skills in the lunchroom. On a somewhat different take on communication skills, Mechling, Pridgen, and

Cronin (2005) used computer-based video training. The focus of their intervention was on answering questions while making a purchase in a fast-food restaurant. They treated two males and one female between the ages of 17 and 20 years, all of whom had moderate ID. All training initially occurred in a secluded room in their high school library. Generalization of skills taught occurred at Wendy's, Hardee's, and McDonald's. These chains were selected due to the frequency of use by the participants. In addition to watching videos, a trainer sat next to the client and provided performance feedback and reinforcement. Items that the participants learned to name were types of sandwiches, drinks, and size of the French fry order. This successful training also included paying the bill.

As noted in the study just discussed, purchasing was one of the tasks trained to the three adults with moderate ID. Depending on the type of purchase made, the complexity of the tasks varied. Haring, Kennedy, Adams, and Pitts-Conway (1987) described one such study. As can be seen from the date of publication, this is a topic that has attracted the attention of researchers for some time. Participants included two males and one female who had ASD and severe ID. The authors stated that according to the Vineland Adaptive Behavior Scales (VABS; Sparrow, Balla, & Cicchetti, 1984), two participants were functioning at the 5-year-old level, while the third participant was assessed at the four-year-old level. Two participants were trained to purchase food in their high school cafeteria, while the third was trained in a convenience store, close to the school. Treatment included verbal praise, the use of tasks analyzed skills, and prompts (both physical and verbal). Typical steps trained in the task analysis were to enter store, get in line, take out money, receive change, and exit store. This approach to training was proven to be effective.

Self-Dressing

Teaching dressing skills to individuals with ID has a long history. In fact, with the advent of behavior modification (now referred to as ABA), self-care skills, particularly independent dressing, has been right at the forefront. One of the early examples of this approach was reported by Azrin, Schaeffer, and Wesolowski (1976). In this groundbreaking study, seven adults with severe ID participated; none of the participants could speak or independently dress or undress themselves. Preferred reinforcers, such as going for a walk, napping, or being with a favorite employee, was provided at the end of the dressing/undressing sequence. During the training itself, reinforcers of shorter duration (e.g., snack items, praise) were used. During training, the participants were seated. Five types of garments, including underpants, shoes, socks, pants, and shirt, were employed. Items were slip-on, so no laces, buttons, belts, or snaps were used. Items were put on and taken off in a natural sequence during a three-hour training session. Prompts, both verbal and physical, were used. All seven participants were able to independently dress and undress after 20 hours of training.

Sisson, Kilwein, and Van Hasselt (1988) also tackled this problem, but in their case, the participants were children. The authors treated two boys, aged 5 and 9. Both children were blind and experienced moderate to severe ID. Assessment and treatment occurred in their bedroom. Tube socks (no heel), pullover shirts, and pants with an elastic waistband were used. A task analysis was used in the training of the three items: 8 steps for putting on socks, 14 steps for putting on pants, and 17 steps for putting on a shirt. Verbal prompts and graduated guidance over a minimum of 10 training trials were used. Training continued over the course of 25 and 95 sessions, until all three skills could be completed at 100%. Independent dressing skills have also been trained by Hughes, Schuster, and Nelson (1993). They taught two students to dress using a shirt, pants, socks, and a jacket.

A treatment package using video modeling and prompts was described by Norman, Collins, and Schuster (2001). The participants included three elementary school students, and the skills taught were cleaning sunglasses, putting on a wrist watch, and zipping a jacket. Trainers evaluated the number of steps in the dressing tasks that were completed correctly before and after observing the video training. The authors reported good results using the methods described.

As noted in this study and the independent dressing studies noted above, breaking tasks down into smaller components is a common practice in self-help interventions. However, other effective methods to teach dressing are available. Young, West, Howard, and Whitney (1986) trained the entire tasks at once instead of breaking it down into components. This model also involved having the participant, two disabled preschool children, receive graduated guidance. This latter approach involved using the minimal amount of physical guidance needed to complete the task. This approach was also accompanied by verbal instructions and praise.

A final dressing study reviewed was published by Day and Horner (1986). Six males with profound ID, between 8 and 23 years of age, were taught how to put on a shirt. Five of the six participants were nonverbal, while the sixth participant had a limited number of simple word responses. Training included prompts (i.e., physical, verbal, and gestural) and praise for correct performance. Errors in the process resulted in interrupting the trial, repositioning the shirt to correct the error, and then providing additional assistance. All participants successfully learned how to put on their shirt, and this skill was generalized to other shirts.

Safety Skills

With the international move to integrated community living for persons with ID, new, more complex safety skills are needed and must be taught. Pedestrian skills would be one example of this trend. Batu, Ergenekon, Erbas, and Akmanoglu (2004) provided one training example in an area in Turkey. They tested five individuals (7–15 years old) with severe ID. Prior to treatment, none of these participants had the skills needed to independently cross the street. Three behaviors were taught: crossing streets using an overcrossing (5 steps), crossing the street using pedestrian lights (4 steps), and crossing the street where there was no traffic patrol or facilitations (9 steps). A simulated model of the crossing was devised in the school gym, which is where the initial training occurred. This simulated approach enhanced safety and eliminated weather issues. Instruction continued until mastery criteria were achieved on all steps of each task using a most-to-least prompt approach. These prompts were both verbal and physical. Verbal praise was reserved until the entire sequence of steps had been performed correctly. Successful generalization of these skills to real traffic situations was achieved.

A range of skills have been studied and reported on in the literature for some time. Martin, Rusch, and Heal (1982) provided a wide-ranging review of topics that have been addressed. They discussed studies on travel, money management, meal preparation, clothing and personal care, telephone use, housekeeping, leisure, social and conversational skills, and self-medication. The use of ABA is almost universal in these studies. Also, the objective is largely to not only promote independence but to enhance community integration.

Personal safety is another important life skill that has been addressed in the literature (Kim, 2010). In this study, the focus was on teaching self-protection skills to persons with ID. In this review, one of the primary focuses was on defending against sexual abuse. In that context, children were taught about body ownership. Additionally, a focus on decision-making, independent thinking, and the recognition about when to obey others versus recognizing inappropriate requests was trained. Social skills training in the context of sexual behaviors has also been addressed, along with the development of educational plans focused on reproduction, parenting, contraception, and sexually transmitted diseases.

Another nice review of the self-help (i.e., safety skills) literature was published by Mechling (2008). She covered 30 years and 36 investigations on the topic. In this review, six topics were targeted: pedestrian/street crossing safety, home accident prevention, applications of first aid and reporting illness, repelling advances of strangers, fire safety, and emergency phone use. She found that behavioral strategies that incorporated the simulation of the skill were common. Additionally, generalization and maintenance strategies were also frequently incorporated in these studies. The author underscored the ability of people with ID to learn a series of complex independent living skills.

Video skills may be the most common form of simulation when teaching self-help skills (Mechling & O'Brien, 2010). This study taught two females and one male high school student (between 19 and 20 years of age) to use public buses. The training video was from the trainee's perspective and showed the participant walking to the bus stop, boarding the bus, sitting down, riding the bus, looking for landmarks from the window, pushing the stop bus signal, and exiting the bus. PowerPoint slides of bus use skills were also included in the successful training program.

Mechling, Gast, and Gustafson (2009) have used a similar treatment model to teach how to extinguish cooking-related fires. They worked with three persons with moderate ID, two females and one male between 9 and 21 years of age. The authors trained three skills, which included scooping and releasing flour, placing a lid on a pot or pan, and using a fire extinguisher. This program, as with the other programs described in this chapter, proved to be effective. Mechling and Seid (2011) also illustrated the use of a handheld personal digital assistant. Using this self-prompting device, the participants were taught pedestrian travel skills across multiple destinations.

While the focus of most self-help studies has been directed toward the trainee, some focus on training parents has also occurred. Feldman and Case (2009) stressed that children can be at risk for impairments in self-help skills when parents display poor career skills. These authors tested self-instructional manuals and audiovisual training. Using low-tech, inexpensive teaching aids, 9 of the 10 parents (who did not have ID) showed marked improvements that were maintained for over half a year.

Parental opinions about safety skills training have been less frequently studied, but are important nonetheless (Agran & Krupp, 2010). In their survey, it was found that most parents (93%) felt that safety training was of critical importance. They also suggested that these skills should be taught at school and at home. Keeping in mind that the children being referenced were in elementary school, parents considered that the following training goals were of most to least: home safety, crime prevention, drug prevention, work safety, and HIV/AIDS prevention.

Horner, Williams, and Steveley (1987) described the use of a telephone, another important skill, with moderate to severely ID high schoolers. With the advent of smartphones, the need to develop some skills in this area becomes an even more important self-help/personal safety skill. In the aforementioned study, four people aged 17–21 years participated. A major focus of the paper was to teach the trainees to use different phones in a variety of settings. This goal is now largely obsolete due to the mobility of most phones now in use.

Hygiene

A final topic included in this overview of treatments for self-help skills is hygiene skills. One topic that has received considerable attention involves menstrual care. Epps, Stern, and Horner (1990) described one of the earliest studies on the topic. The authors noted the high importance of this topic in that there is a high rate of clinic referrals for hysterectomies due to menstrual hygiene issues. In this study, four females participated, three who were 15 years old and one who was 37 years old. For all trainees, menstrual care was not independent, and no previous training on these skills had occurred. The menstrual cycle was described to be "regular," and the mothers or supervisors of participants endorsed the importance of developing these skills. The target behavior was straightforward and involved changing soiled undergarments at designated check times. Verbal prompts, corrections, and praise were also included in this successful program.

Ersoy, Tekin-Iftar, and Kircaali-Iftar (2009) noted that menstrual care is among the most important hygiene skills for adolescents and adult women with ID. They treated three females between 12 and 14 years of age, with mild or moderate ID. In addition to changing soiled clothing, they also targeted other sanitary skills, such as handwashing and drying. Verbal directions, reinforcement, and prompts were successful in the training of an 18-step task analysis.

A bigger study on the same topic was reported by Hamilton, Allen, Stephens, and Daval (1969). The 35 individuals treated had an average IQ of 26 (ranging from 10 to 45) and an average age of 21.5 years. Again, the successful training model involved components of ABA. The topic of personal care has also been addressed by Carnaby and Cambridge (2002). They noted that intimate care issues need to be addressed with respect to specific skill training. Additionally, guidelines for practice and management are needed. The nature of these skills underscores the importance of privacy and choice.

Grooming skills have also been successfully trained. In one study, Salovita and Tuulkari (2000) taught grooming skills to a man with moderate ID. Treatment included picture cues using a task analysis. A least-intrusive prompt and social reinforcement paradigm that focused on toothbrushing, shaving, showering, and washing the body and face were found to be effective. Another grooming skill, learning to shower independently, was targeted with 72 adults (mean age of 32 years). These participants ranged in level of functioning, from moderate to severe ID. The target skill was broken into 27 steps (Matson, DiLorenzo, & Esveldt-Dawson, 1981). The task analysis included acquiring a washcloth, turning on the water, adjusting the temperature, getting wet, washing one's hair, lathering a washcloth, 12 steps on the washing of various body parts, rinsing off the soap, wringing out the washcloth, disposing the washcloth correctly, 5 steps on drying the body, and finally applying deodorant. The trainer, while fully clothed, modeled the task analysis. While the participant was showering, the trainer provided verbal instructions and verbal prompts. This treatment protocol was found to be highly successful.

Summary and Conclusions

Self-help skills have been a training target among persons with the full range of ID (i.e., mild to profound) for decades. Participants of the intervention have included individuals across the life span. Only successful outcomes were reported, which tends to be the nature of journal articles. Likely, this is a bit of a rosy picture, with many people responding to some extent or not at all. However, a number of factors are probably good predictors of success, including level of motivation, the degree of cognitive deficits, and the complexity of the task being trained.

Common elements appear to be evident in most of the wide range of skills that have been taught. These training components routinely involve modeling, verbal and physical prompts, performance feedback, as well as social and other forms of reinforcement. The peak period for training studies were during the 1980s–1990s. The general overall efficacy of this research may account for slowing of the publication pace in recent years. For the most part, the procedures described are now considered tried and true.

References

Agran, M., & Krupp, M. (2010). A preliminary investigation of parents' opinions about safety skills instruction: An apparent discrepancy between importance and expectation. *Education and Training in Autism and Developmental Disabilities, 45*, 303–311.

Akhmetzyanova, A. I. (2014). The development of self-care skills of children with severe mental retardation in the context of Lekoteka. *World Applied Sciences Journal, 29*, 724–727.

Anderson, M. D., Sherman, J. A., Sheldon, J. B., & McAdam, D. (1997). *Picture activity schedules and engagement of adults with mental retardation in a group home*. *Research in Developmental Disabilities* (Vol. 18, pp. 231–250).

Angell, M. E., Bailey, R. L., & Larson, L. (2008). Systemic instruction for social-pragmatic language skills in lunchroom settings. *Education and Training in Developmental Disabilities, 43*, 342–359.

Azrin, N. H., Schaeffer, R. M., & Wesolowski, M. D. (1976). A rapid method of teaching profoundly retarded persons to dress by a reinforcement-guidance method. *Mental Retardation, 14*, 29–33.

Banda, D. R., & Grimmett, E. (2008). Enhancing social and transition behaviors of persons with autism through activity schedules: A review. *Education and Training in Developmental Disabilities, 43*, 324–333.

Batu, S., Ergenekon, Y., Erbas, D., & Akmanoglu, N. (2004). Teaching pedestrian skills to individuals with developmental disabilities. *Journal of Behavioral Education, 13*, 147–164.

Berkowitz, S., Sherry, P. J., & Davis, B. A. (1971). Teaching self-feeding skills to profound retardates using reinforcement and fading procedures. *Behavior Therapy, 2*, 62–67.

Browder, D. M., Hines, C., McCarthy, L. J., & Fees, J. (1984). A treatment package for increasing sight word recognition for use in daily living skills. *Education and Training of the Mentally Retarded, 19*, 191–200.

Cannella-Malone, H. Sigafoos, J., O'Reilly, M., de la Cruz, B., Edrisinha, C., & Lancioni, G. E. (2006). Comparing video prompting to video modeling for teaching daily living skills to six adults with developmental disabilities. Education and Training in Developmental Disabilities*,* 41*,* 344–356.

Carnaby, S., & Cambridge, P. (2002). Getting personal: An exploratory study of intimate and personal care provision for people with profound and multiple intellectual disabilities. *Journal of Intellectual Disability Research, 46*, 120–132.

Day, H. M., & Horner, R. H. (1986). Response variation and the generalization of a dressing skill: Comparison of single instance and general case instruction. *Applied Research in Mental Retardation, 7*, 189–202.

Epps, S., Stern, R. J., & Horner, R. H. (1990). *Comparison of simulation training on self and using a doll for teaching generalized menstrual care to women with severe mental retardation*. *Research in Developmental Disabilities* (Vol. 11, pp. 37–66).

Ersoy, G., Tekin-Iftar, E., & Kircaali-Iftar, G. (2009). Effects of antecedent prompt and test procedure on teaching simulated menstrual care skills to females with developmental disabilities. *Education and Training in Developmental Disabilities, 44*, 54–66.

Falkenstine, K. J., Collins, B. C., Schuster, J. W., & Kleinert, H. (2009). Presenting chained and discrete tasks as non-targeted information when teaching discrete academic skills through small group instruction. *Education and Training in Developmental Disabilities, 44*, 127–142.

Feldman, M. A., & Case, L. (2009). Teaching child-care and safety skills to parents with intellectual disabilities through self-learning. *Journal of Intellectual and Developmental Disabilities, 24*, 27–44.

Ferretti, R. P., Cavalier, A. R., Murphy, M. J., & Murphy, R. (1993). The self-management of skills by persons with mental retardation. *Research in Development al Disabilities, 14*, 189–205.

Fox, R. A., Holtz, C. A., & Moist, A. M. (2009). A community based accommodation program for adults with autism and mental retardation. *Education and Training in Developmental Disabilities, 44*, 118–126.

Hamilton, J., Allen, P., Stephens, L., & Daval, E. (1969). Training mentally retarded females to use sanitary napkins. *Mental Retardation, 7*, 41–43.

Haring, J. G., Kennedy, G. H., Adams, M. J., & Pitts-Conway, V. (1987). Teaching generalization of purchasing skills across community settings to autistic youth using videotape modeling. *Journal of Applied Behavior Analysis, 20*, 89–96.

Horner, R. H., Williams, J. A., & Steveley, J. D. (1987). Acquisition of generalized telephone use by students with moderate and severe mental retardation. *Research in Developmental Disabilities, 8*, 229–247.

Hughes, M. W., Schuster, J. W., & Nelson, C. M. (1993). The acquisition of independent dressing skills by students with multiple disabilities. *Journal of Developmental and Physical Disabilities, 5*, 233–252.

Kim, Y. R. (2010). Personal safety programs for children with intellectual disabilities. *Education and Training in Autism and Developmental Disabilities, 45*, 312–319.

Koyama, T., & Wang, H. T. (2011). Use of activity schedule to promote independent performance of individuals with autism and other intellectual disabilities: A review. *Research in Developmental Disabilities, 32*, 2235–2242.

Kraijer, D. (2000). Review of adaptive behavior studies in mentally retarded persons with autism/pervasive developmental disorder. *Journal of Autism and Developmental Disorders, 30*, 39–47.

Mancil, G. R. (2009). Milieu therapy as a communication intervention: A review of the literature related to children with autism spectrum disorder. *Education and Training in Autism and Developmental Disabilities, 44*, 105–117.

Martin, J. E., Rusch, F. R., & Heal, L. W. (1982). Teaching community survival skills to mentally retarded adults: A review and analysis. *The Journal of Special Education, 16*, 243–267.

Matson, J. L., Dempsey, T., & Fodstad, J. C. (2009). The effect of autism spectrum disorders on adaptive independent living skills in adults with severe intellectual disability. *Research in Developmental Disabilities, 30*, 1203–1211.

Matson, J. L., DiLorenzo, T. M., & Esveldt-Dawson, K. (1981). Independence training as a method of enhancing self-help skills acquisition of the mentally retarded. *Behaviour Research and Therapy, 19*, 399–405.

Matson, J. L., & Kuhn, D. E. (2001). Identifying feeding problems in mentally retarded persons: Development and reliability of the Screening Tool of Feeding Problems (STEP). *Research in Developmental Disabilities, 21*, 165–172.

Matson, J. L., Smalls, Y., Hampff, A., Smiroldo, B. B., & Anderson, S. J. (1998). A comparison of behavioral techniques to teach functional independent living skills with severe and profound mental retardation. *Behavior Modification, 22*, 298–306.

Mechling, L. C. (2008). Thirty year review of safety skill instruction for persons with intellectual disabilities. *Education and Training in Developmental Disabilities, 43*, 311–323.

Mechling, L. C., Gast, D. L., & Gustafson, M. R. (2009). Use of video modeling to teach extinguishing of cooking related fires to individuals with moderate intellectual disabilities. *Education and Training in Autism and Developmental Disabilities, 44*, 67–79.

Mechling, L. C., & O'Brien, E. (2010). Computer-based video instruction to teach students with intellectual disabilities to use public bus transportation. *Education and Training in Autism and Developmental Disabilities, 45*, 230–241.

Mechling, L. C., Pridgen, L. S., & Cronin, B. A. (2005). Computer-based video instruction to teach students with intellectual disabilities to verbally respond to questions and make purchases in fast food restaurants. *Education and Training in Autism and Developmental Disabilities, 40*, 47–59.

Mechling, L. C., & Seid, N. H. (2011). Use of a hand-held personal digital assistant (PDA) to self-prompt pedestrian travel by young adults with moderate intellectual disabilities. *Education and Training in Autism and Developmental Disabilities, 46*, 220–237.

Millar, D. S. (2008). Self-determination in relation to having or not having a legal guardian: Case studies of two school-aged young adults with developmental disabilities. *Education and Training in Autism and Developmental Disabilities, 43*, 279–293.

Munk, D. D., & Repp, A. C. (1994). Behavioral assessment of feeding problems of individuals with severe disabilities. *Journal of Applied Behavior Analysis, 27*(2), 241–250.

Norman, J. M., Collins, B. C., & Schuster, J. W. (2001). Using an instructional package including video technology to teach self-help skills to elementary students with mental disabilities. *Journal of Special Education Technology, 16*, 5–18.

O'Brien, F., Bugle, C., & Azrin, N. H. (1972). Training and maintaining a retarded child's proper eating. *Journal of Applied Behavior Analysis, 5*, 67–72.

O'Neill, J., Brown, M., Gordon, W., & Schonhorn, R. (1985). The impact of deinstitutionalization on activities and skills of severely/profoundly mentally retarded multiply-handicapped adults. *Applied Research in Mental Retardation, 6*, 361–371.

Rehfeldt, R. A., Dahman, D., Young, A., Cherry, H., & Davis, P. (2003). Teaching a simple meal preparation skill to adults with moderate and severe mental retardation using video modeling. *Behavioral Interventions, 18*, 209–218.

Salovita, T. J., & Tuulkari, M. (2000). Cognitive-behavioural treatment package for teaching grooming skills to a man with an intellectual disability. *Scandinavian Journal of Behaviour Therapy, 29*, 140–147.

Schoen, S. F., & Sivil, E. O. (1989). A comparison of procedures in teaching self-help skills: Increasing assistance, time delay, and observational learning. *Journal of Autism and Developmental Disorders, 19*, 57–72.

Shipley-Benamou, R., Lutzker, J. R., & Taubman, M. (2002). Teaching daily living skills to children with autism through instructional video modeling. *Journal of Positive Behavior Interventions, 4*, 165–175.

Sisson, L. A., & Dixon, M. J. (1986). A behavioral approach to the training and assessment of feeding skills in multihandicapped children. *Applied Research in Mental Retardation, 7*, 149–163.

Sisson, L. A., Kilwein, M. L., & Van Hasselt, V. B. (1988). A graduated guidance procedure for teaching self-dressing skills to multihandicapped children. *Research in Developmental Disabilities, 9*, 419–432.

Song, A. Y., & Gandhi, R. (1974). An analysis of behavior during the acquisition and maintenance phases of self-spoon feeding skills of profound retardates. *Mental Retardation, 12*, 25–28.

Sparrow, S. S., Balla, D. A., & Cicchetti, D. V. (1984). *Vineland adaptive behavior scales*. Circle Pines, MN: American Guidance Service.

Stimbert, V. E., Minor, J. W., & McCoy, J. F. (1977). Intensive feeding training with retarded children. *Behavior Modification, 1*, 517–530.

Thommessen, M., Heiberg, A., Kase, B. F., Larsen, S., & Riis, G. (1991). Feeding problems, height, and weight in different groups of disabled children. *Acta Paediatrica, 80*, 522–533.

Young, K. R., West, R. P., Howard, V. F., & Whitney, R. (1986). Acquisition, fluency training, generalization, and maintenance of dressing skills of two developmentally disabled children. *Education and Treatment of Children, 9*, 16–29.

42

Curriculums

Hsu-Min Chiang and Kalli Kemp

Characteristics of Individuals with Intellectual Disability

Individuals with intellectual disability (ID) have various needs in independent living [American Psychiatric Association (APA, 2013)]. They are characterized by impairments in intellectual functioning and adaptive behaviors (APA, 2013). These impairments have impacts on one's functioning, performance, and participation in school, community, and work.

Information from a study using the National Longitudinal Transition Study-2 (NLTS-2) data provides a glimpse into the nationwide school experiences and characteristics of the US secondary (7th–12th grade) students with ID in communication, daily living and functioning, social skills, and academic experiences. Lipscomb et al. (2017) reported that a high percentage of students with ID (72%) live in low-income households and a high percentage of students with ID (75%) cannot perform activities of daily living well. As compared to students with other disabilities, students with ID are more likely to have poor health and chronic conditions and difficulties in communication and understanding. They are also less prepared for independent living and postsecondary employment. Their parents are less likely to expect them to live independently at age 30.

Individuals with ID have delays in social and communication skills (de Bildt et al., 2005). They may be able to initiate social interaction with peers but have problems in sustaining social interactions (Bortoli & Brown, 2008). The students with ID educated in inclusive classrooms have also been found to spend more time alone and have fewer reciprocal friendships compared to their typically developing peers (Kemp & Carter, 2002). There is a positive relationship between ID severity and delays in social and communication areas; the higher the ID severity, the more delays in social and communication skills (de Bildt et al., 2005).

Individuals with ID experience difficulties in academic performance in school settings. Their average academic achievement is lower than that of students with emotional-behavior disorders and students with learning disabilities (Sabornie, Cullinan, Osborne, & Brock, 2005). As compared to students with other disabilities, students with ID are more likely to have problems with course work, need support from teachers, and receive assessment modifications and less likely to be prepared for postsecondary education (Lipscomb et al., 2017).

H.-M. Chiang (✉)
Faculty of Education, University of Macau,
Taipa, Macau, China
e-mail: hchiang@um.edu.mo

K. Kemp
Educational Consultant, New York, New York, USA

J. L. Matson (ed.), *Handbook of Intellectual Disabilities*, Autism and Child Psychopathology
Series, https://doi.org/10.1007/978-3-030-20843-1_42

Individuals with Disabilities Education Act and Students with Intellectual Disability

The Individuals with Disabilities Education Act (IDEA, 2004) regulates special education services provided to students with disabilities including students with ID. ID is one of the disability categories identified in IDEA for which a student may qualify for and receive special education services. The US Department of Education develops regulations to support implementation of IDEA. These regulations define ID as:

> … significantly sub average general intellectual functioning, existing concurrently with deficits in adaptive behavior and manifested during the developmental period, that adversely affects a child's educational performance. [34 CFR Section 300.8 (c) (6)]

Students with ID may also receive special education services under IDEA from the disability category of multiple disabilities. While ID is not inherently part of the multiple disabilities category, many individuals served under this category may have ID. The regulations define multiple disabilities as:

> ... concomitant impairments (such as mental retardation-blindness or mental retardation-orthopedic impairment), the combination of which causes such severe educational needs that they cannot be accommodated in special education programs solely for one of the impairments. Multiple disabilities does not include deaf-blindness. [34 CFR Section 300.8 (c) (7)]

There are numerous requirements detailed in IDEA that impact special education services of students with ID including (1) the Individualized Education Program (IEP), (2) education in the least restrictive environment (LRE), and (3) access to the general curriculum. Each student with a disability should receive an IEP, and an IEP must be developed and reviewed annually. When developing an IEP, the student's educational team must consider the concerns of the parents or guardians, the results of recent evaluations, the strengths of the student, and the academic and functional needs of the student. There are several required components for an IEP, including (1) a description of the student's present levels of academic and functional performance; (2) annual academic and functional goals; (3) methods of progress monitoring of annual goals; (4) special education, related, and supplementary aids and services; (5) explanation of services provided outside of the general education classroom or in settings separate from same-age peers; (6) accommodations provided during state- and district-wide assessments or if the student will take an alternate assessment; (7) the start date, frequency, duration, and location of services; and (8) a transition plan for students who are age 16 or older (IDEA, 2004). The information detailed in the IEP provides a framework for the student's educational programming on an annual basis. Information regarding the impact of the student's disability on participation and progress in the general education curriculum and age-appropriate activities must be included in the present levels of performance. The annual goals are often the academic and functional goals deemed most important for the student to meet their educational or functional needs and to make progress in the general education curriculum.

IDEA (2004) requires that individuals with disabilities be provided services in the LRE, which refers to maximum participation and receipt of services in the general education classroom with same-age peers. Supports and services may be provided to students with disabilities to increase opportunities for inclusion and access to the general education classroom. In the event that special education services will be provided to students outside of the general education classroom, an individual student's IEP must indicate the reasons that services cannot satisfactorily be provided in general education settings. IDEA (2004) has noted that special education should be viewed as a service and not a place in which students with disabilities receive their education.

The requirements that students be educated in the LRE and that the general education classroom be considered the first placement option have led to increased numbers of students with disabilities receiving some or all of their education and services in the general education classroom. The proportion of students with disabilities educated in various education environments is reported on an annual basis. In the 2017 Annual Report to Congress (US Department of Education, 2017), which reports the data from 2015, 16.5% of students with ID spent 80% or more of their school day in the regular classroom, 26.3% spent 40–79% of their day in the regular classroom, and 49.7% of students with ID spent 40% or less of their school day in the regular classroom. Furthermore, 7.4% of students with ID were educated in other placements such as separate schools, residential facilities, home or hospital settings, correctional facilities, or private schools. According to the report, 13.3% of individuals with multiple disabilities are included in the regular classroom for 80% or more of the day, 16.5% spent 40–79% of the day in the regular classroom, 46.2% spent 40% or less of the day in the regular classroom, and 25% were educated in other settings. While data indicates that more students with disabilities are being educated in general education classrooms, individuals with ID and multiple disabilities spend less time in general education classroom than students with other disabilities. However, the Annual Report to Congress regarding educational setting does not provide a high level of detail regarding placement decisions and individual characteristics of students. It is important to consider that students with ID are a diverse group, and information regarding educational setting is not reported for students with ID with varying support needs. Research does indicate that secondary students with ID who are considered to have “low” cognitive functioning are less likely to be educated in regular schools with same-age peers than students with ID who are considered to have “moderate” or “high” cognitive functioning (Yu, Newman, & Wagner, 2009). In a study examining placements of students who took alternate assessments based on alternate achievement standards, those who had lower communication skills, used augmentative or alternative communication, and had lower academic skills in reading and math were less likely to be educated in inclusive classroom settings (Kleinert et al., 2015). It should be noted that the students who took alternate assessments were those who had the most significant cognitive disabilities. Yu et al. (2009) further found that the rate of the secondary students with ID who took at least one special education course was higher than that of the secondary students with ID who took at least one general education course.

IDEA (2004) further requires that individuals with disabilities be provided opportunities to access the general education curriculum. Annual goals must be written to support progress and involvement in the general education curriculum. Furthermore, special education, related, and supplementary aids and services must be described and provided to support access to the general education curriculum that is provided to all students. The Every Student Succeeds Act (ESSA, 2015), which replaced No Child Left Behind, also requires that students with disabilities have access to the general education curriculum and participate in the same statewide accountability measures as their peers. The ESSA also puts a 1% limit on the number of students who may take an alternate assessment based on alternate achievement standards, which is limited to students with significant cognitive disabilities. This limit results in the majority of the students with disabilities taking the same assessments, with or without accommodations, as their same-age peers. Nonetheless, this new policy ensures that students with disabilities continue to have opportunities to access the general education curriculum and their educators have high expectations for these students’ progress and performance in the curriculum.

Research on access to the general education curriculum and education service in the general education classroom provides important findings on the educational experiences of students with ID. In a review of research studies, Freeman and Alkin (2000) found that students with ID who were more fully integrated or spent more substantial amount of time in general education classrooms had better academic and social outcomes than students with ID who spent less time in general education settings. Wehmeyer, Latting, Lapp-Rincker, and Agran (2003) found that students with ID in inclusive settings were more likely to work on the activities connected to academic standards than those in special education settings. Last, Soukup, Whmeyer, Bashinski, and Bovaird (2007) found that students with ID who were included in general education classrooms had more access to the general education curriculum than those who were not.

Curriculum Development for Individuals with ID

A curriculum consists of sequential planned learning activities. It can be conceived in a narrow way as subjects taught or in a broad way as whole learning experiences (Oliva, 2005). A curriculum used in an intervention program for individuals with ID may be considered as the sum of the sequential learning experiences of learners (Johnson, 1968; Shepherd, 1992). Having a well-designed curriculum is the first step to provide effective learning experiences for individuals with ID.

Steps for Developing a Curriculum

There are five steps that may be used for designing a curriculum. The first step is to understand the needs of individuals with ID. The second step is to design intervention goals based on the observed needs. The third step is to select the assessments that will be used to determine whether a teaching activity or an intervention has met its goals. The fourth step is to design lesson/instructional plans for each teaching activity/intervention session. The last step is to select the instructional strategies that will assist a teacher/therapist to effectively deliver a lesson/intervention session.

Step 1. Understand Clients' Needs

Individuals with ID have intellectual and adaptive functioning deficits in conceptual, social, and practical domains (APA, 2013). However, because of family and cultural background and other personal factors (e.g., gender, age, health, abnormalities in gene(s)), intellectual and adaptive functioning abilities may be different in each learner with ID. Thus, it is important to understand a learner's needs and skills before designing a customized curriculum or adopting an existing curriculum.

Various methods (e.g., interview, observation, and assessment) can be used to understand the needs of a target learner with ID.

Interview. Interviewing parents allows teachers/therapists to understand a learner's needs from parents' points of view. When working with parents from culturally diverse backgrounds, culturally sensitive approaches should be applied. For example, an interpreter who is fluent in both of the language used by parents and the language used by the interviewer should be arranged during parent interviews. If such a service is not available, a translated version of the questionnaire(s) to be used during the interview should be made available to the parents, and parents should be encouraged to read and ask questions before the interview. An interviewer should have basic understanding of interviewee's cultural background so he/she knows to avoid culturally inappropriate topics/questions and demonstrate culturally appropriate courtesy. A sample of the list of the questions that may be used during a parent interview is listed below:

(a) *Parents characteristics*. The questions in this section are used to gather information about the characteristics of parents. The questions can include parents' age, educational background, household income, and career.
(b) *Child characteristics*. The questions in this section are used to gather information about the learner's characteristics. The questions can include learner's age, gender, grade level, the most current IQ score as measured by a standardized test, the most current adaptive skills levels as measured by a standardized test, any other diagnosis besides ID, health conditions, interests, strengths, weaknesses, and job preferences if appropriate.
(c) *Needs*. The questions in this section are used to understand a learner's needs based on parents' perspectives. The questions can be divided into two categories: (a) for learners who are in school and (b) for learners who have left school.

For learners who are in school. Sample questions can include: What difficulties does your child have in school? What difficulties does your child have at home? What difficulties does your child have in community? What goals do you have for your child at school? What goals do you have for your child after high school? What do you want this intervention to help your child?

For learners who have left school. Sample questions can include: What difficulties does your child have at work or day rehabilitation center? What difficulties does your child have at home? What difficulties does your child have in community? What goals do you have for your child in the next 5 years? What do you want this intervention to help your child?

Observation. Observation in naturalistic environments can reveal a learner's abilities in real-life situations. Given that behavior expectations can be varied across settings and a learner may function differently in different settings, observation should be conducted across various settings (e.g., school, home, community). A sample of the questions to guide observation may include:

(a) How does the learner function when performing an activity as compared to his/her peers without disabilities? For example, does the learner require a longer time to complete the activity? Does the learner need extra support? Does the learner finish the activity? Does the learner behave appropriately during the activity?
(b) How does the learner feel about the activity? For example, does he/she seem to enjoy the activity? Does he/she use facial expression/body gestures/language to indicate he/she wants to do the activity again? Does he/she seem to not like the activity or feel nervous about it? Does he/she use facial expression/body gestures/language to reject the activity?

Standardized assessments. Standardized assessments can be conducted to reveal a learner's current functioning level as compared to same-age peers without disabilities and his/her relative strengths and weaknesses as compared to his/her own abilities. Several assessment tools are available for examining adaptive behaviors of individuals with ID. Two of the most commonly used assessment tools are:

(a) Adaptive Behavior Assessment System, Third Edition (ABAS-3) (Harrison & Oakland, 2015). This assessment can be used on individuals aged 0–89 years. The administration time for this assessment is 15–20 min.
(b) Vineland Adaptive Behavior Scales, Third Edition (Vineland-3) (Sparrow, Saulnier, & Cicchetti, 2016). This assessment can be used on individuals aged 0–90 years. The administration time is about 20 min for the interview form and 10 min for the parent/caregiver and teacher forms.

Step 2. Set Up Goals

After knowing a learner's needs, the next step is to determine the desired outcomes that an intervention aims to achieve. Depending on the nature of the intervention, various types of goals may be addressed through an intervention. The intervention goals can be generally grouped into two basic categories: remedy weakness and cultivate strength.

*Remedy weakness***.** It is very common that the focus of an intervention for individuals with ID is on their deficits. In fact, why a student with a disability can receive special education and other related services is because his/her disability affects his/her education performance (Public Law 108–446, 2004). Thus, the interventions conducted in school and the IEPs for students with ID are mainly designed to address their deficits. Addressing the deficits of individuals with ID represents an effort to reduce the gap between the abilities of these individuals and their same-age peers without disabilities. However, remedy deficits, this purpose, often bring low expectation on individuals with ID, and low expectation can result in someone not to learn something challenging enough to promote his/her learning motivation, constantly learning the skills he/she has already acquired, or learning not age-appropriate skills.

Deficits are one's *not good* abilities. No one likes people always talking about his/her *not good* abilities. However, individuals with ID may experience their deficits being mentioned over and over when receiving the intervention focusing on remedy deficits. If an intervention does not concern how an individual with ID may feel, his/her self-esteem and self-confidence may potentially be harmed.

Cultivate strength. Cultivate strength takes a positive approach to address the needs of an individual with ID. Instead of focusing on deficits, this approach concerns the strengths of an individual with ID and aims to bring the strengths to the next level and transform the strengths to the skills that an individual with ID can use to improve their quality of life. Because of the label, ID, strengths of individuals with ID are often overlooked. However, each individual with ID has his/her own strengths. Their strengths can be discovered by conducting strength-based assessment (Cosden, Koegel, Koegel, Greenwell, & Klein, 2006). Questions such as *What are the learner's interests? When does the learner enjoy interacting with another person?* can be used to explore learners' interests (Cosden et al., 2006). Focusing on strengths can have several benefits on individuals with ID (e.g., improving learning motivation, self-confidence, self-esteem, and quality of life). However, the number of the interventions focusing on cultivating strengths is much smaller than that focusing on remedying deficits. More interventions that can transform the strengths of individuals with ID to the skills that they feel proud of themselves should be encouraged.

An intervention can aim to address a single goal or multiple goals depending on the length of the intervention and available resources. An intervention goal can be conceptualized as a specific skill or cluster of skills that a learner will acquire during intervention (Sailor & Guess, 1983). For each intervention goal, there should be short-term objectives that are measurable intermedia steps between a client's current level of performance and an intervention goal (Fiscus & Mandell, 1983). Both goals and objectives should be stated in a clear, precise, and measurable way. The *Case Study of Mark* illustrates intervention goals and objectives.

Case Study: Martin

Martin is a 10-year-old boy with ASD who also has moderate ID. He loves drawing and is very good at drawing. He can follow directions and complete assigned tasks with minimal prompting. However, he has significant delays in language skills, social interaction skills, academic skills, and independent living skills.

	Intervention goals	Short-term objectives
Language	Martin will answer *Wh* questions Martin will request desired items verbally	When presenting with *Who* questions, Martin will verbally answer the questions correctly four out of five trials without prompts When presenting with a desired item, Martin will say, *Can I have xx (the name of the item)?* four out of five trials without prompts
Social interaction	Martin will initiate social interactions with peers Martin will respond to social interactions initiated by peers	When seeing peers are playing cars, Martin will say, *Can I play with you?* four out of five trials without prompts When peers ask him to play with them, Martin will join them four out of five trials without prompts
Academics	Martin will solve addition problems with sums up to 50 Martin will write a paragraph including at least three sentences about an interested topic	When presenting with addition problems with sums up to 20, Martin will solve the questions correctly four out of five trials without prompts When presenting with pictures, Martin will write two sentences without errors on four out of five trials without prompts
Independent living	Martin will place cell phone calls Martin will wear a coat when it is cold outside	Given his mom's cell phone number, Martin will successfully place the phone call on four out of five trials without prompts When presenting with weather information, Martin will wear a coat when going outside on four out of five trials without prompts

Step 3. Determine Assessments

In order to understand whether an intervention has achieved its goals, methods to evaluate a learner's learning outcome should be determined. A comprehensive evaluation plan should be determined before starting an intervention. Various evaluation tools can be used, and they can be grouped into two categories: formal standardized tests and informal procedures.

*Formal standardized tests***.** These tests can be either norm-referenced or criterion-referenced (Hickson, Blackman, & Reis, 1995). Norm-referenced tests allow interventionists to compare a learner's performance to that of same-age peers. Criterion-referenced tests allow interventionists to measure a learner's performance against predetermined criteria. These tests often are commercially published tests (e.g., intelligence tests, academic achievement tests, language tests, adaptive behavior tests) which can be purchased through publishers.

*Informal procedures***.** These assessment procedures include observation, work sample analysis, task analysis, checklists, rating scales, interviews, and questionnaires (Hickson et al., 1995). These assessments are often made by interventionists to determine whether learners have acquired target skills that they have been specifically taught.

The effectiveness of an intervention can be determined through carefully following its evaluation plan. The evaluation plan should consist of assessment material(s) and an evaluation design. Depending on the number of participants in an intervention, the evaluation design can be a single-subject design or a pretest-posttest group experimental design.

*Single-subject design***.** This design allows the teacher to determine the intervention effects on a single participant or a small group of participants. There are three commonly used single-subject designs including the reversal or withdraw (ABA) design, multiple baseline design, and changing criterion design (Rusch, Rose, & Greenwood, 1988).

*The reversal or withdraw (ABA) design***.** This design consists of three phases: baseline 1 (A), intervention condition (B), and baseline 2 (A). During baseline 1, the target behavior(s) is/are observed without any presentation of intervention. During intervention, the participant(s)' target behavior(s) is/are observed while receiving intervention. After some stable and substantial changes of the target behavior(s) are observed, the treatment condition will be withdrawn. The baseline condition will then be reintroduced, for example, if one wishes to determine whether using participants' interested items would increase their attending behaviors using an ABA design. If the numbers of participants' attending behaviors during the intervention condition are higher than those during the baseline 1 and baseline 2 conditions, it can be concluded that the intervention was effective.

*The multiple baseline design***.** This design can be used across subjects, behaviors, or settings (Hickson et al., 1995). Baseline data are collected on the same behavior across two or more participants, two or more behaviors on the same participant, or on the same behavior across two or more settings for the same participant (Christensen, Burke Johnson, & Turner, 2011). After baseline data are collected, the intervention will be introduced. If the number of target behavior(s) recorded during the intervention condition is noticeably higher than that during each baseline condition, it can be concluded that the intervention was effective.

*The changing criterion design***.** This design begins with the collection of baseline data. During the first intervention phase, a criterion level is determined. If a participant has successfully reached the criterion level across several trials, a higher criterion level will be introduced during the next intervention phase. If a participant's target behavior improved as the criterion level was increased, this overall pattern of results is the evidence of the positive results of the intervention on the participant (Christensen et al., 2011).

*Pretest-posttest group experimental design***.** There are two types of this design. One allows a comparison of the mean performance of the same participants before the intervention and after the intervention (i.e., one-group pretest-posttest design). Another one allows the comparison of two groups (intervention vs. nonintervention) before and after the intervention (i.e., pretest-posttest control-group design).

One-group pretest-posttest design. Before starting the intervention, all participants receive pretests. After pretests, all participants begin to receive the intervention. Upon completion of the intervention, all participants receive posttests which are the same as the pretests. The differences between the pretest and posttest performances of the participants can be determined by using statistical analyses. If participants' posttest scores are significantly higher than their pretest scores, it may be concluded that the intervention has positive influences on the participants.

Pretest-posttest control-group design. Participants are randomly assigned to either of the intervention group or the nonintervention (control) group. Both groups will then receive pretests. After pretests, only the intervention group will receive the intervention. After the intervention has completed, both groups will then receive posttests. Statistical analyses are used to determine if the intervention effects exceed what would be expected by chance (Hickson et al., 1995).

Step 4. Design Lesson/Instructional Plans

Each intervention session should have a lesson/instructional plan to guide an interventionist to deliver instructional activities to address the needs of a learner(s). This plan specifies the time, duration, teaching materials, and learning content for a to-be-delivered activity(ies). It helps interventionists stay organized and ensure intervention time is used efficiently and effective instruction is delivered to help learners acquire target skills (Selmi, Gallagher, & Mora-Flores, 2015). A lesson plan does not always need to be written in great details, but it has to be completed before starting an intervention session. A lesson/instructional plan should at least include the sequence of learning activities, instructional time for each learning activity, and the learning materials to be used.

Three lesson plan samples targeting different skills at different grade levels are presented below.

Lesson Plan 1: Teaching Functional Math Skills

Title: Count objects to 20	Grade level: K	Time: 45 min
Standard: CCSS.MATH.CONTENT.K.CC.B.5Count to answer "how many?" questions about as many as 20 things arranged in a line, a rectangular array, or a circle, or as many as 10 things in a scattered configuration; given a number from 1 to 20, count out that many objects.	**Objective:** When given a number, students will count the given number from 1 to 20.	
Context: This is an inclusive kindergarten classroom where students with ID are educated with their typically developing peers in the same classroom. There is a general education kindergarten teacher and a paraprofessional in the classroom full time. During math instruction, a special education teacher joins the classroom to co-teach. At the beginning of math instruction, the teacher introduces the skill to the class as a whole group. After the topic has been introduced, students participate in centers that are designed to align to and practice the skill. Teachers support students during the centers. Snack time also occurs during math centers because of the many opportunities to practice math during snack. Whenever possible, the teacher designs an activity at the snack table that corresponds to the math skill. There are four centers, including snack. The teacher divides students into heterogeneous groups of four. Students rotate through centers, spending 8 min at each center.		
Materials: • SMART Board at level that students can reach. The SMART Board is set up as an upside down "T" grid with 20 math manipulative images at the bottom and number dice on left side. • Set of up to 20 manipulatives for each student in a bowl. • Tray for each student. • 5 × 4 grid for some students. • Beanbags. • Hula-hoop. • Multisided dice with numbers. Dice are available in various sizes and with different difficulty. Some dice have smaller sets of numbers (1–6), while other dice have larger numbers (1–20). • Game board. • Math manipulatives. • Number cards. • Grapes for snack. • Trays, plates, and 5 × 4 grid for some students.		

(continued)

Lesson implementation:

1. Each student is seated on the classroom rug in front of the SMART Board. There are seating options for students (e.g., carpet squares, textured cushions, chairs, etc.). Each student has a tray and bowl with up to 20 math manipulatives. For some students, the bowl is attached to the tray. Some students also have a 5 × 4 grid attached to their tray. The teacher introduces the lesson by showing students a game on the SMART Board. The teacher touches the dice on the SMART Board to roll the dice. The teacher and class then count out the number of items shown on the dice. The teacher moves the correct number of manipulatives from the bottom to the right side of the grid. Students count the number of items on to their tray. Students with the grid attached to their tray place the manipulatives in the grid as they count. Teachers help students as needed. After the class has counted, the teacher moves the SMART Board manipulatives back to the bottom of the grid. The teacher repeats this process several times, calling on students to help her at the SMART Board. After 4–5 repetitions, the teacher introduces and models each center to the students.
2. Center 1: Students in this center continue to play the SMART Board game.
3. Center 2: This center takes place on the rug. It is placed far enough away from the SMART Board, so as not to distract students in either center. In this center, students take turns rolling a dice. A variety of dice are available, and a teacher helps students select an appropriate dice. Students then toss the corresponding number of beanbags into a nearby hula-hoop. Students may stand as close to the hula-hoop as needed. A teacher spends time on the rug supporting students at Center 1 and Center 2 as needed.
4. Center 3: Students in this center play a game. The game board has four racetracks with square spaces. Students may select any math manipulative they would like to use. Students take turns drawing a number card. Students then place the correct number of their chosen manipulatives on the square spaces. A teacher is seated at this center to support students and uses modeling and prompting as needed.
5. Center 4: Snack. Students count out 20 grapes at snack. Some students have a 5 × 4 grid available to support counting. A teacher is seated at this center to support students, and uses modeling and prompting as needed. The teacher also facilitates discussion with students during snack.

Assessment:

Teachers keep data sheets at each center. Teachers record the number(s) rolled/drawn and accuracy of counting objects. Teachers also record any prompting that was needed.

Lesson Plan 2: Teaching Functional Language Skills

Title: I love reading	Grade level: 4th	Time: 45 min
Standard: CCSS.ELA-LITERACY.RL.4.3Describe in depth a character, setting, or event in a story or drama, drawing on specific details in the text (e.g., a character's thoughts, words, or actions). CCSS.ELA-LITERACY.SL.4.1 Engage effectively in a range of collaborative discussions (one-on-one, in groups, and teacher-led) with diverse partners on *grade 4 topics and texts*, building on others' ideas and expressing their own clearly.	**Objective:** Student(s) will answer *Who* and *What* questions about characters in a story.	

Context:

This lesson takes place in an inclusive fourth grade classroom. There is one general education teacher and one special education teacher in the classroom. There are four students with disabilities. One of the students has ID. Students in the class read a short story and work in groups to identify and describe the main characters. The learning groups are heterogeneous, and teachers circulate the room to support students.

Materials:

- Short story at fourth grade reading level
- Adapted version of story with visuals added and text reduced
- Audio version of story
- Graphic organizer for students to identify characters and their traits
- Visuals cut out from adapted version of story
- *Wh* question prompt cards

Lesson implementation:

1. Students read the story independently. Some students may also use an audio version of the story. The student with ID uses the adapted version of the story.
2. Students are given time to begin completing their graphic organizer about the story's main characters. A teacher works with the student with ID to complete his graphic organizer. The student has visuals available from the adapted story to complete the graphic organizer. The teacher asks the student *Who* and *What* questions while completing the graphic organizer.
3. Students engage in a group discussion about the characters in the story. Students share with the group their thoughts about the characters by describing their completed graphic organizer. Students ask each other *Wh* questions during the group time. Students may use the *WH* prompt cards.

Assessment:

The teacher collects the graphic organizers. The teacher records the student responses to *Wh* questions, including any prompting needed to answer questions.

Lesson Plan 3: Teaching Community Living Skills

Title: Making lunch with friends	Grade level: 8th	Time: 40 min
Standard: NA	**Objective:** Students will follow multistep directions to make sandwiches for lunch.	

Context:

This lesson takes place during an inclusive lunch group with four students, two with ID and two with typical development. The purpose of this group is to focus on skills of daily living, social skills, community engagement, and academic skills. This group meets for lunch twice a week, as well as participating in other school activities such as working at the school store, tending to the school garden, and participating in other school initiatives. The primary role of students with typical development in the group is to be a peer and friend. These students with typical development have learned strategies for prompting, modeling, reinforcing, and engaging in interactions with students with ID.

Materials:

- Task analysis with images for making a sandwich
- Video model of making a sandwich
- Food items for lunch: lettuce, tomato, bread, cheese, turkey, ham, mayonnaise, mustard
- Drinks for lunch
- Plates
- Napkins
- Cups

Lesson implementation:

1. Prior to the lunch group, the teacher meets with students with ID to watch the video model and cover the steps of the task analysis. The video model and task analysis were designed to align with each other. As students watch the video, the teacher discusses each step of the task analysis. The teacher asks questions as appropriate to probe for comprehension.
2. Students meet in one of the school's cooking labs for lunch.
3. Students split in to two groups; each group has one student with ID and one student with typical development.
4. Students work at one of the cooking tables to make their sandwiches. Students follow the task analysis to gather the materials and make their sandwich. Students check off each step of the task analysis as it is completed. A teacher is present to support students as needed. However, students are prepared to support each other as well.
5. Once each student has made their sandwich, the group sits down to eat lunch together. While eating, the students engage in planned topics of conversation.
6. When students have finished eating, they clean up together and play a game of their choice.

Assessment:

The teacher monitors students while making the sandwich. She keeps a checklist to mark each step the student completed, as well as if the step was completed independently or with prompts.

Step 5. Select Instructional Strategies

After goals are determined, the teaching strategies that will be used during intervention should then be decided. Teaching strategies are the instructional tools that interventionists use to assist learners to acquire target behaviors. They include instructional methods and behavior management techniques (Hickson et al., 1995). Appropriate teaching strategies should be selected based on the philosophical orientations of the intervention program, target behaviors, and students' ability levels.

Instructional methods. There are various evidence-based instructional methods for teaching individuals with ID, and they can be grouped into four categories, including skill acquisition, motivation improvement, skill maintenance, and skill generalization.

Skill acquisition. Various approaches can be used to teach individuals with ID to acquire new skills.

Modeling. This is a teaching strategy that an adult or a peer demonstrates a target behavior to a learner (Wong et al., 2013). For example, the target behavior is saying *water*. An adult demonstrates saying *water* to a learner. Another form of modeling is *video modeling* which offers a visual model of the target behavior via videos (Wong et al., 2013).

Prompts. Prompts, such as verbal, gestural, or physical assistance, are given to a learner to assist him/her to learn a target skill (Wong et al., 2013). For example, a learner is learning to say *car*. An adult presents a toy car and provides a verbal instruction (say car) to verbally prompt the learner to acquire the skill of saying car. An example of the gestural prompt is to point to a toy car and wait for a learner to say car. If an adult wants to use physical assistance to prompt a learner to say car, he/she will hold a learner's hand and put it on a toy car. Adults can provide prompts to learners in several ways: (a) the least-to-most prompting system, (b) the most-to-least prompting system, (c) simultaneous prompting, and (d) flexible prompt fading.

(a) The least-to-most prompting system is that adults systematically increase guidance to assist learners to acquire the target skill(s).

For example, adults first use verbal prompts, then verbal prompts + gestural prompts, and lastly verbal prompts + gestural prompts + physical prompts to guide a learner to learn a target skill (Horner & Keilitz, 1975; Murzynski & Bourret, 2007).

(b) The most-to-least prompting system is that adults start from providing the most assistive prompt and the prompting system ends with no prompt. For example, adults use physical prompt first, then gestural prompt, verbal prompt, and lastly provide no prompt (Leaf et al., 2016).

(c) Simultaneous prompting is a procedure that adults provide a prompt that results in a learner making a correct response 100% of the time immediately following instructions. This makes a learner no chance to error and always receive reinforcement (Leaf, Sheldon, & Sherman, 2010). For example, adults want to teach learners to learn a concept (e.g., long). Adults will say "Which one is long?" and then immediately point to the correct picture and say "This is long." Adults will then wait for 2 s to get a learner's response. If the learner answers correctly, he/she will be rewarded right away. If the learner does not answer correctly, adults will repeat the procedures: "Which one is long?" "This is long" (Celik & Vuran, 2014).

(d) Flexible prompt fading is that adults make moment to moment assessments in deciding which type of prompt to be provided and when to assist a learner to learn a target skill, as opposed to adhering to a predetermined system (Leaf et al., 2016). For example, adults want to teach a learner some object labels. Adults will predetermine how likely the learner will say the names correctly. If adults determine that the learner is likely to say the names correctly, adults will not provide any prompt to the learner. However, if the learner will likely to respond correctly with a more assistive prompt, adults will provide this type of prompt to the learner (Leaf et al., 2016).

Reinforcement. A learner's desired item or activity is given to him/her once he/she has produced the target behavior. For example, a learner says *water* to request drinking water. An adult heard the learner saying water. He/she then allows the learner to drink water. Reinforcement should be provided to a learner immediately after he/she has displayed a target skill. After a learner has successfully shown a target skill several times, adults should begin to fade the reinforcement delivery gradually. The goal of fading reinforcement is to train a learner to display a target behavior independently without relying on any prompt.

Time delay. This is a strategy that adults allow time between the presentation of an instruction request and the presentation of a prompt (Snell & Gast, 1988). There are two types of time delay: (a) constant time delay and (b) progressive time delay.

(a) Constant time delay is a procedure that adults allow a fixed delay between a task request and prompt (Ault, Wolery, Gast, Doyle, & Eizenstat, 1988). For example, adults want to teach a learner to write his/her name. Adults demonstrate how to write the learner's name and then ask the learner to write his/her name. Adults will wait 5 s for the learner's response. If the leaner successfully writes his/her name, adults will provide a reinforcement. If the learner does not successfully write his/her name, adult will use a prompt to assist the learner to demonstrate the target behavior. Adults will wait for 5 s for the learner's response every time they have made a task request.

(b) Progressive time delay is a procedure that adults gradually increase the amount of the time between the presentation of a task request and the presentation of a prompt (Wolery, Gast, Kirk, & Schuster, 1988). Using the example described above that adults want to teach a learner to write his/her name, adults will wait for 5 s for a learner's response for the first trial. They will then increase the waiting time to 10 s for the second trial, 15 s for the third trial, 20 s for the fourth trial, and go on if more trials are required.

Task analysis. It is a strategy that an instructor breaks down a target behavior into several small continuous steps and teaches these steps to students (Anderson, Taras, & Cannon, 1996). Adults can teach the behavior from the first step to the last one or backward (from the last step to the first one) using modeling and prompting procedures (Anderson et al., 1996). For example, the procedures of making photocopies can be broken down to these steps: (a) turn on the photocopy machine, (b) place paper on copier, (c) select quantity, (d) hit the copy button, and (e) collect the copies. If an adult aims to teach a learner how to make photocopies, he/she can model each of the steps from step (a) to step (e) and then ask the learner to practice each step. While the learner is practicing each step, adults can take notes on the prompt level required by the learner. For example, if a learner can complete step (a) independently, the individual instruction for the learner can skip step (a). If a learner only has difficulty in placing paper on the copier, the individual instruction for this learner may only focus on this behavior.

Performance feedback. This is a strategy that adults provide supportive and corrective responses to a learner's performance during instruction (Gilson, Carter, & Biggs, 2017). For example, using this strategy to teach a learner to learn math, adults will provide performance feedback immediately following the learner's response. If a learner solves a math problem correctly, adults will let the learner know whether his/her answer is correct and provide a reinforcement to the learner. However, if a learner does not solve a math problem correctly, adults will use supportive comments (e.g., "I see you get this part right, but the other part should be done in this way") to give feedback.

Device-assisted instruction. This strategy uses computer or other technological devices (e.g., tablet, mobile phone, digital watch) to facilitate a learner's learning (Gilson et al., 2017). For example, adults can teach learners with ID who have not developed verbal skills to use an aided augmentative and alternative communication (AAC) device (e.g., communication board, computer-based technology with voice output) to communicate.

Mnemonics. This strategy uses words that provide acoustic reconstructions of unfamiliar information (Gilson et al., 2017). For example, to help learners remember the continents, Africa, Antarctica, Australia, Europe, Asia, North America, and South America, adults can take the first letter out of these continent words and arrange them into a new word, AAAEANS. Adults will use a silly sentence: Three ants eat apples North and South to help students remember it (Frank & Smith-Rex, 2015).

Peer-mediated instruction. This type of instruction utilizes peers of learners with ID to promote their target skills. For example, adults can train typically developing peers of learners with ID to use time delay to help these learners' improve their grade-aligned science skills (Jimenez, Browder, Spooner, & Dibiase, 2012).

Incidental teaching. This is a naturalistic instruction that adults arrange the environment and materials to increase a learner's initiated communication and respond to the learner's communication with modeling, prompts, and reinforcement (Hart & Risley, 1982). For example, adults want to teach a learner to request markers. Adults can put markers in a clear container where it is located at a bookshelf where the learner can see but cannot reach. When the learner uses facial expression and/or gestures to request adults to deliver the markers to him/her, adults model "I want markers." and instruct the learner to say the sentence. Adults will wait for the learner's response. If the learner correctly says the sentence, adults will deliver the makers to the learner. However, if the leaner does not say the sentence correctly, adults will use prompts to facilitate the learner to say the correct sentence.

Motivation improvement. Motivation is one of the pivotal areas that can have positive influence in the learning of children with autism spectrum disorder and ID (Koegel, Koegel, Harrower, & Carter, 1999). Motivation can be defined as "observable characteristics of a child's responding" (Koegel et al., 1999, p. 178). Improvement in motivation can be described as increased responsiveness to social and environmental stimuli (Koegel, Carter, & Koegel, 1998). If a learner has high level of motivation in learning, it will be easier for interventionists to provide instruction, and it will also be easier for the learner to learn new skills. Thus, it is important to apply strategies to improve learners' learning motivation. Several strategies can be used to improve learner's learning motivation. They include child choice, interspersal of acquisition and maintenance tasks, natural reinforcement, and reinforcing attempts.

Child choice. Allow learners to choose preferred items or activities, and the items or activities chosen by them are then used in or as the learning activities (Koegel et al., 1999). This simple strategy can effectively improve learners' learning motivation. For example, an adult asks a child if he/she would like to use a pen or marker to do writing. If the child wants to use a marker, he/she is allowed to use it for writing (Koegel, Singh, & Koegel, 2010).

Interspersal of acquisition and maintenance tasks. Students often are not interested in completing the tasks that they feel difficult. Learning new tasks often make students feel challenging and want to escape. However, in order to achieve intervention goals, students are required to complete challenging tasks. Blending difficult tasks with easy tasks can increase students' learning motivation. Numerous previously learned tasks are interspersed with the target skill that the learner has not already learned (Koegel & Koegel, 2012). For example, a learner can add two numbers less than 10 to yield a sum smaller than 10 but is learning the tasks adding two numbers less than 10 to yield a sum greater than 10. The easy questions can be interspersed with the difficult ones to increase students' learning motivation.

Natural reinforcement. Another strategy to improve motivation is to use natural reinforcers. A natural reinforcer is the item/activity that is directly and functionally related to the target task (Koegel & Koegel, 2012). Once a learner displays a target behavior, he/she should receive the item or activity that is directly related to the target behavior. For example, a cup of water is given to a learner once he/she said *water*.

Reinforcing attempts. Before a learner can successfully display a target behavior, he/she may need to practice the behavior many times. Depending on the learner's learning speed, his/her learning process may be long. If interventionists only provide reinforcement to learners when they successfully produce target behaviors, they may lose motivation to learn new skills. Thus, reinforcing learning attempts while a learner is trying to acquire a new skill is important. Reinforcing attempts means that a learner's goal-oriented attempts are reinforced (Koegel & Koegel, 2012). For example, a learner is learning to name balloon and wants to play with a balloon, but he/she has difficulty sounding the whole word. He/she looked at the adult and said, *ba*. The adult reinforced his/her attempt by giving him/her a balloon.

Skill maintenance. Maintenance refers to learners continue to display learned behaviors after an intervention has completed. Several strategies may be used to improve skill maintenance (Westling & Fox, 2009).

Continue practice. After learners have acquired a target skill, he/she should be taught to continue to practice the skill.

Multiple opportunities to practice. Learners should be given opportunities to practice one skill at different times, instead of all at once.

Intermittent reinforcement. Once a learner has learned a target skill, reinforcement should not be given to the learner immediately but every couple trials or minutes.

Self-monitoring. This instruction teaches learners to self-monitor their own behaviors (Strain, Kohler, Storey, & Danko, 1994). For example, a learner often forgets to complete his/her homework. Adults can work with the learner to create a homework checklist and teach him/her to write down the list of homework and cross out the ones he/she has completed. The learner can earn a desired item as a reward after he/she has successfully completed all the homework.

Skill generalization. Generalizing a learned skill to a novel situation or environment may be difficult for individuals with ID (Oreilly & Chadseyrusch, 1992). They may not know how to apply a learn skill to a non-training setting or non-training interaction partner. Specific strategies should be applied to promote skill generalization in learners with ID. Several strategies could be used to improve skill generalization (Stokes & Osnes, 1986).

1. The target skills should be useful to the learner's daily life.
2. The target skills should result in receiving natural reinforcers.
3. Use a wide variety of stimulus exemplars.
4. Use a wide variety of response exemplars.
5. Provide training in various training conditions and settings.

Behavior management techniques. Individuals with ID and/or autism who are nonverbal or have limited spoken language use challenging behaviors (e.g., tantrum, aggression) as a form of expressive communication (Chiang, 2008). Learners with a more severe degree of ID are more likely to display challenging behaviors than learners with a milder degree of ID (McClintock, Hall, & Oliver, 2003). Children's problem behaviors may bring challenges for interventionists to deliver interventions to them and decrease children's learning time. Thus, it is critical to apply appropriate strategies to manage learners' behaviors.

Functional analysis of problem behaviors. The first step to design a program to manage learners' problem behaviors is to conduct a thorough functional analysis of the problem behaviors (Hickson et al., 1995). This analysis is to identify the antecedent conditions that trigger the problem behavior, define the challenge (i.e., a description of the problem behavior), and identify the consequent conditions that maintain the problem behavior (Horner, Albin, Todd, Newton, & Sprague, 2011). Once these information is obtained, intervention alternatives may be introduced to learners. For example, if a learner is likely to bite his teacher to escape from the task demands. Changing the nature of the task demands to be more interesting to the learners may decrease his biting behavior. Also, teaching the learner to say *No* or point to a picture with *No* sign when he wants to reject an uninterested task may replace the biting behavior.

Extinction with positive reinforcement of other behaviors. Extinction means that interventionists intentionally withhold reinforcement following a learner's behavior or ignore a learner's behavior (Snell & Brown, 2011). Extinction is most effective when positive reinforcing other behaviors is also used (Kerr & Nelson, 1989). For example, if a learner uses screaming to get adults' attention, in order to decrease the challenging behavior, adults will not look at the learner when he screams but will look at him to praise him when he does something good.

Issues to be Considered When Developing a Curriculum

Educators must consider several important factors when developing and implementing curriculum for learners with ID in their classrooms. IDEA requires that educators develop annual goals and provide appropriate aids and services that support progress of students in the general education curriculum. This means that educators must consider methods to promote and improve access to grade-level standards and curriculum for their students. In order to do so, Ryndak, Moore, Orlando, and Delano (2008) suggest that educators must consider general education contexts (e.g., general education classrooms, peers, teachers, etc.), general education content (e.g., content standards and processes), high expectations, and participation in state- and district-wide accountability measures. Educators should adopt the strategies that can support students with ID to participate in the general education curriculum as well as help them to show progress in functional skills (Hunt, McDonnell, & Crockett, 2012).

Researchers have developed frameworks and models for promoting access to the general education curriculum for students with ID. Hunt et al. (2012) recommend using an ecological curricular framework to support student access to the general education curriculum and standard-based academics. As part of process of developing curricula as part of an ecological framework, educators consider goals that are high priority, focus on quality of life outcomes, and are relevant and meaningful to individuals, which may include academic content knowledge and skills. Hunt et al. (2012) describe a process for applying the ecological framework, which requires educators to consistently consider the needs of students with ID and their personal quality of life outcomes while promoting access to the general education curriculum. Hunt et al. (2012) suggest identifying the “critical function” of grade-level content standards in order to develop individualized performance outcomes for students with ID. Once outcomes are developed that connect the content standard to the student’s personal quality of life goals, educators must teach the skills using activities that are meaningful and relevant to the student. Trela and Jimenez (2013) also suggest the use of an ecological framework to support access to the general education curriculum for students with ID. They suggest using curricular modifications that are “personally relevant” by making connections between the student’s school community and the content being taught.

Educators may also consider promoting self-determination in order to facilitate access to the general education curriculum. The Self-Determined Learning Model of Instruction prompts students through a problem-solving process that supports self-directed learning (Wehmeyer, Palmer, Agran, Mithaug, & Martin, 2000). During instruction, students answer a series of questions that are designed to support the student to set goals and make plans to achieve their goals. Furthermore, students utilize self-directed learning strategies such as self-monitoring and self-reinforcement. The Self-Determined Learning Model of Instruction has been used successfully with students to increase outcomes such as improving public speaking and question asking, as well as academic outcomes such as scientific inquiry, understanding maps, and learning about organ systems (Agran, Cavin, Wehmeyer, & Palmer, 2006; Agran, Wehmeyer, Cavin, & Palmer, 2010). Based on the positive results of this model, educators may consider promoting self-determined behaviors and self-directed learning to promote access to the general education curriculum for students with ID.

Universal Design for Learning (UDL) should also be utilized by educators when developing curriculum and supporting access to the general education curriculum for students with ID. UDL is a framework that promotes access to the curriculum and learning environments for a variety of learners by providing flexibility and options (Meyer, Rose, & Gordon, 2014). There are three principles of UDL that are used to guide educators when developing curriculum, designing learning environments, and selecting materials and instructional methods. These principles (1) provide multiple means of representation, (2) provide multiple means of engagement, and (3) provide multiple means of action and expression (Meyer et al., 2014). Guidelines have been further provided in each of the three principles to provide suggestions for educators to consider the needs of their learners and provide flexibility when designing curriculum and instruction. Educators provide a number of options for how content is presented and delivered in their classrooms in order to meet the principles of multiple means of representation. In order to provide multiple means of engagement, educators should consider student interests, provide choices, and create relevant activities. Finally, educators provide options for students to demonstrate their knowledge in order to provide multiple means of action and expression. When educators use the UDL principles and guidelines, they create learning environments that support all of their students to access instruction and reduce barriers to learning.

Educators should also consider the need for accommodations and modifications for their students with ID when developing curriculum and supporting access to the general education curriculum. Wehmeyer, Lattin, Lapp-Rincker, and Agran (2003) describe strategies for adapting and augmenting the curriculum to support access. Curriculum adaptations are strategies that modify presentation and engagement with the curriculum. Several strategies of curriculum adaptations have been described including (1) strategies that support organization such as graphic and advance organizers, chunking, and outlines; (2) strategies that promote understanding such as comparisons, examples, and diagrams; (3) strategies that augment content presentation such as videos and stories; (4) strategies that improve demonstration such as role-play; and (5) strategies that promote recall such as keywords and visual images (Lee et al., 2006; Wehmeyer et al., 2003). Curriculum augmentation strategies are learning strategies that promote success and independence. Some augmentation strategies overlap with adaptation strategies such as the use of graphic organizers and keywords to support students with retrieval of information (Wehmeyer et al., 2003). Other curriculum augmentation strategies include self-directed learning strategies such as goal-setting and problem-solving (Lee et al., 2006).

Existing Curricula

Practitioners can develop the curriculum specially designed to address a particular intervention program philosophy and to meet the needs of the learners with ID in their program or adopt an existing available curriculum specially designed for teaching learners with ID. Several searches using a combination of ID related terms [e.g., cognitive disability(ies), developmental disability(ies), intellectual disability(ies), mental retardation, Down Syndrome] and curriculum or teaching as keywords were conducted in WorldCat, PsycINFO, and Web of Science. These searches were limited to publications in the past 20 years and English language. A total of 13 curricula for teaching learners with ID was found. The list of these curricula is presented below:

- Accessible Literacy Learning (ALL) Curriculum (Light & McNaughton, 2009)
- This curriculum is used to improve literacy skills in learners with autism, cerebral palsy, Down syndrome, and other disabilities. It includes sections on assessing skills, teaching component skills, teaching simple sentence and story reading, building comprehension, troubleshooting, writing skills, and six skill binders (e.g., sound blending, phoneme segmentation, letter-sound correspondence, single word decoding, sight word recognition, and shared reading).
- Health U: A Nutrition Curriculum for Teenagers with Intellectual and Developmental Disabilities (Bandini, Curtin, Fleming, Maslin, & Scampini, 2012)
- This curriculum is used to provide age-appropriate nutrition education for adolescents and young adults with mild to moderate intellectual disabilities. It includes ten lessons covering topics such as added sugar in food and drinks, added fats, making healthy choices, and healthy portions. Each lesion introduces new concepts and contains hands-on learning activities, movement/physical activities, and a taste test to encourage students expand their food repertoires.
- Conversation Skills on the Job and in the Community: A Curriculum for Adolescents and Adults with Developmental Disabilities (Banks, 2005)
- This curriculum teaches learners with developmental disabilities conversation skills on the job and in the community. It contains 50 plus self-managed lessons, such as learn the names of your coworkers, be the first to ask a question, and be the first to give a compliment.
- Conversation Skills II: Extending Conversations (Banks, 2008)
- This curriculum teaches learners with developmental disabilities to remember names, develop friendships at work and in the community, and extend conversations.
- Activities for Students with Special Needs: A Social Skills Curriculum (grades 3–5)(Bray, Morton, & Novotny, 2008)
- This curriculum is for students diagnosed with Autism, ADHD, intellectual disabilities, learning disabilities, behavioral difficulties, and emotional difficulties. It contains 11 units that are designed to help school-age children with special needs to learn and practice appropriate social skills.
- Feed All My Sheep: A Guide and Curriculum for Adults with Developmental Disabilities (Clark, 2000)
- This curriculum teaches adults and older youth with disabilities about the church and provides advice about how to set up church classes for these adults and how to integrate them into the church community.
- Creating a Meaningful Day: An Innovative Curriculum for Adults with Significant Intellectual Disabilities (Cofield-Van Dyke, 2008)

- This curriculum includes lesson plans for socializing, athletics, nature awareness, drama, visual arts, creative movement, sensory events, and storytelling. Each lesson plan has objectives, a material list, and areas of documentation.
- Improving Social Behaviors in the Classroom: An Easy Curriculum for Teachers of Young Children with Autism, Developmental Disabilities, and Typical Children (Freeman, 2011)
- This structured and structured curriculum contains day-by-day lessons for 25 weeks to be incorporated into the regular school day. It contains four categories: possessive understanding/acknowledgement of others, basic initiation skills, turn taking and simple social play, and cooperation.
- Effective Strategy-Based Curriculum for Abuse Prevention and Empowerment (ESCAPE) for Individuals with Developmental Disabilities
- (Hickson, Khemka, Golden, & Chatzistyli, 2015; Khemka & Hickson, 2002; Khemka & Hickson, 2008)
- This curriculum teaches individuals with intellectual and developmental disabilities to make effective, self-protective decisions in a wide range of sexual, physical, and verbal abuse situations.
- Attainment's Health Advocacy Program: An Activity-Based Curriculum for Adults with Developmental Disabilities (Holburn, Cea, & Gordon, 2008)
- This curriculum consists of 20 1-hour lessons covering topics, such as healthy eating, exercise, relationships, sexuality, stress, and medical issues like high blood pressure, diabetes, and depression.
- Teaching Math to People with Down Syndrome and Other Hands-On Learners (Horstmeier, 2008, 2016)
- This book provides strategies and activities/lesson plans relevant to daily living and covers topics, such as pre-number concepts, counting, recognition and writing of numbers, time and measurement, addition, subtraction, multiplication and division, fractions, money, and calculator use.
- Women be Healthy: A Curriculum for Women with Mental Retardation (Lunsky, Straiko, & Armstrong, 2003; Lunsky, Straiko, & Armstrong, 2002)
- This curriculum was designed for women with mental retardation and other developmental disabilities who had difficulty with gynecological and breast examinations or who had never had an exam. Its primary emphasis is on early detection of breast and cervical cancer. It contains three components: health education, anxiety reduction, and assertiveness and empowerment training.
- Health Matters: The Exercise and Nutrition Health Education Curriculum for People with Developmental Disabilities (Marks, Sisirak, & Heller, 2010)
- This curriculum is used to help adults with developmental disabilities to build healthy lifestyles. It contains 59 1-hour sessions that help people make the best choices about health, exercise, and nutrition.

Conclusion

Learners with ID need effective instruction to cultivate their strength and to help them develop skills for independent living. Developing or adopting a curriculum that can address intervention goals is the foundation of an effective intervention. This chapter presents information regarding how to develop a curriculum to meet the needs of individuals with ID, demonstrates sample lesson plans, and compiles a list of existing curricula published within the last 20 years. Practitioners may find the information presented in this chapter useful for their clinical work.

References

Agran, M., Cavin, M., Wehmeyer, M., & Palmer, S. (2006). Participation of students with moderate to severe disabilities in the general curriculum: The effects of the self-determined learning model of instruction. *Research and Practice for Persons with Severe Disabilities, 31*(3), 230–241. https://doi.org/10.1177/154079690603100303

Agran, M., Wehmeyer, M., Cavin, M., & Palmer, S. (2010). Promoting active engagement in the general education classroom and access to the general education curriculum for students with cognitive disabilities. *Education and Training in Autism and Developmental Disabilities, 45*(2), 163–174.

American Psychiatric Association. (2013). *Diagnostic and statistical manual of mental disorders* (5th ed.). Arlington, VA: American Psychiatric Publishing.

Anderson, S. R., Taras, M., & Cannon, B. O. (1996). Teaching new skills to young children with autism. In C. Maurice, G. Green, & S. C. Luce (Eds.), *Behavioral intervention for young children with autism* (pp. 181–194). Austin, TX: Pro-Ed.

Ault, M. J., Wolery, M., Gast, D. L., Doyle, P. M., & Eizenstat, V. (1988). Comparison of response prompting procedures in teaching numeral identification to autistic subjects. *Journal of Autism and Developmental Disorders, 18*(4), 627–636.

Bandini, L., Curtin, C., Fleming, R., Maslin, M., & Scampini, R. (2012). *Health U: A nutrition curriculum for teenagers with intellectual and developmental disabilities*. Worcester, MA: University of Massachusetts Medical School Eunice Kennedy Shriver Center.

Banks, M. (2005). *Conversation skills on the job and in the community: A curriculum for adolescents and adults with developmental disabilities*. Verona, Wisconsin: Attainment Company.

Banks, M. (2008). *Conversation skills II: Extending conversations*. Verona, Wisconsin: Attainment Company.

Bortoli, A. M., & Brown, P. M. (2008). The social attention skills of preschool children with an intellectual disability and children with a hearing loss. *Australasian Journal of Early Childhood, 33*(4), 25–33.

Bray, E., Morton, N., & Novotny, S. (2008). *Activities for students with special needs: A social skills curriculum (grades 3–5)*. Warminster, PA: Marco Products.

Celik, S., & Vuran, S. (2014). Comparison of direct instruction and simultaneous prompting procedure on teaching concepts to individuals with intellectual disability. *Education and Training in Autism and Developmental Disabilities, 49*(1), 127–144.

Chiang, H. M. (2008). Expressive communication of children with autism: The use of challenging behaviour. *Journal of Intellectual Disability Research, 52*, 966–972. https://doi.org/10.1111/j.1365-2788.2008.01042.x

Christensen, L. B., Burke Johnson, R., & Turner, L. A. (2011). *Research methods, design, and analysis*. Boston, MA: Allyn & Bacon.

Clark, D. C. (2000). *Feed all my sheep: A guide and curriculum for adults with developmental disabilities* (1st ed.). Louisville, Ky.: Geneva Press.

Cofield-Van Dyke, L. (2008). *Creating a meaningful day: An innovative curriculum for adults with significant intellectual disabilities*. Homewood, IL: High Tide Press.

Cosden, M., Koegel, L. K., Koegel, R. L., Greenwell, A., & Klein, E. (2006). Strength-based assessment for children with autism spectrum disorders. *Research and Practice for Persons with Severe Disabilities, 31*(2), 134–143.

de Bildt, A., Serra, M., Luteijn, E., Kraijer, D., Sytema, S., & Minderaa, R. (2005). Social skills in children with intellectual disabilities with and without autism. *Journal of Intellectual Disability Research, 49*, 317–328. https://doi.org/10.1111/j.1365-2788.2005.00655.x

Every Child Succeeds Act (ESSA) of 2015, Public Law No. 114-95, S.1177, 114th Cong. (2015). Retrieved from https://www.congress.gov/114/plaws/publ95/PLAW-114publ95.pdf

Fiscus, E. D., & Mandell, C. J. (1983). *Developing individualized education programs*. St Paul: West Publishing Company.

Frank, K. T., & Smith-Rex, S. J. (2015). *102 Practical strategies for reducing the deficit* (2nd ed.). Chapin, SC: YouthLight.

Freeman, S. (2011). *Improving social behaviors in the classroom: An easy curriculum for teachers of young children with autism, developmental disabilities, and typical children*. New York, NY: DRL Books, Inc.

Freeman, S. F. N., & Alkin, M. C. (2000). Academic and social attainments of children with mental retardation in general education and special education settings. *Remedial and Special Education, 21*(1), 2–18. https://doi.org/10.1177/074193250002100102

Gilson, C. B., Carter, E. W., & Biggs, E. E. (2017). Systematic review of instructional methods to teach employment skills to secondary students with intellectual and developmental disabilities. *Research and Practice for Persons with Severe Disabilities, 42*(2), 89–107. https://doi.org/10.1177/1540796917698831

Harrison, P. L., & Oakland, T. (2015). *Adaptive behavior assessment system* (3rd ed.). Torrance, CA: Western Psychological Services.

Hart, B. M., & Risley, T. R. (1982). *How to use incidental teaching for elaborating language*. Lawrence, Ks: H & H Enterprises.

Hickson, L., Blackman, L. S., & Reis, E. M. (1995). *Mental retardation: Foundations of educational programming*. Needham Heights: A Simon & Schuster Company.

Hickson, L., Khemka, I., Golden, H., & Chatzistyli, A. (2015). Randomized controlled trial to evaluate an abuse prevention curriculum for women and men with intellectual and developmental disabilities. *American Journal on Intellectual and Developmental Disabilities, 120*(6), 490–503. https://doi.org/10.1352/1944-7558-120.6.490

Holburn, S., Cea, C. D., & Gordon, A. (2008). *Attainment's health advocacy program: An activity-based curriculum for adults with developmental disabilities*. Verona, Wisconsin: Attainment Company.

Horner, R. D., & Keilitz, I. (1975). Training mentally-retarded adolescents to brush their teeth. *Journal of Applied Behavior Analysis, 8*(3), 301–309. https://doi.org/10.1901/jaba.1975.8-301

Horner, R. H., Albin, R. W., Todd, A. W., Newton, J. S., & Sprague, J. R. (2011). Designing and implementing individualized positive behavior support. In M. E. Snell & F. Brown (Eds.), *Instruction of students with* *severe disabilities* (7th ed., pp. 257–303). Upper Saddle River, NJ: Pearson.

Horstmeier, D. (2008). *Teaching math to people with Down Syndrome and other hands-on learners* (1st ed.). Bethesda, MD: Woodbine House.

Horstmeier, D. (2016). *Teaching math to people with Down syndrome, and other hands-on learners* (2nd ed.). Bethesda, MD: Woodbine House.

Hunt, P., McDonnell, J., & Crockett, M. A. (2012). Reconciling an ecological curricular framework focusing on quality of life outcomes with the development and instruction of standards-based academic goals. *Research and Practice for Persons with Severe Disabilities, 37*(3), 139–152. https://doi.org/10.2511/027494812804153471

Individuals with Disabilities Education Improvement Act of 2004, P.L. 108-446, 20 U.S.C. § 1400 et seq (2004).

Jimenez, B. A., Browder, D. M., Spooner, F., & Dibiase, W. (2012). Inclusive inquiry science using peer-mediated embedded instruction for students with moderate intellectual disability. *Exceptional Children, 78*(3), 301–317. https://doi.org/10.1177/001440291207800303

Johnson, H. T. (1968). *Foundations of curriculum*. Columbus: Ohio, C.E. Merrill Pub. Co.

Kemp, C., & Carter, M. (2002). The social skills and social status of mainstreamed students with intellectual disabilities. *Educational Psychology, 22*(4), 391–411.

Kerr, M. M., & Nelson, C. M. (1989). *Strategies for managing behavior problems in the classroom* (2nd ed.). Columbus: Merrill.

Khemka, I., & Hickson, L. (2002). *ESCAPE: An effective strategy-based curriculum for abuse prevention and empowerment*. New Tork, NY: Center for Opportunities and Outcomes for People with Disabilities Teachers College Columbia University.

Khemka, I., & Hickson, L. (2008). *An effective strategy-based curriculum for abuse prevention and empowerment for adults with developmental disabilities*. New York, NY: Center for Opportunities and Outcomes for People with Disabilities, Teachers College Columbia University.

Kleinert, H., Towles-Reeves, E., Quenemoen, R., Thurlow, M., Fluegge, L., Weseman, L., & Kerbel, A. (2015). Where students with the most significant cognitive disabilities are taught: Implications for general curriculum access. *Exceptional Children, 81*(3), 312–328. https://doi.org/10.1177/0014402914563697

Koegel, L. K., Koegel, R. L., Harrower, J. K., & Carter, C. M. (1999). Pivotal response intervention I: Overview of approach. *Journal of the Association for Persons with Severe Handicaps, 24*(3), 174–185. https://doi.org/10.2511/rpsd.24.3.174

Koegel, L. K., Singh, A. K., & Koegel, R. L. (2010). Improving motivation for academics in children with autism. *Journal of Autism and Developmental Disorders, 40*(9), 1057–1066. https://doi.org/10.1007/s10803-010-0962-6

Koegel, R. L., Carter, C. M., & Koegel, L. K. (1998). Setting event to improve parent-teacher coordination and motivation for children with autism. In J. K. Luiselli & M. J. Cameron (Eds.), *Antecedent control: Innovative approaches to behavioral support* (pp. 167–186). Baltimore: Paul H. Brookes.

Koegel, R. L., & Koegel, L. K. (2012). The PRT pocket guide: Pivotal response treatment for autism spectrum disorders. In *Baltimore*. Maryland: Paul H. Brookes Publishing Co.

Leaf, J. B., Leaf, J. A., Alcalay, A., Kassardjian, A., Tsuji, K., Dale, S., … Leaf, R. (2016). Comparison of most-to-least prompting to flexible prompt fading for children with autism spectrum disorder. *Exceptionality, 24*(2), 109–122. https://doi.org/10.1080/09362835.2015.1064419

Leaf, J. B., Sheldon, J. B., & Sherman, J. A. (2010). Comparison of simultaneous prompting and no-no prompting in two-choice discrimination learning with children with autism. *Journal of Applied Behavior Analysis, 43*(2), 215–228. https://doi.org/10.1901/jaba.2010.43-215

Lee, S.-H., Amos, B. A., Gragoudas, S., Lee, Y., Shogren, K. A., Theoharis, R., & Wehmeyer, M. L. (2006). Curriculum augmentation and adaptation strategies to promote access to the general curriculum for students with intellectual and developmental disabilities. *Education and Training in Developmental Disabilities, 41*(3), 199–212.

Light, J. C., & McNaughton, D. (2009). *ALL (Accessible Literacy Learning): Evidence-based reading instruction for learners with Autism, Cerebral Palsy, Down Syndrome and other disabilities*. Solana Beach, CA: Mayer-Johnson.

Lipscomb, S., Haimson, J., Liu, A. Y., Burghardt, J., Johnson, D. R., & Thurlow, M. L. (2017). *Preparing for life after high school: The characteristics and experiences of youth in special education. Findings from the National Longitudinal Transition Study 2012. Volume 2: Comparisons across disability groups: Full report (NCEE 2017-4018)*. Retrieved from Washington, DC:

Lunsky, Y., Straiko, A., & Armstrong, S. (2003). Women be healthy: Evaluation of a women's health curriculum for women with intellectual disabilities. *Journal of Applied Research in Intellectual Disabilities, 16*(4), 247–253. https://doi.org/10.1046/j.1468-3148.2003.00160.x

Lunsky, Y., Straiko, A., & Armstrong, S. C. (2002). *Women be healthy: A curriculum for women with mental retardation and other developmental disabilities*. Chapel Hill, NC: NC North Carolina office on disability and health.

Marks, B., Sisirak, J., & Heller, T. (2010). Health matters: The exercise and nutrition health education curriculum for people with developmental disabilities. In *Baltimore*. Maryland: Paul H. Brookes Publishing Co.

McClintock, K., Hall, S., & Oliver, C. (2003). Risk markers associated with challenging behaviours in people with intellectual disabilities: Ameta-analytic study. *Journal of Intellectual Disability Research, 47*, 405–416. https://doi.org/10.1046/j.1365-2788.2003.00517.x

Meyer, A., Rose, D. H., & Gordon, D. T. (2014). *Universal design for learning: Theory and practice*. Wakefield, MA: CAST Professional Publishing.

Murzynski, N. T., & Bourret, J. C. (2007). Combining video modeling and least-to-most prompting for establishing response chains. *Behavioral Interventions, 22*(2), 147–152. https://doi.org/10.1002/bin.224

Oliva, P. F. (2005). *Developing the curriculum* (6th ed.). Boston: Allyn and Bacon.

Oreilly, M. F., & Chadseyrusch, J. (1992). Teaching a social skills problem-solving approach to workers with mental-retardation: An analysis of generalization. *Education and Training in Mental Retardation and Developmental Disabilities, 27*(4), 324–334.

Public Law 108-446 (2004). Individuals with Disabilities Education Act Amendments of 2004,PL 108–446, *et seq*.

Rusch, F. R., Rose, T., & Greenwood, C. R. (1988). *Instruction to behavior analysis in special education*. Englewood Cliffs, NJ: Prentice-Hall.

Ryndak, D. L., Moore, M. A., Orlando, A. M., & Delano, M. (2008). Access to the general curriculum: The mandate and role of context in research-based practice for students with extensive support needs. *Research and Practice for Persons with Severe Disabilities, 33*(34), 199–213. https://doi.org/10.2511/rpsd.33.4.199

Sabornie, E. J., Cullinan, D., Osborne, S. S., & Brock, L. B. (2005). Intellectual, academic, and behavioral functioning of students with high-incidence disabilities: A cross-categorical meta-analysis. *Exceptional Children, 72*(1), 47–63.

Sailor, W., & Guess, D. (1983). *Severely handicapped students: An instructional design*. Boston: Houghton Miffin.

Selmi, A. M., Gallagher, R. J., & Mora-Flores, E. R. (2015). *Early childhood curriculum for all learners: Integrating play and literacy activities*. Thousand Oaks, CA: SAGE Publications.

Shepherd, G. D. R. W. B. (1992). *Modern elementary curriculum* (7th ed.). Fort Worth: Harcourt Brace Jovanovich.

Snell, M. E., & Brown, F. (2011). Selecting teaching strategies and arranging educational environments. In M. E. Snell & F. Brown (Eds.), *Instruction of students with severe disabilities* (7th ed., pp. 122–185). Upper Saddle River, NJ: Pearson.

Snell, M. E., & Gast, D. (1988). Applying the time delay procedure to the instruction of the severely handicap. *Education and Treatment of Children, 11*, 29–44.

Soukup, J. H., Wehmeyer, M. L., Bashinski, S. M., & Bovaird, J. A. (2007). Classroom variables and access to the general curriculum for students with disabilities. *Exceptional Children, 74*(1), 101–120. https://doi.org/10.1177/001440290707400106

Sparrow, S. S., Saulnier, C. A., & Cicchetti, D. V. (2016). *Vineland-3: Vineland adaptive behavior scales* (3rd ed.). Minneapolis, MN: Pearson Assessments.

Stokes, T. F., & Osnes, P. G. (1986). Programming the generalization of children's social behavior). In P. S. Strain, M. Guralnick, & H. Walker (Eds.), *Children's social behavior: Development, assessment and modification*. Orlando, FL: Academic Press.

Strain, P. S., Kohler, F. W., Storey, K., & Danko, C. D. (1994). Teaching preschoolers with autism to self-monitor their social interactions: An analysis of results in-home and school settings. *Journal of Emotional and Behavioral Disorders, 2*(2), 78–88. https://doi.org/10.1177/106342669400200202

Trela, K., & Jimenez, B. A. (2013). From Different to Differentiated: Using "Ecological Framework" to Support Personally Relevant Access to General Curriculum for Students with Significant Intellectual Disabilities. Research and Practice for Persons with Severe Disabilities, 38(2), 117–119. https://doi.org/10.2511/027494813807714537

U.S. Department of Education. (2017). *39th Annual Report to Congress on the Implementation of the Individuals with Disabilities Education Act, 2017*. Retrieved from Washington, DC

Wehmeyer, M. L., Lattin, D. L., Lapp-Rincker, G., & Agran, M. (2003). Access to the general curriculum of middle school students with mental retardation: An observational study. *Remedial and Special Education, 24*(5), 262–272. https://doi.org/10.1177/07419325030240050201

Wehmeyer, M. L., Palmer, S. B., Agran, M., Mithaug, D. E., & Martin, J. E. (2000). Promoting causal agency: The self-determined learning model of instruction. *Exceptional Children, 66*(4), 439–453. https://doi.org/10.1177/001440290006600401

Westling, D. L., & Fox, L. (2009). *Teaching students with severe disabilities*. Upper Saddle River, NJ: Pearson Education.

Wolery, M., Gast, D. L., Kirk, K., & Schuster, J. (1988). Fading extra-stimulus prompts with autistic children using time delay. *Education and Treatment of Children, 11*(1), 29–44.

Wong, C., Odom, S. L., Hume, K., Cox, A. W., Fettig, A., Kucharczyk, S., & Schultz, T. R. (2013). *Evidence-based practices for children, youth, and young adults with autism spectrum disorder*. Chapel Hill: The University of North Carolina, Frank Porter Graham Child Development Institute.

Yu, J., Newman, L., & Wagner, M. (2009). *Secondary school experiences and academic performance of students with mental retardation*. Menlo Park, CA: SRI International.

43 Emerging Technology for Students with Intellectual Disability

Luis J. Mena, Vanessa G. Felix, Rodolfo Ostos, and Gladys E. Maestre

Introduction

Development of technological innovations is an engine of integration with the capacity to mitigate the obstacles that continue to hinder full insertion of individuals with intellectual disabilities in all areas of our social context (Brown & Duguid, 2017). The use of new technologies has emerged as a transformative tool to mediate and enhance the inclusion of people with disabilities, becoming an effective alternative to break the physical and cognitive barriers that limit the learning and communication of this sector of the population (Carver, Ganus, Ivey, Plummer, & Eubank, 2016; Leopold, Lourie, Petras, & Elias, 2015; Lancioni & Singh, 2014). Thus, in the last decade, the use of new supports and digital applications in the school environment has significantly increased, acting as effective mediators in the teaching and learning process (Altınay-Gazi & Altınay-Aksal, 2017; Díaz, 2017; Kirkwood & Price, 2014; Teo & Zhou, 2017).

Emerging technology-aided learning interventions can be used creatively to improve the development of abilities of people with academic skill deficiencies in math, reading, and/or writing. Under an interactionist conception that shifts its focus from the individual characteristics of students to a curricular support model, these types of interventions are expanding (Crossley & Mcnamara, 2016; Felix, Mena, Ostos, & Maestre, 2017; Kim, Park, & Coleman, 2017; Weng, Maeda, & Bouck, 2014). Through the interaction of software, peripherals, and an adequate methodology, the facilitator of learning – professor, companion, and family member – helps everyone in the classroom to develop their potential. This interaction is carried out in the zone of proximal development and converts what many call the problem of diversity – from the perspective of the deficit-based model – into an opportunity to broaden and enrich the educational context. The goal is to provide a suitable response to the didactic needs of students with intellectual disabilities and help them to be included in the physical and digital environments with creativity and full equality of opportunities (Cortiella & Horowitz, 2014; Felix et al., 2017; Shalev, Asmus, Carter, & Moss, 2016).

Although the role of technology as a mediator of learning is widely accepted, new forms of

---

L. J. Mena (✉) · V. G. Felix · R. Ostos
Department of Computer Sciences, Universidad Politecnica de Sinaloa, Mazatlan, Mexico
e-mail: lmena@upsin.edu.mx; vfelix@upsin.edu.mx; rostos@upsin.edu.mx

G. E. Maestre
Department of Neurosciences and Human Genetics, University of Texas Rio Grande Valley School of Medicine, Brownsville, TX, USA
e-mail: gladys.maestre@utrgv.edu

J. L. Matson (ed.), *Handbook of Intellectual Disabilities*, Autism and Child Psychopathology Series, https://doi.org/10.1007/978-3-030-20843-1_43

social exclusion have emerged from different fronts and for various reasons. The absence of specific policies on digital inclusion, the difficulties in accessing technological infrastructures, insufficient training in and for the use of emerging technologies, the lack of references and supports, or the scarce application and promotion of design standards and guidelines for all people are some of the causes of what is rightly called *digital exclusion* (Chadwick & Wesson, 2016; Ditchman, Kosyluk, Lee, & Jones, 2016; Newman, Browne-Yung, Raghavendra, Wood, & Grace, 2017).

To guarantee access to technological resources to all people, including those who suffer some type of disability, it is not enough to develop emerging assistive technologies adjusted for each type of disability. It is also necessary that the design of technological resources considers standards and recommendations that facilitate the adoption of new technologies (Ayres, Mechling, & Sansosti, 2013; Carey, Friedman, & Bryen, 2005; Standen, Brown, & Cromby, 2001). Consequently, the design of technological aids or adaptations should be oriented to the development of simpler materials and high accessibility in their contents. Therefore, the desirable characteristics include using clear language, not overloading the user interface with information, using graphic icons or enough description to help in navigation, and using alternative supports of comprehension through auditory resources, graphics, and/or text, among others (Dekelver et al., 2015; Davies et al., 2015; Stock, Davies, Wehmeyer, & Palmer, 2008).

The aspiration toward greater accessibility has led to the idea of a *design for all* or *universal design* (Alnahdi, 2014; Hall, Cohen, Vue, & Ganley, 2015; Hartman, 2015). *Universal design* implies that learning is flexible and supportive for all learners, including those with intellectual disabilities, so that instructional goals, assessments, methods, and materials are usable and accessible by all (Hall, Meyer, & Rose, 2012; Rose, 2000). This framework aims to take into account in the design phase the accessibility requirements derived from the different types and degrees of people's functional capacity. Hence, failure to meet this objective in design processes could lead to discrimination, exclusion, and problems of social participation for groups with learning disabilities.

This chapter provides an overview of some technological developments that claim to assist students with disabilities, ensuring that users can access them regardless of their cognitive characteristics and also can use them efficiently to meet minimum criteria of usability and accessibility. Devices such as microswitch-based program technology to enhance adaptive behaviors, computer-assisted learning to improve cognitive and academic performances, and potential future developments, such as educational robotics, will be reviewed and discussed.

Microswitch-Based Program Technology

Persons with severe to profound intellectual and developmental disabilities may exhibit challenging behaviors and few adaptive responses that seriously hamper their acceptance and social desirability in daily context (Cannella, O'Reilly, & Lancioni, 2006; Lancioni et al., 2009; Spittle & Orton, 2014). Therefore, when individuals with serious limitations in adaptive response also exhibit problematic behaviors, technological interventions should try to increase constructive responding and reduce negative behaviors (Lancioni et al., 2005, 2008, 2009).

To overcome both issues, emerging assistive technology includes the implementation of microswitch cluster approaches (Lancioni et al., 2009). Basically, microswitches are electronic tools or devices designed to assist persons with severe and multiple developmental disabilities to independently access the preferred stimuli through the exhibition of small or minimal motor responses (Holburn, Nguyen, & Vietze, 2004; Lancioni, Sigafoos, O'Reilly, & Singh, 2012; Mechling, 2006; Saunders et al., 2003). The latest microswitch cluster interventions simultaneously monitor adaptive responding and atypical behaviors and deliver preferred stimuli automatically on positive responses performed without the presence of aberrant behaviors (Lancioni et al., 2006, 2009;

Stasolla, Damiani, & Caffo, 2014). Additionally, positive stimulation delivered for a constructive response would be interrupted when challenging behavior is demonstrated.

For example, Lancioni et al. (2013) assessed the use of microswitch clusters strategy to promote adaptive responding and decrease inappropriate posture consisting of the head and trunk leaning forward in two participants: a 10-year-old boy with a congenital encephalopathy diagnosis and reportedly profound intellectual disability and a 64-year-old woman presenting a neurodegenerative condition with decline in all aspects of life. Optic, tilt, and vibration microswitches were used to record their adaptive responses, while optic and tilt microswitches monitored their posture. Initially, the participants accessed brief periods of preferred stimulation for each cleaning response regardless of whether their posture was correct or not. Subsequently, the adaptive responses led to positive stimulation only if the inappropriate posture was absent. Moreover, in the case of appearing inadequate posture, the stimulation was interrupted. Results showed that both participants had large increases in adaptive responding and a drastic reduction in inappropriate posture during the first and second intervention phases and improved their problematic posture at the 2-week post-intervention check.

Microswitch technology can be implemented as educational intervention in school settings to improve the academic skills of students with multiple disabilities (Lancioni et al., 2004, 2006, 2008, 2010; Lancioni, Singh, Oliva, Scalini, & Groeneweg, 2003; Stasolla et al., 2015; Wacker, Wiggins, Fowler, & Berg, 1988). Individuals with extensive intellectual and communication disabilities can experience a reduced level of participation and/or active role within school environment, affecting their academic progress and, consequently, deteriorating their quality of life (Lancioni et al., 2006; Schalock et al., 2002). Therefore, a suitable/effective microswitch intervention strategy may foster their motivation and self-determination by learning and acquiring adaptive responses, thereby reducing passivity and preventing isolation (Lancioni et al., 2006; Stasolla et al., 2015).

Stasolla et al. (2015) evaluated six participants (mean age 10.7 years) with intellectual and extensive motor disabilities on a combined computer and microswitch-based program aimed at promoting academic activities in a school setting. The study provided educational activities and positive stimulation as primary reinforcements. By activating pressure and keyboard emulator microswitches (Fig. 43.1), participants were able to request and choose geography, literature,

Fig. 43.1 Keyboard emulator and pressure microswitches from Stasolla et al. (2015)

mathematics, and literacy activities through a computer interface. Each academic option was linked to specific stimuli, and the student could choose which response to perform and which stimuli to pursue. A final response was necessary in order to confirm a definitive choice. Results indicated that all students significantly improved their academic skills, positive participation, and indices of happiness during intervention phases compared to baselines. At the 3-month post-intervention check, a maintenance/generalization phase indicated that all participants consolidated the performances at their homes involving their parents. Finally, effectiveness was corroborated by a social validation assessment including support teachers as raters, who considered that the combined intervention was more favorable than those singularly learned.

One the other hand, Lancioni et al. (2010) assessed the effectiveness and acceptability of a microswitch-based program technology to support the literacy performance of a 12-year-old girl with extensive neuromotor disabilities to write words. Educational intervention included using a touch/pressure panel and voice-detecting microswitches connected to a computer with a scanning keyboard emulator. Panel activation enabled her to light up the keyboard and select/write letters and the vocalization to perform the scanning of the letters until reaching the one needed. Results indicated that the use of the microswitch strategy allowed the participant to successfully manage both scanning and letter selection/writing in an independent manner. It was a less tiring performance with respect to the traditional solution of expanded keyboard connected to a computer screen. Furthermore, effectiveness of the microswitch intervention was corroborated by a social validation assessment involving university psychology students as raters who manifested a significant personal support for the use of microswitch technology instead of the pre-existing solutions.

Earlier studies were consistently positive and support the use of microswitch technology in educational programs for individuals with severe to profound intellectual and developmental disabilities to provide constructive engagement, self-determination, and independence (Algozzine, Browder, Karvonen, Test, & Wood, 2001; Lachapelle et al., 2005; McDougall, Evans, & Baldwin, 2010; Stasolla et al., 2014). Thus, microswitch-based interventions may be of relevant and even of crucial importance to facilitate the acquisition of an active role and to develop some forms of constructive/functional response in an independent manner to increase academic performance, to improve general mood, and to enhance social image, desirability, and acceptability by others (Lancioni & Singh, 2014; Lancioni et al., 2012; Petry, Maes, & Vlaskamp, 2005). Within school settings, microswitch intervention strategies can be offered to students with learning difficulties to support a wide range of academic activities, pursuing the dual objective of improving integration and inclusion and reducing stereotypic and/or challenging behavior, with beneficial effects for the entire classroom (Lancioni & Singh, 2014). More extensive forms of microswitch sessions involving home context could promote positive interactions between teachers and parents and consolidate generalization and maintenance of academic achievements of students (Lancioni et al., 2006; McLay et al., 2015; Stasolla et al., 2015).

Although microswitch-based approaches represent a constructive and effective way to help people with intellectual and developmental disabilities to improve their academic performance, their implementation in educational settings is insufficient (Stasolla et al., 2015). Therefore, a research effort must be aimed to understand the barriers and facilitators of implementation of microswitch technology within learning environments. Design of new microswitch programs should deal with the following issues: (1) envisaging the interaction with other emerging technologies as mobile computing, pattern recognition, and virtual reality, among others, (2) performing a post-intervention phase involving home context to maintain learned abilities and generalize academic skills, and (3) enlarging social validation assessment with teachers and parents as raters to endorse the intervention program with greater validity and reliability (Lancioni et al., 2006; Stasolla et al., 2014).

Computer-Assisted Learning

Individuals with intellectual disability have different capacities and skills for understanding and learning (Daunhauer, Fidler, & Will, 2014). Thus, their learning potential depends on their educational environment and on the encouragement and support received in learning situations. Accordingly, they need more reinforcement and stimulation to function independently within school settings (Carlson, 2009). Therefore, it is necessary to design additional strategies for students with intellectual disabilities to improve their independence and self-determination, enabling them to carry out successfully education tasks, facilitating academic progress with beneficial effects on school staff and peers (Barnard-Brak, Thompson, Wei, & Richman, 2014).

Specific skills or abilities are learned through exposure to various situations, experiences, and learning opportunities. Some suggest that for individuals with intellectual disabilities to learn new skills, activities need to be broken down into smaller steps, and more repetition and structure are required for retention (Hulme & Mackenzie, 2014). Every person with cognitive disabilities demonstrates individual abilities, strengths, and weaknesses and has their own learning profile. These personal patterns of learning can be used to develop teaching strategies and therapies that are particularly effective for each individual (Felix et al., 2017). Given this set of educational characteristics, the use of computer-assisted interventions emerges as a suitable method for teaching students with intellectual disability (Clarfield & Stoner, 2005; Fletcher-Watson, 2014; Kulik, 2003; Tanimoto, Thompson, Berninger, Nagy, & Abbott, 2015).

Computer technologies have been found to be beneficial in the instruction of persons with intellectual disabilities for several reasons. People with learning difficulties often show attention problems because they have difficulty screening out unnecessary sensory information. Viewing of instructions through electronic media may maximize attention by eliminating or reducing information that might distract from the main task, allowing individuals to work at their own pace (Sansosti, Doolan, Remaklus, Krupko, & Sansosti, 2015; Williams, Wright, Callaghan, & Coughlan, 2002). Computer-assisted teaching can counteract difficulties in motivation and fatigue that often occur in individuals with learning disabilities. Creative and playful activities using computers can increase their confidence and motivation (Bosseler & Massaro, 2003; Goldsmith & LeBlanc, 2004; Ortega & Gómez, 2006). The use of computers promotes individualized teaching, allowing adaption of the work to the skill level of each student. Additionally, a computer system can offer an extensive choice of rewards and stimuli, which can be personalized to the student's needs and interests (Constantin, Johnson, Smith, Lengyel, & Brosnan, 2017).

Computer-assisted instruction enables repetition, errorless learning, self-correction, and immediate feedback about response accuracy. Academic software can be designed to provide support at each step, allowing students to learn a sequence of steps to achieve success. A computer can be a patient and repetitive teacher, providing students with multiple opportunities for learning and assessment, creating an environment that appeals to individuals with intellectual disabilities as less threatening, and ultimately decreasing anxiety and expectation of failure (Felix et al., 2017; Sansosti, Powell-Smith, & Cowan, 2010). Computers offer students the possibility of increasing their autonomy and personal independence, to be able to learn alone, and they need less help from others. They can accomplish tasks with less effort, greater speed, and higher quality, reducing work avoidance, frustration, and inappropriate behavior (Meyers, 1988). The computer can also store achievement data for each student, providing an objective record of their progress. Finally, computers can act as an interface between the individual with cognitive disability and other people, ameliorating social communication and interaction deficits and reducing social anxiety (Constantin et al., 2017).

Research evidence has identified that individuals with learning disabilities not only demonstrate significant skill acquisition when taught via computers but also have a preference for instruction delivered through such devices over standard print materials (Ortega & Gómez, 2006; Shaw & Lewis, 2005; Walcott, Marett, & Hessel, 2014). Nevertheless, some research has suggested that there may be differential effects for the effectiveness of the computer-assisted learning based upon the academic skill targeted and the age of the population (Kulik, 2003; Murphy et al., 2002). Moreover, several studies evaluated the benefits of computer-assisted learning approaches for students within special education without comparing participants' outcomes with those of a control group (Butcher & Jameson, 2016). Additionally, many studies showed that students with learning difficulties spent more time on task and completed more problems during the computerized intervention but demonstrated similar levels of academic performance using paper-and-pencil methods (Lewandowski, Wood, & Miller, 2016). This could be explained by the studies that show the main difference between teaching approaches was simply the presentation of academic instructions (computer vs. printed text), rather than offering, for example, a game format based on multimedia instructional design utilizing touch-screen technology (Felix et al., 2017).

To effectively assess the positive impact of computer learning intervention in students with intellectual disabilities, it is necessary to examine academic benefits from well-designed studies based on group experimental research within natural classroom settings (Butcher & Jameson, 2016; Dunst & Hamby, 2012; Gersten et al., 2005; Sansosti et al., 2015). It is also important to evaluate academic achievement and educational outcomes in specific scholar domains. Selection of digital approaches based on emerging computer technologies seems to be justified in supporting students' learning and cognitive processes and outcomes within special education settings (Fernández-López et al. 2013; Liu, Wu, & Chen, 2013; Shah, 2011).

In this context, Felix et al. (2017) conducted a pilot study on an experimental and control group to assess the effectiveness of a computer-assisted learning tool for children with Down syndrome. Complementary learning strategies included mobile computing, multimedia instructional design principles (Mayer, 2005; Sorden, 2005), and automated handwriting and speech recognition using an artificial neural network (Graves, Mohamed, & Hinton, 2013; Jiao, Liu, Yuan, & Bai, 2012) aimed at improving literacy skills of participants, through speech and drawing activities within a playful environment. Software design was assisted by special education therapists, and graphical use interface included animated characters with features of people with Down syndrome (Fig. 43.2). The treatment group

Fig. 43.2 Graphical user interface of computer learning approach from Felix et al. (2017)

was taught using a digital approach on tablet computers, and the control group received individual instruction on the same material but for paper-and-pencil tasks. The differences between adjusted posttest scores for both groups were statistically significant in favor of the intervention group on single-word reading ($p = 0.048$) and handwriting form ($p = 0.046$) measures, with large effect sizes ($d > 0.80$).

Felix et al. (2017) concluded that effectiveness of the computer instructional approach was possibly attributable to its ability to present very tightly controlled stimuli in an entertaining game-like format, providing enough repetitions for learning and helping to avoid frustration and stress; integration of speech and touchscreen input to perform personalized task sequences that address attention deficit and working memory limitations; adjustable neural network recognition thresholds that helped to make adaptations in progress to facilitate teaching according to learning profile of each student; and the presentation of animated objects that aided overcome deficits in abstract thinking.

Instead, Bakker, Heuvel-Panhuizen, and Robitzsch (2016) examined the effects of a teacher-delivered intervention with online mathematics mini-games on special education students' multiplicative reasoning ability. A pretest/posttest control group design was employed with three experimental and two control schools of special primary education. The only difference between the two groups was that in the experimental group part of the academic program for multiplicative reasoning was replaced by the online digital intervention. Computer mini-games were adapted versions from the mathematics games website RekenWeb (Jonker, Wijers, & Van Galen, 2009). In accordance with the needs of special education students, adaptations were easy to learn, contained simple graphics and minimal sound effects, and provided rewards, and the mathematics content was integrated into the main activity of the games. A scoring mechanism that increased scores as students finished the game successfully was also added. Furthermore, many of the games allowed for different difficulty levels. Results indicated that students in the experimental condition had higher learning outcomes than control students on declarative knowledge of multiplication facts ($d = 0.39$, $p = 0.047$).

Bakker et al. (2016) concluded that positive effects of computer intervention may be because mathematical knowledge was more easily transferred from the mini-games since these included less distracting features and required only a little reading. Additionally, it appears important for special education students that games provide continuous rewards and that they allow for different difficulty levels – for example, by providing choices in the types of problems to be solved or making available supportive features. Therefore, games delivered an engaging environment in which students were motivated to practice basic multiplicative number facts. Finally, they argued that findings were in agreement with Kroesbergen and Van Luit's (2003) conclusion that for students with intellectual disabilities, basic skill interventions are more effective than interventions targeting more complex mathematics tasks.

Despite the potential of computer emerging technologies for helping students with intellectual disabilities to improve their academic performance and motivation, consistent evidence to support their implementation in the classroom and evaluating their efficacy is lacking (Fletcher-Watson, 2014; Lewandowski et al., 2016). A possible explanation is that computer innovations are evolving much faster than the research needed to demonstrate their effectiveness as a learning strategy in special education. In this sense, the rapid pace of technology has delayed the separation of the leading features of new technologies that are most significant in supporting students with learning disabilities to improve their academic achievement. Consequently, it is possible that many of the new technological developments actually help non-disability students more than students with intellectual disabilities, creating a digital divide with respect to their educational outcomes (Lussier-Desrochers et al., 2017; Lewandowski et al., 2016).

Beyond the positive claims suggesting that computer-assisted learning effectively supports academic skill acquisition of students with intellectual disabilities, the field of new technologies for special education has yet to accumulate evidence-based practices (Gersten & Edyburn, 2007). Because several studies found that very few computer-aided instruction approaches met the standards of quality required for evidence-based interventions (Anderson, Anderson, & Cherup, 2009; Edyburn, 2013; Fitzgerald, Koury, & Mitchem, 2008; Weng et al., 2014), it is difficult to determine which features of computer technology resulted in specific academic gains for students in special education. Therefore, it is important to build a robust research base demonstrating generalizable efficacy of emerging computer-based learning interventions for students with intellectual disability, meeting quality criteria needed to derive evidence-based learning strategy, developing collaborative approaches between special education staff and software developers, and examining the positive impact of digital technology use for both non-disability students and students with learning disabilities within school settings.

Educational Robotics

Emerging technology advances and research developments in the area of robotics have allowed robots to become an integral component of our society. In the case of applications for education, students with disability have received particular attention due to evidence from several studies and more reasonable new robotic platforms (Conti, Di Nuovo, Buono, & Di Nuovo, 2017; Hedgecock, Standen, Beer, Brown, & Stewart, 2014; Pennington, Welch, & Scott, 2014).

In general, educational robotics is the set of pedagogical activities that support and strengthen specific areas of knowledge and develop competencies in the student through the conception, creation, assembly, and implementation of robotic prototypes and specialized programs for pedagogical purposes (Kazakoff, Sullivan, & Bers, 2013). Within special education, it has shown the potential to be implemented as a learning strategy to reinforce social, emotional, and academic skills in students with concentration problems and learning disabilities (Den Brok & Sterkenburg, 2015).

Students with intellectual disabilities require a high degree of repetition and face-to-face time with an instructor, as well as consistent, timely, and precise feedback (Cacioppo & Petty, 1979; Cook, Meng, Gu, & Howery, 2002; Park & Kwon, 2016; Reardon, Zhang, Wright, & Parker, 2015). Robots can tirelessly provide consistent and continuous academic interactions with students and avoid mistakes, which are an advantage over human teachers who may experience fatigue and common teaching errors (Park & Kwon, 2016; Reardon et al., 2015).

Learning and retention for academic subjects such as mathematics or science can be improved when students actively participate in direct purposeful practices, as opposed to watching demonstrations, because these emphasize the integration of several hands-on activities while communicating educational concepts (Adams, Alvarez, & Rios-Rincon, 2017; Petress, 2008). Students with intellectual and developmental disabilities may miss the hands-on component of learning due to their limitations in handling with precision objects used in hands-on lessons. Therefore, developing robots used for augmentative manipulation in education activities can contribute to the hands-on learning of students with multiple disabilities, acting as a practical learning partner to interact with the same objects as their peers in the classroom, and increase the active component of learning experience in the real world (Adams et al., 2017; Chang, Lee, Wang, & Chen, 2010; Cook, Bentz, Harbottle, Lynch, & Miller, 2005).

However, the most significant advantage of learning strategy from robots is the tendency of positive acceptance and motivation for students with intellectual disability (Den Brok & Sterkenburg, 2015; Park & Kwon, 2016; Standen et al., 2014). People with learning disabilities can have difficulties with understanding and showing interest in the outside world. Robots seem to operate as a key tool able to draw the attention of students with learning difficulties. They appear to encourage a process of interaction, by increasing joint attention, communication, and autonomous

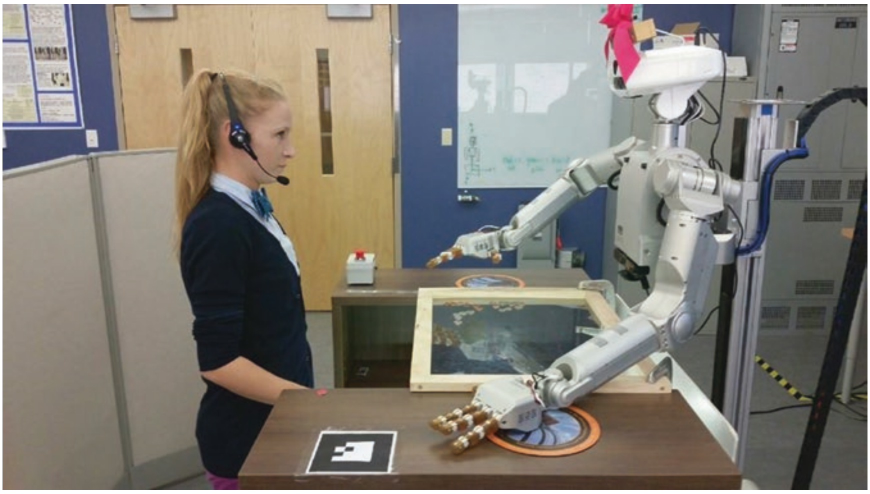


Fig. 43.3 Interaction setting for intelligent robot instruction from Reardon et al. (2015)

social behavior, moreover of supporting the work of teachers and parents (Costa, Soares, Santos, Pereira, & Moreira, 2016; Den Brok & Sterkenburg, 2015; Fridin, 2014; Lehmann, Iacono, Dautenhahn, Marti, & Robins, 2014). In addition, research evidence indicates that robots can stimulate the social development of children with multiple disabilities because affective touching, imitation, and expressing emotions toward the robot were encouraged (Kozima, Nakagawa, & Yasuda, 2007; Pioggia et al., 2008; Robins, Dautenhahn, Te Boekhorst, & Billard, 2005).

Cho and Shin (2011) demonstrated that cognitive-disabled children tend to accept social robots more enthusiastically than other kinds of toys, agents, or even humans. Studies found that using robots as a tool to perform explorative learning activities is more motivating than microswitch interventions or computer learning programs (Cook et al., 2005; Fridin, 2014; Kim et al., 2013). Therefore, this tendency toward positive acceptance of robots by learners could be helpful when dealing with different learning needs.

As an example, Reardon et al. (2015) proposed the creation of an autonomous, intelligent robot instructor to teach social and life skills based on mathematical competencies to students with intellectual disabilities. The primary robotic hardware was a mobile humanoid robot equipped with a couple of elastic arms with hands, a sensor head with 2 degree of freedom movement, high-definition cameras, Bluetooth microphone, and stereo speakers (Fig. 43.3). Two computer systems provided real-time functionality of the motion, vision, and audio components. The selected instructional task was the ability to calculate the correct quantities and denominations of currency that should be exchanged after a cash transaction. Participants in this study were three college-age students attending a postsecondary education program for young adults with intellectual and developmental disabilities.

During baseline, each participant performed at least three trials related to counting amounts of change back to researchers for purchases of items under US$1. No feedback, prompting, or assistance was provided. Under the baseline condition, none of the students were able to correctly perform the skills related to making change independently. The general outline of experimental trial began with the robot delivering the task instruction and then asking the student to show the correct change for 1 dollar from a randomly selected price less than US$ 1. Robotics approach observed the performance of the requested task through a real-time object tracking system and provided feedback by means of speech and gestures by using response prompting methodology as part of a cognitive process (Wolery, Ault, & Doyle, 1992).

The robotics system interpreted the answer of the student and provided precise instruction, reevaluating and presenting an updated prompt, if necessary. In the case of a correct response, the robot delivered a positive and differential reinforcement tailored to the level of intrusiveness of the prompt that was required. After reinforcement, the student was asked if he or she would like to continue with another lesson. By repetitions of this process, the goal was for the student to require less and less prompting, to the point of performing the task independently.

Experimental results showed that all students were able to achieve accurate performance to identify the correct amount of change for a given price and to provide the correct amount of change using the least amount of coins possible. An acceptability study was conducted to examine the opinions of students under the robot's tutelage. Post-instruction survey results indicated that participants felt a beneficial interaction and showed greater willingness to work with robots again. Authors concluded that intelligent autonomous robot instructors have the potential to teach a non-trivial life skill to students with intellectual disabilities with the purpose of empowering them to lead more independent lives.

Positive acceptance and motivation toward robotics learning approaches by students could provide an affective component to learners and encourage their participation in educational settings, increasing their overall well-being. Nevertheless, despite its full potential to allow more scalable didactic supports to improve student engagement, implementation of robotics academic intervention in real special education environments is still scarce (Adams et al., 2017; Den Brok & Sterkenburg, 2015). Current evidence consists of mostly case studies in relatively artificial conditions (Adams et al., 2017), where students used the robotic approaches for a short period at school but with a research team present. One possible explanation is that although students with learning difficulties have shown a positive perception and a significantly higher willingness to use robots within educational contexts, it seems that there is still some skepticism among the specialized teachers in the treatment of intellectual disabilities, who perceive the robot as an expensive and limited learning tool (Adams & Cook, 2017; Conti et al., 2017; Encarnação et al., 2017).

In a recent study that examined the views of disability service organization employees toward social robotics, many expressed concerns about safety, normality for disabled people, and artificial interactions (Wolbring & Yumakulov, 2014). Furthermore, findings of Encarnação et al. (2017) indicated that special education teachers had more reservations about the difficulties related to the use of robotics technology in the educational context when compared to regular education teachers. Even in some cases where teachers agreed that a robot system was useful and had a positive impact on students with intellectual disabilities, they also felt that the amount of time it took to do things with the robot was not always practical in the classroom due to time constraints and the energy level of the participants to complete the activities (Adams & Cook, 2017; Encarnação et al., 2017). In addition, the cost of an autonomous, intelligent robot instructor with a range of functions large enough to engage students with learning difficulties is considered prohibitive for most special education schools (Hedgecock et al., 2014; Pennington et al., 2014).

More research involving teachers into designing, developing, and implementing of robotics intervention in special education settings is needed. Only with their active participation, the synergistic integration of the pedagogical and academic protocols will be up to the standards and expectations of educational staff. On the other hand, the professional experience of educators could also help to identify practical issues, to adapt robotic solution to the needs and skills of learners, and to provide effective outcome measures for assessing the use of robotics learning approaches for students with intellectual disability.

Conclusions and Future Directions

In this chapter we have reviewed some of the most recent emerging technological developments that support learning for students with intellectual disabilities. In general, available research has shown that technological innovations used by students with learning difficulties have the potential for many positive academic outcomes. However, further control group instead of single-case studies are needed to assess the benefits and challenges of using emerging technological interventions for educational development and academic achievement in special education settings.

From an empirical point of view, it is clear that emerging technologies have made teaching for students with learning disabilities more flexible and adaptive. Nevertheless, it is also necessary to demonstrate which technology supports which kind of learning strategy or specific academic outcome better. Each technology can facilitate academic learning in different ways depending on its own features. Therefore, we desperately need robust research initiatives that focus on user experience design and validation of studies because current evidence in these research aims for most emerging technologies embedded in special education settings is scarce and weak.

Therefore, in agreement with Butcher & Jameson (2016) it is necessary to build a pedagogical framework that guides design and validation into emerging technology interventions for supporting academic performance and learning outcomes for students with intellectual disabilities. The lack of this framework makes it difficult to assess the effectiveness and validity of academic gains reported in individual interventions or small-group studies to determine the true impact of emerging technologies on special education sessions. This framework should be focused on identifying best practices in user-centered interface and experimental design. Thus, this pedagogical framework could provide a solid understanding of which technology or features of technology are the more effective to promote a specific academic achievement for students with learning disabilities. Moreover, this will help to optimize the use of technology for educational support and to accelerate the accumulation of valid knowledge for effective intervention (Butcher & Jameson 2016).

However, emerging technological developments cannot be considered as a panacea for teaching students with intellectual disabilities because the actual benefits of technology-assisted learning depend greatly on the quality and adequacy of the academic intervention. Therefore, to reach educational goals promoting motivational and independent learning, emerging technological approaches must be developed through the collaboration of an interdisciplinary team comprised of special education teachers, researchers, developers, therapists, and parents, as well as intervention programs must be expanded in different settings involving school and home and maintenance and generalization of academic achievement in post-intervention phases be systematically designed.

Finally, it is essential to emphasize that the development and implementation of emerging technologies within special education environments are mainly aimed at providing teachers with improved support and feedback for students with intellectual disabilities in order to facilitate successful social integration. In this sense, future research should also address how to assess the acquired academic competences by emerging technology intervention within meaningful environments which can potentially result in increased access of these traditionally marginalized persons to an expanded social context.

References

Adams, K. D., Alvarez, L., & Rios-Rincon, A. M. (2017). Robotic systems for augmentative manipulation to promote cognitive development, play, and education. In A. Cook & P. Encarnação (Eds.), *Principles and practice of rehabilitation robots* (pp. 219–260). Boca Raton, FL: CRC Press.

Adams, K. D., & Cook, A. M. (2017). Performing mathematics activities with non-standard units of measurement using robots controlled via speech-generating devices: Three case studies. *Disability and Rehabilitation: Assistive Technology, 12*(5), 491–503.

Algozzine, B., Browder, D., Karvonen, M., Test, D. W., & Wood, W. M. (2001). Effects of interventions to promote self-determination for individuals with disabilities. *Review of Educational Research, 71*(2), 219–277.

Alnahdi, G. (2014). Assistive technology in special education and the universal design for learning. *Turkish Online Journal of Educational Technology, 13*(2), 18–23.

Altınay-Gazi, Z., & Altınay-Aksal, F. (2017). Technology as mediation tool for improving teaching profession in higher education practices. *Eurasia Journal of*

Mathematics, Science & Technology Education, 13(3), 803–813.

Anderson, C. L., Anderson, K. M., & Cherup, S. (2009). Investment vs. return: Outcomes of special education technology research in literacy for students with mild disabilities. *Contemporary Issues in Technology and Teacher Education, 9*(3), 337–355.

Ayres, K. M., Mechling, L., & Sansosti, F. J. (2013). The use of mobile technologies to assist with life skills/independence of students with moderate/severe intellectual disability and/or autism spectrum disorders: Considerations for the future of school psychology. *Psychology in the Schools, 50*(3), 259–271.

Bakker, M., van den Heuvel-Panhuizen, M., & Robitzsch, A. (2016). Effects of mathematics computer games on special education students' multiplicative reasoning ability. *British Journal of Educational Technology, 47*(4), 633–648.

Barnard-Brak, L., Thompson, S., Wei, T., & Richman, D. (2014). Assistive technology as a predictor of general or alternate assessment among elementary-aged students with autism spectrum disorders. *Assistive Technology, 26*(2), 81–87.

Bosseler, A., & Massaro, D. W. (2003). Development and evaluation of a computer-animated tutor for vocabulary and language learning in children with autism. *Journal of Autism and Developmental Disorders, 33*(6), 653–672.

Brown, J. S., & Duguid, P. (2017). *The social life of information: Updated, with a new preface*. Boston, MA: Harvard Business Review Press.

Butcher, K. R., & Jameson, J. M. (2016). Computer-based instruction (CBI) within special education. In J. Luiselli & A. Fischer (Eds.), *Computer-assisted and web-based innovations in psychology, special education, and health* (pp. 211–254). San Diego, CA: Academic Press.

Cacioppo, J. T., & Petty, R. E. (1979). Effects of message repetition and position on cognitive response, recall, and persuasion. *Journal of Personality and Social Psychology, 37*(1), 97–109.

Cannella, H. I., O'Reilly, M. F., & Lancioni, G. E. (2006). Treatment of hand mouthing in individuals with severe to profound developmental disabilities: A review of the literature. *Research in Developmental Disabilities, 27*(5), 529–544.

Carey, A. C., Friedman, M. G., & Bryen, D. N. (2005). Use of electronic technologies by people with intellectual disabilities. *Mental Retardation, 43*(5), 322–333.

Carlson, L. (2009). *The faces of intellectual disability: Philosophical reflections*. Bloomington, IN: Indiana University Press.

Carver, J., Ganus, A., Ivey, J. M., Plummer, T., & Eubank, A. (2016). The impact of mobility assistive technology devices on participation for individuals with disabilities. *Disability and Rehabilitation: Assistive Technology, 11*(6), 468–477.

Chadwick, D., & Wesson, C. (2016). Digital inclusion and disability. In A. Attrill & C. Fullwood (Eds.), *Applied cyberpsychology* (pp. 1–23). London: Palgrave Macmillan.

Chang, C. W., Lee, J. H., Wang, C. Y., & Chen, G. D. (2010). Improving the authentic learning experience by integrating robots into the mixed-reality environment. *Computers & Education, 55*(4), 1572–1578.

Cho, K., & Shin, C. (2011). Caregiving intervention for children with autism spectrum disorders using an animal robot. In *Proceedings of the 6th International Conference on Human-Robot Interaction (HRI), March 2011, Lausanne, Switzerland* (pp. 399–400). New York, NY: ACM.

Clarfield, J., & Stoner, G. (2005). The effects of computerized reading instruction on the academic performance of students identified with ADHD. *School Psychology Review, 34*(2), 246.

Constantin, A., Johnson, H., Smith, E., Lengyel, D., & Brosnan, M. (2017). Designing computer-based rewards with and for children with autism spectrum disorder and/or intellectual disability. *Computers in Human Behavior, 75*, 404–414.

Conti, D., Di Nuovo, S., Buono, S., & Di Nuovo, A. (2017). Robots in education and care of children with developmental disabilities: A study on acceptance by experienced and future professionals. *International Journal of Social Robotics, 9*(1), 51–62.

Cook, A. M., Bentz, B., Harbottle, N., Lynch, C., & Miller, B. (2005). School-based use of a robotic arm system by children with disabilities. *IEEE Transactions on Neural Systems and Rehabilitation Engineering, 13*(4), 452–460.

Cook, A. M., Meng, M. H., Gu, J. J., & Howery, K. (2002). Development of a robotic device for facilitating learning by children who have severe disabilities. *IEEE Transactions on Neural Systems and Rehabilitation Engineering, 10*(3), 178–187.

Cortiella, C., & Horowitz, S. H. (2014). *The state of learning disabilities: Facts, trends and emerging issues*. New York, NY: National Center for Learning Disabilities.

Costa, S. C. C., Soares, F., Santos, C., Pereira, A. P. D. S., & Moreira, M. F. (2016). Lego robots & autism spectrum disorders: A potential partnership? *Revista de Estudios e Investigación en Psicología y Educación, 3*(1), 50–59.

Crossley, S. A., & Mcnamara, D. S. (Eds.). (2016). *Adaptive educational technologies for literacy instruction*. New York, NY: Routledge.

Daunhauer, L. A., Fidler, D. J., & Will, E. (2014). School function in students with Down syndrome. *The American Journal of Occupational Therapy, 68*(2), 167–176.

Davies, D. K., Stock, S. E., King, L. R., Brown, R. B., Wehmeyer, M. L., & Shogren, K. A. (2015). An interface to support independent use of Facebook by people with intellectual disability. *Intellectual and Developmental Disabilities, 53*(1), 30–41.

Dekelver, J., Kultsova, M., Shabalina, O., Borblik, J., Pidoprigora, A., & Romanenko, R. (2015). Design of mobile applications for people with intellectual disabilities. *Communications in Computer and Information Science, 535*, 823–836.

Den Brok, W. L. J. E., & Sterkenburg, P. S. (2015). Self-controlled technologies to support skill attainment in persons with an autism spectrum disorder and/or an intellectual disability: A systematic literature review. *Disability and Rehabilitation Assistive Technology, 10*(1), 1–10.

Díaz, L. D. E. (2017). The teaching and learning process of mathematics in the primary education stage: A constructivist proposal within the framework of key competencies. *International Electronic Journal of Mathematics Education, 12*(7), 709–713.

Ditchman, N., Kosyluk, K., Lee, E. J., & Jones, N. (2016). How stigma affects the lives of people with intellectual disabilities: An overview. In K. Scior & S. Werner (Eds.), *Intellectual disability and stigma* (pp. 31–47). London, UK: Palgrave Macmillan.

Dunst, C. J., & Hamby, D. W. (2012). Guide for calculating and interpreting effect sizes and confidence intervals in intellectual and developmental disabilities research studies. *Journal of Intellectual and Developmental Disability, 37*(2), 89–99.

Edyburn, D. L. (2013). Critical issues in advancing the special education technology evidence base. *Exceptional Children, 80*(1), 7–24.

Encarnação, P., Leite, T., Nunes, C., Nunes da Ponte, M., Adams, K., Cook, A., et al. (2017). Using assistive robots to promote inclusive education. *Disability and Rehabilitation: Assistive Technology, 12*(4), 352–372.

Felix, V. G., Mena, L. J., Ostos, R., & Maestre, G. E. (2017). A pilot study of the use of emerging computer technologies to improve the effectiveness of reading and writing therapies in children with Down syndrome. *British Journal of Educational Technology, 48*(2), 611–624.

Fernández-López, Á., Rodríguez-Fórtiz, M. J., Rodríguez-Almendros, M. L., & Martínez-Segura, M. J. (2013). Mobile learning technology based on iOS devices to support students with special education needs. *Computers & Education, 61*, 77–90.

Fitzgerald, G., Koury, K., & Mitchem, K. (2008). Research on computer-mediated instruction for students with high incidence disabilities. *Journal of Educational Computing Research, 38*(2), 201–233.

Fletcher-Watson, S. (2014). A targeted review of computer-assisted learning for people with autism spectrum disorder: Towards a consistent methodology. *Review Journal of Autism and Developmental Disorders, 1*(2), 87–100.

Fridin, M. (2014). Storytelling by a kindergarten social assistive robot: A tool for constructive learning in preschool education. *Computers & Education, 70*, 53–64.

Gersten, R., & Edyburn, D. (2007). Defining quality indicators for special education technology research. *Journal of Special Education Technology, 22*(3), 3–18.

Gersten, R., Fuchs, L. S., Compton, D., Coyne, M., Greenwood, C., & Innocenti, M. S. (2005). Quality indicators for group experimental and quasi-experimental research in special education. *Exceptional Children, 71*(2), 149–164.

Goldsmith, T. R., & LeBlanc, L. A. (2004). Use of technology in interventions for children with autism. *Journal of Early and Intensive Behavior Intervention, 1*(2), 166–178.

Graves, A., Mohamed, A. R., & Hinton, G. (2013). Speech recognition with deep recurrent neural networks. In *Proceedings of International Conference on Acoustics, Speech and Signal Processing (ICASSP), May 2013* (pp. 6645–6649). Vancouver, Canada: IEEE.

Hall, T. E., Cohen, N., Vue, G., & Ganley, P. (2015). Addressing learning disabilities with UDL and technology: Strategic reader. *Learning Disability Quarterly, 38*(2), 72–83.

Hall, T. E., Meyer, A., & Rose, D. H. (Eds.). (2012). *Universal design for learning in the classroom: Practical applications*. New York, NY: Guilford Press.

Hartman, E. (2015). Universal design for learning (UDL) and learners with severe support needs. *International Journal of Whole Schooling, 11*(1), 54–67.

Hedgecock, J., Standen, P. J., Beer, C., Brown, D., & Stewart, D. S. (2014). Evaluating the role of a humanoid robot to support learning in children with profound and multiple disabilities. *Journal of Assistive Technologies, 8*(3), 111–123.

Holburn, S., Nguyen, D., & Vietze, P. M. (2004). Computer-assisted learning for adults with profound multiple disabilities. *Behavioral Interventions, 19*(1), 25–37.

Hulme, C., & Mackenzie, S. (2014). *Working memory and severe learning difficulties*. Hove, UK: Psychology Press.

Jiao, L., Liu, Y., Yuan, A., & Bai, G. (2012). Offline handwritten English character recognition based on convolutional neural network. In *Proceedings of the 10th IAPR International Workshop on Document Analysis Systems (DAS)* (pp. 125–129). Gold Coast, Australia: IEEE.

Jonker, V., Wijers, M., & Van Galen, F. (2009). The motivational power of mini-games for the learning of mathematics. In *Proceedings of the 3rd European Conference on Game Based Learning (ECGBL). October 2009* (pp. 202–210). Graz, Austria: Academic Conferences Ltd.

Kazakoff, E. R., Sullivan, A., & Bers, M. U. (2013). The effect of a classroom-based intensive robotics and programming workshop on sequencing ability in early childhood. *Early Childhood Education Journal, 41*(4), 245–255.

Kim, E. S., Berkovits, L. D., Bernier, E. P., Leyzberg, D., Shic, F., Paul, R., & Scassellati, B. (2013). Social robots as embedded reinforcers of social behavior in children with autism. *Journal of Autism and Developmental Disorders, 43*(5), 1038–1049.

Kim, M. K., Park, Y., & Coleman, M. B. (2017). The quality of evidence in tablet-assisted interventions for students with disabilities. *Journal of Computer Assisted Learning, 33*(6), 547–561.

Kirkwood, A., & Price, L. (2014). Technology-enhanced learning and teaching in higher education: What is ‘enhanced’ and how do we know? A critical literature review. *Learning, Media and Technology, 39*(1), 6–36.

Kozima, H., Nakagawa, C., & Yasuda, Y. (2007). Children–robot interaction: A pilot study in autism therapy. *Progress in Brain Research, 164*, 385–400.

Kroesbergen, E. H., & Van Luit, J. E. H. (2003). Mathematics interventions for children with special educational needs: A meta-analysis. *Remedial and Special Education, 24*(2), 97–114.

Kulik, J. A. (2003). *Effects of using instructional technology in elementary and secondary schools: What controlled evaluation studies say*. Arlington, VA: SRI International.

Lachapelle, Y., Wehmeyer, M. L., Haelewyck, M. C., Courbois, Y., Keith, K. D., Schalock, R., et al. (2005). The relationship between quality of life and self-determination: An international study. *Journal of Intellectual Disability Research, 49*(10), 740–744.

Lancioni, G., O’Reilly, M., Singh, N., Green, V., Chiapparino, C., De Pace, C., et al. (2010). Use of microswitch technology and a keyboard emulator to support literacy performance of persons with extensive neuro-motor disabilities. *Developmental Neurorehabilitation, 13*(4), 248–257.

Lancioni, G. E., Comes, M. L., Stasolla, F., Manfredi, F., O'reilly, M. F., & Singh, N. N. (2005). A microswitch cluster to enhance arm-lifting responses without dystonic head tilting by a child with multiple disabilities. *Perceptual and Motor Skills, 100*(3), 892–894.

Lancioni, G. E., Sigafoos, J., O'Reilly, M. F., & Singh, N. N. (2012). *Assistive technology: Interventions for individuals with severe/profound and multiple disabilities*. New York, NY: Springer Science & Business Media.

Lancioni, G. E., Singh, N. N., O’Reilly, M. F., Sigafoos, J., Oliva, D., Boccasini, A., et al. (2013). Persons with multiple disabilities increase adaptive responding and control inadequate posture or behavior through programs based on microswitch-cluster technology. *Research in Developmental Disabilities, 34*(10), 3411–3420.

Lancioni, G. E., & Singh, N. N. (Eds.). (2014). *Assistive technologies for people with diverse abilities*. New York, NY: Springer Science & Business Media.

Lancioni, G. E., Singh, N. N., O’reilly, M. F., Oliva, D., Smaldone, A., Tota, A., … Groeneweg, J. (2006). Assessing the effects of stimulation versus microswitch-based programmes on indices of happiness of students with multiple disabilities. *Journal of Intellectual Disability Research, 50*(10), 739–747.

Lancioni, G. E., Singh, N. N., O’Reilly, M. F., Sigafoos, J., Didden, R., Manfredi, F., et al. (2009). Fostering locomotor behavior of children with developmental disabilities: An overview of studies using treadmills and walkers with microswitches. *Research in Developmental Disabilities, 30*(2), 308–322.

Lancioni, G. E., Singh, N. N., O’Reilly, M. F., Sigafoos, J., Oliva, D., Gatti, M., et al. (2008). A microswitch-cluster program to foster adaptive responses and head control in students with multiple disabilities: Replication and validation assessment. *Research in Developmental Disabilities, 29*(4), 373–384.

Lancioni, G. E., Singh, N. N., Oliva, D., Scalini, L., & Groeneweg, J. (2003). Microswitch clusters to enhance non-spastic response schemes with students with multiple disabilities. *Disability and Rehabilitation, 25*(6), 301–304.

Lancioni, G. E., Singh, N. N., O'Reilly, M. F., Oliva, D., Scalini, L., Vigo, C. M., & Groeneweg, J. (2004). Microswitch clusters to support responding and appropriate posture of students with multiple disabilities: Three case evaluations. *Disability and Rehabilitation, 26*(9), 501–505.

Lancioni, G. E., Singh, N. N., O'Reilly, M. F., Sigafoos, J., Didden, R., Oliva, D., & Cingolani, E. (2008). A girl with multiple disabilities increases object manipulation and reduces hand mouthing through a microswitch-based program. *Clinical Case Studies, 7*(3), 238–249.

Lehmann, H., Iacono, I., Dautenhahn, K., Marti, P., & Robins, B. (2014). Robot companions for children with down syndrome: A case study. *Interaction Studies, 15*(1), 99–112.

Leopold, A., Lourie, A., Petras, H., & Elias, E. (2015). The use of assistive technology for cognition to support the performance of daily activities for individuals with cognitive disabilities due to traumatic brain injury: The current state of the research. *NeuroRehabilitation, 37*(3), 359–378.

Lewandowski, L., Wood, W., & Miller, L. A. (2016). Technological applications for individuals with learning disabilities and ADHD. In J. Luiselli & A. Fischer (Eds.), *Computer-assisted and web-based innovations in psychology, special education, and health* (pp. 61–93). San Diego, CA: Academic Press.

Liu, G. Z., Wu, N. W., & Chen, Y. W. (2013). Identifying emerging trends for implementing learning technology in special education: A state-of-the-art review of selected articles published in 2008–2012. *Research in Developmental Disabilities, 34*(10), 3618–3628.

Lussier-Desrochers, D., Normand, C. L., Romero-Torres, A., Lachapelle, Y., Godin-Tremblay, V., Dupont, M. È., … Bilodeau, P. (2017). Bridging the digital divide for people with intellectual disability. *Cyberpsychology: Journal of Psychosocial Research on Cyberspace, 11*(1). https://doi.org/10.5817/CP2017-1-1

Mayer, R. E. (2005). Principles for managing essential processing in multimedia learning: Segmenting, pretraining, and modality principles. In R. Mayer (Ed.), *The Cambridge handbook of multimedia learning* (pp. 169–182). New York, NY: Cambridge University Press.

McDougall, J., Evans, J., & Baldwin, P. (2010). The importance of self-determination to perceived quality of life for youth and young adults with chronic conditions and disabilities. *Remedial and Special Education, 31*(4), 252–260.

McLay, L., van der Meer, L., Schäfer, M. C., Couper, L., McKenzie, E., O'Reilly, M. F., & Sutherland, D. (2015). Comparing acquisition, generalization, maintenance, and preference across three AAC options in four children with autism spectrum disorder. *Journal of Developmental and Physical Disabilities, 27*(3), 323–339.

Mechling, L. C. (2006). Comparison of the effects of three approaches on the frequency of stimulus activations, via a single switch, by students with profound intellectual disabilities. *The Journal of Special Education, 40*(2), 94–102.

Meyers, L. F. (1988). Using computers to teach children with Down syndrome spoken and written language skills. In L. Nadel (Ed.), *The psychobiology of Down syndrome* (pp. 247–265). New York, NY: NDSS.

Murphy, R., Penuel, W. R., Means, B., Korbak, C., Whaley, A., & Allen, J. E. (2002). *E-DESK: A review of recent evidence on the effectiveness of discrete educational software*. Palo Alto, CA: SRI International.

Newman, L., Browne-Yung, K., Raghavendra, P., Wood, D., & Grace, E. (2017). Applying a critical approach to investigate barriers to digital inclusion and online social networking among young people with disabilities. *Information Systems Journal, 27*(5), 559–588.

Ortega, J. M., & Gómez, C. J. (2006). Computer-assisted teaching and mathematical learning in down syndrome children. *Journal of Computer Assisted Learning, 22*(4), 298–307.

Park, E., & Kwon, S. J. (2016). I can teach them: The ability of robot instructors to cognitive disabled children. *Journal of Psychological and Educational Research, 24*(1), 101–114.

Pennington, R., Welch, K. C., & Scott, R. (2014). Using robot-assisted instruction to teach students with intellectual disabilities to use personal narrative in text messages. *Journal of Special Education Technology, 29*(4), 49–58.

Petress, K. (2008). What is meant by "active learning?". *Education, 128*(4), 566–570.

Petry, K., Maes, B., & Vlaskamp, C. (2005). Domains of quality of life of people with profound multiple disabilities: The perspective of parents and direct support staff. *Journal of Applied Research in Intellectual Disabilities, 18*(1), 35–46.

Pioggia, G., Igliozzi, R., Sica, M. L., Ferro, M., Muratori, F., Ahluwalia, A., & De Rossi, D. (2008). Exploring emotional and imitational android-based interactions in autistic spectrum disorders. *Journal of CyberTherapy & Rehabilitation, 1*(1), 49–61.

Reardon, C., Zhang, H., Wright, R., & Parker, L. E. (2015). Response prompting for intelligent robot instruction of students with intellectual disabilities. In *Proceedings of 24th International Symposium on Robot and Human Interactive Communication (RO-MAN)*. August 2015, Kobe, Japan. IEEE, pp. 784–790.

Robins, B., Dautenhahn, K., Te Boekhorst, R., & Billard, A. (2005). Robotic assistants in therapy and education of children with autism: Can a small humanoid robot help encourage social interaction skills? *Universal Access in the Information Society, 4*(2), 105–120.

Rose, D. (2000). Universal design for learning. *Journal of Special Education Technology, 15*(4), 47–51.

Sansosti, F. J., Doolan, M. L., Remaklus, B., Krupko, A., & Sansosti, J. M. (2015). Computer-assisted interventions for students with autism spectrum disorders within school-based contexts: A quantitative meta-analysis of single-subject research. *Review Journal of Autism and Developmental Disorders, 2*(2), 128–140.

Sansosti, F. J., Powell-Smith, K. A., & Cowan, R. J. (2010). *High functioning autism/Asperger syndrome in schools: Assessment and intervention*. New York, NY: Guilford.

Saunders, M. D., Timler, G. R., Cullinan, T. B., Pilkey, S., Questad, K. A., & Saunders, R. R. (2003). Evidence of contingency awareness in people with profound multiple impairments: Response duration versus response rate indicators. *Research in Developmental Disabilities, 24*(4), 231–245.

Schalock, R. L., Brown, I., Brown, R., Cummins, R. A., Felce, D., Matikka, L., et al. (2002). Conceptualization, measurement, and application of quality of life for persons with intellectual disabilities: Report of an international panel of experts. *Mental Retardation, 40*(6), 457–470.

Shah, N. (2011). Special education pupils find learning tool in iPad applications. *Education Week, 30*(22), 1–16.

Shalev, R. A., Asmus, J. M., Carter, E. W., & Moss, C. K. (2016). Attitudes of high school students toward their classmates with severe disabilities: A pilot study. *Journal of Developmental and Physical Disabilities, 28*(4), 523–538.

Shaw, R., & Lewis, V. (2005). The impact of computer-mediated and traditional academic task presentation on the performance and behaviour of children with ADHD. *Journal of Research in Special Educational Needs, 5*(2), 47–54.

Sorden, S. D. (2005). A cognitive approach to instructional design for multimedia learning. *Informing Science Journal, 8*, 263–279.

Spittle, A. J., & Orton, J. (2014). Cerebral palsy and developmental coordination disorder in children born preterm. *Seminars in Fetal and Neonatal Medicine, 19*(2), 84–89.

Standen, P. J., Brown, D. J., & Cromby, J. J. (2001). The effective use of virtual environments in the education and rehabilitation of students with intellectual disabilities. *British Journal of Educational Technology, 32*(3), 289–299.

Standen, P. J., Brown, D. J., Roscoe, J., Hedgecock, J., Stewart, D., Trigo, M. J. G., & Elgajiji, E. (2014). Engaging students with profound and multiple disabilities using humanoid robots. In *8th International Conference on Universal Access in Human-Computer Interaction (UAHCI). June 2014, Crete, Greece* (pp. 419–430). Cham: Springer.

Stasolla, F., Boccasini, A., Perilli, V., Caffo, A. O., Damiani, R., & Albano, V. (2014). A selective overview of microswitch-based programs for promoting adaptive behaviors of children with developmental disabilities. *International Journal of Ambient Computing and Intelligence, 6*(2), 56–74.

Stasolla, F., Damiani, R., & Caffo, A. O. (2014). Promoting constructive engagement by two boys with autism spectrum disorders and high functioning through behavioural interventions. *Research in Autism Spectrum Disorders, 8*(4), 376–380.

Stasolla, F., Damiani, R., Perilli, V., D'Amico, F., Caffo, A. O., Stella, A., et al. (2015). Computer and microswitch-based programs to improve academic activities by six children with cerebral palsy. *Research in Developmental Disabilities, 45*, 1–13.

Stasolla, F., Perilli, V., Damiani, R., Caffo, A. O., Di Leone, A., Albano, V., et al. (2014). A microswitch-cluster program to enhance object manipulation and to reduce hand mouthing by three boys with autism spectrum disorders and intellectual disabilities. *Research in Autism Spectrum Disorders, 8*(9), 1071–1078.

Stock, S. E., Davies, D. K., Wehmeyer, M. L., & Palmer, S. B. (2008). Evaluation of cognitively accessible software to increase independent access to cellphone technology for people with intellectual disability. *Journal of Intellectual Disability Research, 52*(12), 1155–1164.

Tanimoto, S., Thompson, R., Berninger, V. W., Nagy, W., & Abbott, R. D. (2015). Computerized writing and reading instruction for students in grades 4–9 with specific learning disabilities affecting written language. *Journal of Computer Assisted Learning, 31*(6), 671–689.

Teo, T., & Zhou, M. (2017). The influence of teachers' conceptions of teaching and learning on their technology acceptance. *Interactive Learning Environments, 25*(4), 513–527.

Wacker, D. P., Wiggins, B., Fowler, M., & Berg, W. K. (1988). Training students with profound or multiple handicaps to make requests via microswitches. *Journal of Applied Behavior Analysis, 21*(4), 331–343.

Walcott, C. M., Marett, K., & Hessel, A. B. (2014). Effectiveness of a computer-assisted intervention for young children with attention and reading problems. *Journal of Applied School Psychology, 30*(2), 83–106.

Weng, P. L., Maeda, Y., & Bouck, E. C. (2014). Effectiveness of cognitive skills-based computer-assisted instruction for students with disabilities: A synthesis. *Remedial and Special Education, 35*(3), 167–180.

Williams, C., Wright, B., Callaghan, G., & Coughlan, B. (2002). Do children with autism learn to read more readily by computer assisted instruction or traditional book methods? A pilot study. *Autism, 6*(1), 71–91.

Wolbring, G., & Yumakulov, S. (2014). Social robots: Views of staff of a disability service organization. *International Journal of Social Robotics, 6*(3), 457–468.

Wolery, M., Ault, M. J., & Doyle, P. M. (1992). *Teaching students with moderate to severe disabilities: Use of response prompting strategies*. New York, NY: Longman Publishing Group.

Part X

Treatment for Dual Diagnosis

44

Anxiety and Phobias in Individuals with Intellectual Disabilities

Lauren J. Moskowitz, Megan Braconnier,
and Melissa Jeffay

Background

History of Anxiety in ID

Prior to the last 40 years, it was generally assumed that mental health problems such as anxiety disorders could not exist in individuals with intellectual disabilities (ID) (Matson, Belva, Hattier, & Matson, 2012; Reardon, Gray, & Melvin, 2015). Anxiety is a construct that involves (a) subjective mental state inferred from verbal reports of *emotions* (e.g., subjective fear and panic experienced) and *cognitions* (e.g., beliefs, thoughts, images, such as worrying, "I'm going to get hurt"), (b) *behaviors* indicating fear or anxiety (e.g., physically escaping or avoiding the feared stimulus or situation, such as running out of the room, whining, crying, or pacing), and (c) *physiological arousal* (e.g., heart racing, palms sweating, muscles tightening) (Barlow, 2000). The difficulty diagnosing anxiety disorders in individuals with ID may be due in large part to the communication deficits often present in this population, since diagnosis of an anxiety disorder typically requires verbal report of cognitions and affect. In addition to difficulties with communication and introspection, anxiety may also be unrecognized or overlooked in this population because of the atypical manifestation of anxiety disorders in individuals with ID (Helverschou & Martinsen, 2011); the misattribution of symptoms of anxiety to the intellectual disability (i.e., "diagnostic overshadowing," Reiss, Levitan, & Szyszko, 1982) or to problem behavior (i.e., "behavioral overshadowing"); and the reluctance of behavior analysts to use the construct of anxiety to describe or explain behavior in those with ID, given that—unlike behaviors—the cognitive/affective and physiological components of anxiety cannot be directly observed (Groden, Cautela, Prince & Berryman, 1994).

Prevalence of Anxiety in ID

In spite of these difficulties identifying anxiety in ID, it is increasingly recognized that individuals with ID do experience fear and anxiety, at increased rates in comparison to the general population. The prevalence of anxiety disorders ranges from 3% to 22% in youth with ID (see Reardon et al., 2015 for a review) and 2–17% in adults with ID (e.g., Deb, Thomas, & Bright, 2001; Reid, Smiley, & Cooper, 2011), depending on the methods used to diagnose anxiety and on the sample (e.g., whether including individuals with severe or profound ID). The rate of phobias is significantly higher in adults with ID compared

---

L. J. Moskowitz (✉) · M. Braconnier · M. Jeffay
Department of Psychology, St. John's University,
Queens, NY, USA
e-mail: Moskowil@stjohns.edu

J. L. Matson (ed.), *Handbook of Intellectual Disabilities*, Autism and Child Psychopathology Series, https://doi.org/10.1007/978-3-030-20843-1_44

to the general population (Deb et al., 2001). Even higher rates of anxiety are reported in some genetic syndromes associated with ID, most notably fragile X syndrome (FXS), with 70–83% of individuals meeting criteria for at least one anxiety disorder, the most common being specific phobia and social phobia (Bailey, Raspa, Olmstead, & Holiday, 2008; Cordeiro, Ballinger, Hagerman, & Hessl, 2011), and Williams syndrome (WS), with a rate of anxiety disorders at approximately 48%, the most common being specific phobia and generalized anxiety disorder (GAD) (Royston, Howlin, Waite, & Oliver, 2017). Anxiety was also found to be three times as common in individuals with ID and comorbid autism spectrum disorder (ASD) (Gillott & Standen, 2007).

Several researchers have found that children without ID (IQ ≥ 70) were rated by parents as displaying more symptoms of anxiety than those with ID (IQ < 70) (Sukhodolsky et al., 2008; Witwer & Lecavalier, 2010) and that anxiety disorders are more common in those with mild ID than moderate to profound ID (Cooper, Smiley, Morrison, Williamson, & Allan, 2007; Dekker, Koot, van der Ende, & Verhulst, 2002). This may be because individuals with higher intellectual functioning are likely to have a greater understanding of their condition than those with lower intellectual functioning, which presumably leads to increased anxiety. Alternatively, these findings may be due to the aforementioned difficulty of identifying anxiety in individuals with more severe ID. Thus, it may be that parents, teachers, staff, or other informants may not recognize the person's fear/anxiety and may even attribute the person's aggression, self-injury, or tantrums to disobedience, oppositionality, or anger/irritability rather than to fear or anxiety.

Definition of the Anxiety Disorders

DSM-5 Anxiety Disorders

The *Diagnostic and Statistical Manual of Mental Disorders* (*DSM-5*; APA, 2013) defines anxiety as "the apprehensive anticipation of future danger or misfortune accompanied by a feeling of worry, distress, and/or somatic symptoms of tension." Although some researchers (e.g., Brown, Aman, & Lecavalier, 2004) have argued that *DSM* criteria are applicable to individuals with mild or moderate ID, other researchers have questioned the applicability of the existing diagnostic system to individuals with ASD, DD, and ID (e.g., Leyfer, Woodruff-Borden, Klein-Tasman, Fricke, and Mervis, 2008). For example, results of a study by Leyfer et al. (2008) suggested the need either to modify the definition of GAD or create a new category of anxiety to capture the "anticipatory" anxiety exhibited by a large proportion of children with WS. To begin to address the limitations in applying *DSM-5* criteria to those with ID, the *Diagnostic Manual for Diagnosing Mental Disorders in Persons with ID* (*DM-ID-2*) was created to adapt *DSM-5* diagnostic criteria for individuals with ID. In the *DM-ID-2*, Cooray et al. (2016) make the point that – to receive a diagnosis of most *DSM* anxiety disorders – the person must have some ability to recognize and describe his emotions and/or thoughts (more so for anxiety and worry than for fear), which may be difficult or impossible for many people with ID. However, with visual prompts, some individuals with mild to moderate ID may be able to describe worries and rate time spent worrying and related distress (Cooray et al., 2016). Further, fear can often be observed through behavioral and/or physiological indicators for individuals who are unable to self-report, as described for the following *DSM* disorders. Caretakers are often able to recognize subtle facial expressions and other behaviors associated with anxiety in individuals with ID (e.g., Helverschou & Martinsen, 2011). As such, some researchers have attempted to identify behavioral descriptors of DSM anxiety symptoms (e.g., Charlot et al., 2007).

The *DSM-5* defines 11 types of anxiety disorders, which we will now describe. All of the disorders with the exception of selective mutism involve fear ("the emotional response to real or perceived danger"), anxiety ("anticipation of future threat"), and/or worry ("apprehensive expectation"). To receive a diagnosis for any of the anxiety disorders, the person's fear or anxiety must be severe enough to cause significant distress or impairment, interfering with his/her social, occupational, or other areas of functioning. To meet criteria for most of the anxiety disorders, the fear/anxiety must be out of proportion to the actual danger posed by the situation, be persistent (≥ 6 months), and not be better explained by another mental disorder.

Separation Anxiety Disorder (SAD). SAD is defined by developmentally inappropriate and excessive fear or anxiety regarding separation from an attachment figure, as evidenced by at least three of eight symptoms (APA, 2013). The *DM-ID-2* does not recommend any modifications for diagnosing SAD in individuals with ID, given that fear can be observed rather than subjectively described for three of the symptoms (e.g., distress when anticipating or experiencing separation from home or attachment figures, reluctance/refusal to go away from home or go to sleep without attachment figure). However, assessing whether nightmares and complaints of physical symptoms pertain specifically to separation and assessing the more cognitive symptoms of SAD (e.g., worry about harm befalling attachment figures) may still be challenging in individuals with ID. For example, in Moskowitz et al. (2013), although a child with ASD and ID exhibited recurrent excessive distress when he anticipated or experienced separation from his parents, it was difficult for his parents to know whether he was worried about losing them or harm befalling them or that an event would lead to separation, as he was unable to articulate such worries.

Selective Mutism (SM). SM is defined as failure to speak in social situations in which the person is expected to speak (e.g., school) for at least 1 month, despite speaking to a select few people in other situations (APA, 2013). The failure to speak is not due to a lack of knowledge of the spoken language required in the social situation. The *DM-ID-2* does not recommend any modifications for diagnosing SM in individuals with ID, given that failure to speak can be directly observed (no subjective inferences required).

Specific Phobia (SP) and Social Phobia (SoP). Individuals with SP display marked fear, anxiety, or avoidance about a circumscribed object or situation (e.g., heights, animals) that is almost always immediately provoked by the phobic situation, is persistent, and is out of proportion to the actual risk posed (APA, 2013). Individuals with SoP, or social anxiety disorder, are fearful or anxious about social situations in which they might be scrutinized by others, including social interactions (e.g., meeting unfamiliar people), being observed, and performing in front of others (e.g., giving a speech) (APA, 2013). This involves fear of being negatively evaluated by others (i.e., being embarrassed, humiliated, and rejected or offending others). These social situations are avoided or endured with intense fear/anxiety. For children who cannot verbalize fear or anxiety, the *DSM-5* allows fear or anxiety to be expressed by "crying, tantrums, freezing, or clinging" for both SP and SoP (APA, 2013). Although the *DSM-5* does not specify any such modification for individuals with ID, Cooray et al. (2016) recommend using this modifier for adults with ID who are functioning at a similar developmental level so that fear can be observed rather than subjectively described. Although with the *DSM-5* it may still be difficult for individuals with ID to recognize or articulate fear of embarrassment or humiliation or rejection, one positive development is that the criterion for both SP and SoP was revised from "the person recognizes that the fear is excessive or unreasonable" in the *DSM-IV* to "the fear or anxiety is out of proportion to the actual threat" in the *DSM-5*, which may make it easier for individuals with ID to meet this criterion without having to express "recognition."

Panic Disorder (PD). PD involves recurrent unexpected panic attacks, which are abrupt surges of intense fear or discomfort that reach a peak within minutes accompanied by physical symptoms (e.g., pounding heart, sweating, trembling, shortness of breath, chest pain, nausea, dizziness, chills) and/or cognitive symptoms (derealization or depersonalization, fear of losing control or "going crazy," fear of dying). To meet criteria for PD, at least one attack must be followed by ≥1 month of persistent worry about having more panic attacks and/or a maladaptive change in behavior to try to avoid having panic attacks. Given that an individual with even mild ID might have difficulty with that temporal sequencing, the *DM-ID-2* recommends identifying 1 month of time using a time frame or anchor events or description of time frame from informant report (Cooray et al., 2016). Individuals with ID may not be able to articulate the more abstract cognitive symptoms of being detached from oneself, feelings of unreality, fear or losing control, and/or fear of dying, making it more difficult for them to reach the criterion threshold for PD. The *DM-ID-2* recommends modifying the *DSM-5* criteria for those with moderate to profound ID so that a panic attack can be either observed or reported and recommends reducing the criteria from ≥4 to ≥3 symptoms for those with severe to profound ID. The *DM-ID-2* also suggests relying on direct behavioral observations for those with severe to profound ID, such as identifying racing heartbeats by taking pulse, shortness of breath by observing overbreathing or hyperventilation, or chills or heat sensations by observing flushing (Cooray et al., 2016).

Agoraphobia. Agoraphobia involves marked fear or anxiety regarding two or more of five situations: using public transportation, being in open spaces, being in enclosed places, standing in line or being in a crowd, and being outside of the home alone (APA, 2013). To meet the *DSM-5* criteria for agoraphobia, the individual must be able to express that he fears/avoids these situations because of thoughts that escape might be difficult or help might not be available if he/she develops panic-like or other incapacitating symptoms (APA, 2013) – thoughts that may be difficult for individuals with more severe ID to express. Thus, the *DM-ID-2* recommends modifying the criteria for agoraphobia for those with severe or profound ID so that fear can be observed rather than subjectively described. In addition, *DSM-5* requires that the agoraphobic situations are actively avoided, require the presence of a companion, or are endured with intense fear or anxiety. The *DM-ID-2* cautions that avoidance of agoraphobic situations may not be prominent in individuals with moderate to profound ID if they are not given the opportunity to make choices such as where they go, what they do, and what they avoid (Cooray et al., 2016). Further, the *DM-ID-2* recommends establishing that individuals with ID require the presence of a companion due to fear/anxiety rather than due to low adaptive functioning.

Generalized Anxiety Disorder (GAD). GAD is characterized by excessive, persistent, and uncontrollable anxiety and worry about a variety of areas (e.g., work, school performance) (APA, 2013). Given that it may be difficult for people with severe or profound ID to verbalize thoughts of anxiety and/or worry, the *DM-ID-2* recommends modifying the criteria for GAD so that fear or anxiety can be observed rather than subjectively described (as with SoP, PD, and agoraphobia) and eliminating the criterion that the person finds it difficult to control the worry (Cooray et al., 2016). In addition, to meet *DSM-5* criteria for GAD, one must frequently experience three out of six physical symptoms including restlessness, being easily fatigued, irritability, sleep disturbance, muscle tension, and difficulty concentrating or mind going blank. Individuals with ID may be unable to articulate these symptoms, particularly the last two. As such, the *DM-ID-2* recommends including a modifier for individuals with severe to profound ID that only one of these six physical symptoms is required, a modification that the *DSM-5* includes for children.

Obsessive-Compulsive Disorder (OCD). Although OCD is now included in its own section in the *DSM*-5 due to its distinction from the aforementioned anxiety disorders (it was subsumed under anxiety disorders in *DSM-IV*), we will mention OCD in this chapter because obsessions and compulsions are frequently observed in those with ID (Matson & Dempsey, 2009). OCD is an anxiety disorder characterized by the presence of obsessions (recurrent and persistent thoughts, urges, or images that are intrusive and unwanted) and/or compulsions (repetitive behaviors or mental acts aimed at preventing/reducing anxiety or distress) that are time-consuming or cause clinically significant distress or impairment in important areas of functioning (APA, 2013). While anxiety or distress occurs in most individuals with OCD, this is no longer required for a diagnosis in *DSM-5*, which facilitates making the diagnosis of OCD in individuals with ID. Although the *DM-ID-2* does not recommend specific adaptations to the *DSM-5* criteria, they note that obsessions may not be experienced as intrusive/unwanted for some individuals with ID and that they may not attempt to ignore or suppress or neutralize their obsessions (Cooray et al., 2016). They further note that repetitive behaviors or mental acts may be difficult to elicit due to cognitive deficits and limited expressive language skills and suggest considering ordering, telling, asking, or repetitive physical acts as compulsions. Importantly, they caution that it might not be possible to ascertain the function of the compulsion (i.e., to prevent or reduce distress) in some individuals with ID, which is traditionally the main way that compulsions have been discriminated from stereotypic/repetitive behavior in ASD and/or ID.

Other Specified Anxiety Disorder (OSAD). Many individuals with ID may be diagnosed with OSAD (formerly *anxiety disorder not otherwise specified* in *DSM-IV*) because their fears and anxieties do not clearly fit into any *DSM-5* category. Although some of the *DSM-5* anxiety disorders may be applicable to those with ID, traditional *DSM*-consistent symptoms are more often present in individuals with intact intellectual and language ability (Kerns et al., 2014). Many anxiety-like behaviors occur in people with ID and other DDs – particularly those with ASD – that may not meet the criteria for any *DSM-5* disorder except OSAD (Kerns et al., 2014). For example, Kerns et al. (2014) found that almost half of youth with ASD (IQ, 67–158) displayed “atypical anxiety” – impairing symptoms of anxiety that were not consistent with any *DSM* disorders, the most common type (22%) being worry around routine, novelty, and restricted interests (e.g., changes in daily schedule, losing access to a special interest, rule-breaking). Other atypical anxieties included unusual specific fears (12%, e.g., fear of radio jingle), social fearfulness without the awareness of social judgment required to meet SoP (8.5%), and compulsive behavior in the absence of a clear desire to prevent distress or feared outcome as in OCD (8.5%) (Kerns et al., 2014). These atypical symptoms may reflect a more diffuse form of anxiety that is not addressed by the *DSM* but which still may be impairing (Kerns et al., 2014). As such, Kerns et al. (2016) proposed a four-step framework for differential diagnosis of anxiety disorders in ASD (many of whom have ID). For example, an individual with ASD and/or ID who presents with anxiety about transitions and routine changes might not experience the range of worries or feared consequences (“what if?”) that are characteristic of GAD yet nonetheless experiences anticipatory anxiety that is (1) excessive for his developmental level, (2) causes significant impairment to functioning, (3) is distinguishable from sensory difficulties, and (4) is above and beyond what can be explained by ASD or ID alone, which would warrant a *DSM-5* diagnosis of OSAD (excessive worry about change) (Kerns et al., 2016).

Finally, within the anxiety disorder category, the *DSM-5* also includes diagnoses for *substance-/medication-induced anxiety disorder* (which involves anxiety due to substance intoxication/withdrawal or to a medication treatment), *anxiety disorder due to another medical condition* (anxiety symptoms are attributed to the physiological consequences of another medical condition such as hyperthyroidism), and *unspecified anxiety disorder*.

In sum, although the *DSM-5* has rearranged the anxiety disorders into separate groups of classical anxiety disorders that now include SM and SAD, the *DSM-5* criteria still largely depend on adequate cognitive function and communication, which makes anxiety disorders difficult to diagnose in individuals with more severe ID (Cooray et al., 2016). While the *DM-ID-2* might be useful for adults with mild to moderate ID in that its use helped to avoid the OSAD category, resulting in more specific diagnosis, Cooray et al. (2016) suggest the use of behavioral equivalents for diagnosing anxiety in adults with severe or profound ID and point out that there are no studies looking at utility of *DSM* criteria for anxiety disorders in children with ID.

Although using behavioral equivalents may be helpful in identifying fear or anxiety in individuals with ID, it is important to note that it still may be difficult to differentiate anxiety from avoidance or challenging behavior in individuals with ID.

For example, Khreim and Mikkelsen (1997) observed that fear in adults with ID may manifest itself as "agitation, screaming, crying, withdrawal, freezing, or regressive clingy behavior." Although these behaviors or other behaviors might indicate fear or anxiety in an individual with ID at certain times in certain contexts, that same individual might also cry, scream, withdraw, cling, become tense, or behave irritably because he is tired, in pain, angry, sad, overstimulated, or otherwise distressed or because he dislikes an object, person, or situation (or simply prefers another situation). As such, Hagopian and Jennett (2014) differentiated between "simple avoidance" in which the individual avoids non-preferred stimuli or situations (such as a chore or academic task) and "anxious avoidance" in which the individual exhibits avoidant behavior accompanied by traditional symptoms of anxiety, including facial expressions of fear, increased physiological arousal, and, if possible, self-reported anxiety. Along these lines, Moskowitz et al. (2013) developed a multimethod assessment strategy to operationally define the construct of "anxiety" in children with ASD and ID. They assessed (1) the *behavioral* component of anxiety by collecting data on idiosyncratic behaviors that were reported to indicate anxiety for each child, (2) the *physiological* component of anxiety by collecting data on heart rate and respiratory sinus arrhythmia, and (3) the *cognitive/affective* or "subjective" component of anxiety by assessing the context in which the behaviors and physiological arousal occurred, given that labeling one's state of affective arousal as "anxiety" or any other emotion is highly influenced by the situational context in which the arousal occurs (Bandura, 1988). When multiple sources are taken together, converging data may support that the behavior is indicative of anxiety.

Intervention for Fear and Anxiety in ID

Goals for Intervention

From a diagnostic standpoint, sometimes it may be difficult to discriminate whether an individual with ID is running away from a blender while crying and covering his ears because he is anxious/afraid of the blender or because he is hypersensitive to auditory stimuli (e.g., Koegel et al., 2004). However, from a pragmatic behavioral standpoint, one could argue that it may not matter. That is, regardless of whether running away from the blender is due to a specific phobia of blenders or hyperacusis, our intervention goals would likely be the same: to encourage the individual to approach rather than avoid the given stimulus (e.g., the blender), to engage in the formerly avoided activity, or at the very least to calmly tolerate that stimulus or situation without aggression, self-injury, tantrums, or other problem behaviors.

Review of Evidence-Based Treatments

Although about a quarter of intervention studies that treat individuals with ID or cognitive delay mentioned specific *DSM* anxiety disorders (e.g., SoP, SM, GAD; Suveg et al., 2006), most studies that treat fear or anxiety in individuals with ID do not apply *DSM* criteria or other formal diagnostic criteria to establish the presence of anxiety disorders in these individuals. Rather, most intervention studies treating fear or anxiety in individuals with ID rely on overt avoidance behavior (i.e., avoidance of a phobic stimulus), verbalizations or vocalizations of fear, and other behavioral indicators of fear or anxiety (e.g., crying, screaming, running away, shaking, wide eyes), from which the presence of anxiety may be inferred. Therefore, we will review the literature on treating anxiety and/or "phobic avoidance" more broadly in individuals with ID, rather than restricting our focus to the treatment of specific *DSM* anxiety disorders.

Additionally, similar to other reviews (e.g., Rosen, Connell, & Kerns, 2016), we reviewed intervention studies that target individuals with impaired intellectual, adaptive, and/or developmental abilities even when standardized IQ testing was unavailable. This is because, in some studies, participants were untestable on standardized IQ tests or because, in some older behavior analytic studies, standardized measures of IQ or adaptive functioning were likely not seen as vital to describe participants' functioning. Regardless, with the new *DSM-5*, there is an increased emphasis on adaptive functioning over IQ, given that it is adaptive functioning that determines the level of supports required. With these considerations in mind, we will now review the evidence on interventions that address anxiety or phobic avoidance in individuals with ID.

In Vivo Exposure. The power of stimuli to evoke fear reactions can be reduced by exposing individuals to their feared stimuli or situations, known as in vivo exposure, which is typically the most critical component of treatment for any anxiety disorder. Most researchers would agree that engaging in exposure is necessary for positive treatment outcome when treating anxiety (Kendall et al., 2005). Indeed, 76% of the intervention studies that targeted fear or anxiety in individuals with ID used in vivo exposure (see Tables 44.1 and 44.2). This exposure is typically done in a gradual fashion, also known as "graduated exposure" or "in vivo desensitization" or "contact desensitization," in which brave behavior (i.e., approach/acceptance response) is positively reinforced, while fearful/avoidant responses are extinguished, as the individual is gradually exposed to increasing proximity, intensity, or amounts of the feared stimulus or situation. These exposures are typically conducted in a hierarchical fashion from situations that are rated as less anxiety-provoking to those rated as more anxiety-provoking. For example, in Moskowitz et al. (2017), Jon was exposed to people singing happy birthday, progressing from a cake with the candles unlit to a cake with the candles lit, and gradually increasing proximity to the cake.

Contingent Reinforcement. This refers to providing the individual with some type of positive reinforcement, such as social reinforcement (e.g., praise) and/or tangible reinforcement (e.g., toy, tokens to be exchanged for desired item), contingent upon approaching the feared stimulus or engaging in a feared activity/task, or contingent on remaining in a feared situation without resisting or engaging in problem behavior. Most of the intervention studies (68%) that target fear or anxiety in individuals with ID involved some type of reinforcement for approach or completing an exposure. For example, in Love, Matson, and West (1990), the therapist praised the child and dispensed positive reinforcement (e.g., verbal praise, sticker, stuffed dog toys, Gummi Bears) to the child for his successful partial approach toward the feared stimulus. Of note, contingent positive reinforcement includes both differential reinforcement of alternative behavior (DRA), in which approach behavior or compliance is reinforced (e.g., sitting still during blood draw; Hagopian et al., 2001), and differential reinforcement of other behavior (DRO), in which the absence of escape/avoidance behavior is reinforced.

Hierarchy/Shaping/Stimulus Fading. A hierarchy refers to breaking down the task of approaching or engaging with the feared stimulus or situation into small steps, with gradual increases in response requirements or successive approximations toward the final goal/step. Steps can be broken down sequentially into a task analysis (as in the steps in the hierarchy of a dental examination; e.g., sit in dental chair, and lean back in dental chair, Luscre & Center, 1996), or steps can include time exposed to the stimulus, amount of the stimulus, size of the stimulus (e.g., increasingly larger dogs; Erfanian & Miltenberger, 1990), intensity of the stimulus (e.g., increasing volume of audio recordings of thunder), or distance to the stimulus (e.g., decreasing distance between the needle and the tip of the child's finger, i.e., stimulus fading; Shabani & Fisher, 2006). Over half of the intervention studies (56%) that target fear or anxiety in individuals with ID involved hierarchy/shaping/stimulus fading. To illustrate, in Koegel et al. (2004), Steps 1–2 of the hierarchy for one child with a fear of vacuums involved placing a turned-off vacuum in the child's environment to expose him to seeing the feared stimulus, Steps 3–12 exposed him to an out-of-sight vacuum that became increasingly louder as it was moved closer to the child, and Steps 13–14 exposed the child to both seeing and hearing the vacuum. In most studies, progression to the next step of the hierarchy is typically contingent on a certain number of successfully

Table 44.1 Treatment studies summary

Study	*N*	Participant characteristics (age, diagnoses, functioning)	Anxiety disorder or feared stimuli or presenting problem	Dependent variables/ outcome measures	Study design	Intervention	Results/primary findings
Bouvet and Coulet (2016)	30	Adults with mild to moderate ID (15 in relaxation group, 15 in waitlist control group)	General anxiety management	Anxiety (state-trait anxiety inventory [STAI-Y]), self-esteem, emotional regulation	Randomized controlled trial	Relaxation therapy	Significantly reduced anxiety and improved self-esteem and cognitive reappraisal in relaxation group vs. control group
Burgio, Willis, and Burgio (1986)	1	21yo male with severe ID	Stair avoidance	Percentage of staff requests complied with for stair ascent and descent on staircases of varying lengths	Multiple baseline design across settings; within settings, a withdrawal of intervention (ABAB) was also used	Prompting, contingent reinforcement (praise and edibles), and physical guidance	Treatment was effective in increasing stair use in both settings (5 and 11 stairs)
Chok et al. (2010)	1	15yo male with ASD, moderate ID	Specific phobia, animal type (dogs)	Distance from dog, presence of behavioral escape/avoidance, heart rate	Case study (AB design)	Reinforced practice (graduated exposure, positive reinforcement), hierarchy, prompting	Approached and pet 4 dogs without elevated heart rate; maintained at 6-month follow-up
Conyers et al. (2004)	6	3 male and 3 female adult subjects (ages 33–54) with profound or severe ID	Fear of dental procedures Crying, yelling, hitting, biting, grabbing, or refusing to enter dentist's office	Participants' compliance with an 18-step task analysis associated with a dental examination	Multiple baseline design across participants; 3 received desensitization, and 3 received video modeling; crossover to other treatment for participants that did not complete task analysis with first treatment	In vivo desensitization (including verbal and physical prompts) and video modeling	Desensitization increased compliance with dental procedures for all 5 participants; video modeling increased compliance for 1 of 3 participants

Davis, Kurtz, Gardner, and Carman (2007)	1	7yo male with developmental delays (IQ or adaptive behavior not reported) and severe behavior problems	Phobia of water and heights	Behavioral avoidance, specific phobia symptoms (anxiety disorders interview schedule-IV-parent [ADIS-IV-P]), subjective fear (fear survey schedule for children-revised [FSSC-R]), negative vocalizations	Multiple baseline design across water and height phobias	CBT (in vivo exposure, participant modeling, cognitive challenges to catastrophic beliefs, reinforcement); one-session treatment for each phobia	Reduced or eliminated behavioral avoidance, specific phobia symptoms, and subjective fear; reduced negative vocalizations in height exposure
Dixon and Gunary (1986)	9	Adults with ID (ages 32–62)	Phobia of dogs	Ability to cope with dogs	Pre-post design	Gradual desensitization (exposure to 4 different dogs), video and modeling procedures	Majority showed an increased ability to cope with dogs, maintained after 9 months for those who practiced
Efranian and Miltenberger (1990)	2	39yo male with moderate ID 18yo male with profound ID	Phobia of dogs	Avoidance and approach behavior	Multiple baseline design across participants	Contact desensitization	Elimination of avoidance behavior in both adults; behavior generalized to outdoor setting and maintained at 2-month follow-up
Florez and Bethay (2017)	1	28yo female with mild ID, GAD, and intermittent explosive disorder	Challenging behaviors, emotional dysregulation, and GAD	Frequency of challenging behavior	Case study	Adapted DBT (individual therapy and social skills training)	Decrease in challenging behaviors; maintained average of <1 challenging behavior per month during year after treatment
Hagopian, Crockett, and Keeney (2001)	1	19yo male with moderate ID, intermittent explosive disorder, and cerebral palsy	Blood-injury-injection phobia (BIIP)	Refusals (verbal and physical), inappropriate behavior (aggression and disruption), compliance	Changing criterion design	Fading (graduated exposure), modeling, noncontingent and differential reinforcement, presession anxiolytic medication, and topical analgesic cream	During final two sessions, participant sat unrestrained in chair and complied with having blood drawn

(continued)

Table 44.1 (continued)

Study	*N*	Participant characteristics (age, diagnoses, functioning)	Anxiety disorder or feared stimuli or presenting problem	Dependent variables/ outcome measures	Study design	Intervention	Results/primary findings
Hall et al. (2013)	7	Adults with learning disabilities	Anxiety and emotion regulation	Anxiety, depression, mindfulness, caretaker component	Qualitative and repeated measures design	Group DBT skills training	Treatment successfully reduced anxiety and depression and increased the use of mindfulness skills
Idusohan-Moizer, Sawicka, Dendle, and Albany (2015)	15	Adults with borderline, mild, or moderate ID and anxiety and/or depression (ages 21–44)	Anxiety, depression, and self-compassion	Anxiety and depression (hospital anxiety and depression scale), self-compassion and compassion for others (the compassion scale)	Pre-post design	Mindfulness-based cognitive therapy (MBCT)	Improvement in anxiety, depression, self-compassions, and compassion for others, maintained at 6-week follow-up
Koegel, Openden, and Koegel (2004)	3	3yo female with ASD (VABS = 1y7m) 2yo male with ASD (VABS = 2y1m) 2yo with ASD (VABS = 1y5m)	Fear of auditory stimuli (vacuum cleaner, blender, hand mixer, toilet flushing, and animal sounds)	(a) Number of hierarchical steps completed with the child's anxiety level rated as comfortable per week, (b) mean level of anxiety per session	Changing criterion design for each child, clinical replication for all 3 children, and multiple baseline across stimuli design for 1 child	Systematic desensitization implemented by teachers or parents with clinician assistance or directly by clinician	Number of steps completed in hierarchy increased Anxiety level decreased from intolerable to comfortable level
Lindsay (1999)	15	Adults with mild/ moderate ID	General anxiety management	Simplified versions of standardized self-report anxiety instruments Intensity and duration of maladaptive cognitions	Quantitative case series	CBT was simplified and adapted but included essential elements of Beck et al.'s (1979) cognitive therapy (e.g., challenging negative automatic thoughts, reviewing evidence for/ against dysfunctional beliefs, role-playing more positive ways of thinking)	Large and significant decreases in scores on standard measures and maladaptive cognitions; maintained at 6-month follow-up
Lindsay and Baty (1986)	3	3 adults with ID	General anxiety management	Anxiety levels	Case studies	Behavioral relaxation training	Treatment successful in reducing anxiety levels

Lindsay and Baty (1989)	20	Adults with moderate and severe ID (ages 25–69; IQ < 55)	General anxiety management	Anxiety score (composed of 10 behavioral ratings) and pulse rate	Randomly allocated to 2 groups: Abbreviated progressive relaxation (APR) or behavioral relaxation training (BRT)	Abbreviated progressive relaxation (APR) or behavioral relaxation training (BR)	While both relaxation procedures produced improvement, BRT was quick, consistent, and significantly better than APR; no differences in pulse rate
Lindsay, Baty, Michie and Richardson (1989)	50	Adults with moderate to severe ID (ages 25–69; IQ 30–55)	General anxiety management	Anxiety score (composed of 10 behavioral ratings) and pulse rate	Randomly allocated to 5 groups: Individual behavioral relaxation training (BRT), individual abbreviated progressive relaxation training (APR), group BRT, group APR, or control	Abbreviated progressive relaxation (APR, focusing on internal states of muscular tension and release) or behavioral relaxation training (BRT; copy relaxed behaviors)	Though both BRT and APR improved more than control group, BRT resulted in lower levels of rated anxiety than APR; no significant differences in pulse rate
Lindsay, Michie, Baty, and McKenzie (1988)	2	Women with ID	Phobia of dogs	Overall ratings of fear, number of positive approaches to a dog, number of negative reactions to a dog, number of requests to remove a dog, and self-assessments of anxiety	Unknown	Anxiety management training, exposure, and modeling	Both women responded to treatment
Love, Matson, and West (1990)	2	4.5yo male with ASD (VABS = 1y10m) 6yo male with ASD (VABS = 1y8m)	Avoided going outside alone, avoided running shower water	(a) Number of approach steps completed, (b) verbalizations and vocalizations of fear, (c) appearance of fear	Multiple baseline design across participants	In vivo desensitization, modeling of approach steps, prompting approach response, positive reinforcement contingent upon approach	Increase in approach, decrease in fear verbalizations, and decrease in ratings of appearance of fear
Luscre and Center (1996)	3	9yo, 9yo, and 6yo males with ASD and severe ID	Fear of dental procedures	Completion of the steps in a dental exam (in vivo and in an analog dental office)	Multiple baseline design across participants	Desensitization with guided mastery, video peer modeling, and reinforcement	Successful completion of the steps in a dental exam and clinically significant increase in steps completed in vivo

(continued)

Table 44.1 (continued)

Study	*N*	Participant characteristics (age, diagnoses, functioning)	Anxiety disorder or feared stimuli or presenting problem	Dependent variables/ outcome measures	Study design	Intervention	Results/primary findings
Maguire, Lange, Scherling, and Grow (1996)	4	4 adult males (ages 28–40) with mild to severe ID	Dental procedure resistance	Aggression and noncompliance Target behaviors: Sit calmly in dental chair, be vocally quiet, keep hands in lap, keep mouth open, tolerate procedure	Multiple baseline design across participants	Increasing predictability (i.e., tell-show-do), monetary and social reinforcement, brief and frequent dental visits, increased client control	Significant increases in cooperativeness with dental procedures, maintained over several months with 1 booster session per month
Matson (1982)	3	3 adult males with mild ID	Obsessive-compulsive behaviors	Clothes and body checking for extended periods of time	Multiple baseline design across behaviors, settings, and targets	Differential reinforcement of incompatible behavior, overcorrection as response suppression	Perceived improvement in appropriateness of target behaviors and rapid reduction in target behaviors and self-ratings of anxiety
Matson (1981a)	3	8yo, 8yo, and 10yo females with moderate ID	Fear of talking to or being near unfamiliar adults	Approaching and talking to unfamiliar adults and child ratings of overall fear	Multiple baseline design across participants	Participant modeling by mother with gradual fading of physical/verbal prompts, contingent reinforcement (tangible and praise)	Treatment was effective at reducing fear, and gains were maintained at 6-month follow-up
Matson (1981b)	24	24 adults with mild to moderate ID (ages 25–45)	Fear of participating in community-based activities	Fear level of going into a grocery store	Pre-post design	Participant modeling, behavioral rehearsal, shaping, in vivo exposure, graduated hierarchy	Participant modeling was significantly more effective at reducing fear than no treatment
Moskowitz et al. (2017)	3	6yo, 8yo, and 9yo males in the low range of intellectual and/or adaptive functioning with ASD, anxiety, and behavior problems	Fear of happy birthday, fear of separation from parents, fear of left/right turns in car	Anxious behaviors, problem behaviors, heart rate, respiratory sinus arrhythmia (RSA), ratings of appearance of anxiety	Multiple baseline design across participants	Graduated exposure, psychoeducation, cognitive restructuring, choices, counterconditioning, incorporating perseverative interests, prompting, positive reinforcement, extinction	Decreases in anxiety and problem behaviors and increases in RSA in anxiety-provoking situations

Neil, Vause, Jaksic, and Feldman (2017)	1	11yo male with ASD and IQ in borderline range	Ripping and sniffing paper, counting his fingers in a specific order	Obsessive-compulsive behaviors (OCBs) (repetitive behavior scale-revised [RBS-R], children's Yale-Brown obsessive-compulsive scale [CY-BOCS], child obsessive-compulsive impact scale-revised [COIS-RP])	Multiple baseline design across behaviors	Functional behavior-based CBT: CBT (e.g., psychoeducation, skills training, exposure and response prevention) plus FBA and FB intervention	Clinically significant decreases in OCBs, increase in psychosocial functioning, and high consumer satisfaction
Rapp, Vollmer, and Hovanetz (2005)	1	14yo female with ASD and severe ID	Swimming pool avoidance	Elopement, flopping, face hitting, choking, screaming, depth of water at which most time was spent	Within-subject design including multiple reversals	Blocking for flopping and elopement; reinforcement for movement toward pool; exposure to varying water depths	Eliminated pool avoidance and other disruptive behaviors
Riccardi, Luiselli, and Camare (2006)	1	8yo male with ASD and language and cognitive delays	Specific phobia of animatronic objects	Approach and time spent at a specified distance from the feared stimulus	Changing criterion design	Contact desensitization (reinforcing approach responses), shaping	Able to encounter previously avoided stimuli
Runyan, Stevens, and Reeves (1985)	3	1 adult male and 2 adult females (ages 33–41) with profound to severe ID	Escalator avoidance	Avoidance behavior toward escalators	Multiple baseline design across participants	Contact desensitization and contingent social approval	Reduced avoidance behavior toward escalators
Schmidt, Luiselli, Rue, and Whalley (2013)	1	16yo male with ASD and severe ID	Setting and activity avoidance of gymnasium, music room, and gross-motor exercise room	Approach distance or duration in setting before displaying problem behaviors Participation in activities within each setting	Multiple baseline design across settings and changing criterion design	Graduated exposure and positive reinforcement	Able to enter settings without resistance and participate in activities within each setting
Shabani and Fisher (2006)	1	18yo male with ASD, ID, and type 2 diabetes	Phobia of needles	Percentage of successful blood draw trials	ABAB reversal design	Stimulus fading and differential reinforcement	Successful in obtaining daily blood samples

(continued)

Table 44.1 (continued)

Study	*N*	Participant characteristics (age, diagnoses, functioning)	Anxiety disorder or feared stimuli or presenting problem	Dependent variables/ outcome measures	Study design	Intervention	Results/primary findings
Suveg, Comer, Furr, and Kendall (2006)	1	8yo female with cognitive delays (IQ = 78; borderline)	Social phobia (SoP), selective mutism (SM), generalized anxiety disorder (GAD)	Anxiety symptoms (anxiety disorders interview schedule for children [ADIS-C/P-IV], state-trait anxiety scale for children [STAIC-P], child behavior checklist for children [CBCL], teacher rating form [TRF], coping questionnaire [CQ])	Pre-post design	CBT (e.g., imaginal and in vivo exposure, modeling, role-play, reinforcement, relaxation)	Significantly reduced anxiety symptoms; participant no longer met criteria for GAD or SM
Waranch, Iwata, Wohl, and Nidiffer (1981)	1	21yo male with Down's syndrome and mild ID	Phobia of mannequins	Approach responses	Multiple baseline design across mannequins	Shaping (mannequin gradually pulled closer), verbal prompts to touch mannequin, reinforcement	100% approach responses to different sized mannequins; effects generalized and maintained at 6-month follow-up
Wolff and Symons (2013)	1	41yo male with ASD, ID, and a history of medical noncompliance	Phobia of needles	Compliance with needle-to-skin contact	Changing criterion design	Exposure, differential reinforcement, and safety signal	Compliance with needles was achieved and maintained at follow-up; differential reinforcement and safety signal added value
Wright (2013)	1	19yo male with ASD, ID, and social anxiety	Social anxiety	Idiosyncratic measure of anxiety Global anxiety symptoms (brief symptom inventory [BSI])	A-B design	CBT (psychoeducation, relaxation, cognitive restructuring)	Anxiety severity significantly reduced, reaching and maintaining zero; BSI showed reduction posttreatment

Note. Studies highlighted in blue were included in calculations of empirically supported treatments (EST) according to Chambless and Hollon (1998) criteria because they were either a randomized control trial (RCT) or a single-case experiment with at least three participants; case studies, pre-post designs, or single-case experiments with fewer than three participants were not included in EST calculations

Table 44.2 Treatment components by study

Article	Treatment components												
	In vivo/ exposure	Hierarchy	Contingent reinforcement	Prompting	Modeling	Extinction/ blocking	Noncontingent reinforcement	Increasing predictability	Providing choices	Perseverative interests	CBT	Relaxation	Acceptance-based
Bouvet and Coulet (2016)												X	
Burgio, Willis, and Burgio (1986)	X		X	X	X								
Chok et al. (2010)	X	X	X	X									
Conyers et al. (2004)	X	X	X	X	X[a]			X					
Davis, Kurtz, Gardner, and Carman (2007)	X	X	X	X	X						X		
Dixon and Gunary (1986)	X	X			X[a]						X		
Efranian and Miltenberger (1990)	X	X	X	X	X		X						
Florez and Bethay (2017)			X	X	X								X
Hagopian, Crockett, and Keeney (2001)	X	X	X		X		X						

(continued)

Table 44.2 (continued)

Article	Treatment components												
	In vivo/ exposure	Hierarchy	Contingent reinforcement	Prompting	Modeling	Extinction/ blocking	Noncontingent reinforcement	Increasing predictability	Providing choices	Perseverative interests	CBT	Relaxation	Acceptance-based
Hall et al. (2013)													
Idusohan-Moizer, Sawicka, Dendle, and Albany (2015)													X
Koegel, Openden, and Koegel (2004)	X	X					X						
Lindsay (1999)	X										X	X	
Lindsay and Baty (1986)												X	
Lindsay, Baty, Michie, and Richardson (1989)												X	
Lindsay and Baty (1989)												X	
Lindsay, Michie, Baty, McKenzie (1988)	X	X			X							X	
Love, Matson, and West (1990)	X	X	X	X	X								

Luscre and Center (1996)	X	X	X	X	X[a]		X						
Maguire, Lange, Scherling, and Grow (1996)	X		X				X	X	X				
Matson (1982)	X		X		X								
Matson (1981a)	X	X	X	X	X								
Matson (1981b)	X		X	X	X								
Moskowitz et al. (2017)	X	X	X	X	X[a]	X	X	X	X	X	X		
Neil, Vause, Jaksic, and Feldman (2017)	X		X			X				X	X		
Rapp, Vollmer, and Hovanetz (2005)	X		X	X		X							
Riccardi, Luiselli, and Camare (2006)	X	X	X				X						
Runyan, Stevens, and Reeves (1985)	X	X	X	X	X								
Schmidt, Luiselli, Rue, and Whalley (2013)	X	X	X	X	X	X							

(continued)

Table 44.2 (continued)

Article	Treatment components												
	In vivo/ exposure	Hierarchy	Contingent reinforcement	Prompting	Modeling	Extinction/ blocking	Noncontingent reinforcement	Increasing predictability	Providing choices	Perseverative interests	CBT	Relaxation	Acceptance-based
Shabani and Fisher (2006)	X	X	X	X									
Suveg, Comer, Furr, and Kendall (2006)	X	X	X		X						X	X	
Waranch, Iwata, Wohl, and Nidiffer (1981)	X	X	X	X			X						
Wolff and Symons (2013)	X	X	X	X			X	X					
Wright (2013)	X										X	X	
Total %	76%	56%	68%	50%	50%	12%	26%	12%	6%	6%	21%	24%	6%

[a] Denotes use of video modeling

completed trials of the previous step. Of note, although this treatment component overlaps with the previous component of gradual exposure, this component refers to creating the steps in the fear/avoidance hierarchy, whereas gradual exposure refers to progressively moving the individual through each step in that hierarchy.

Prompting. Prompting is the presentation of any antecedent stimulus – a physical, verbal, or sensory stimulus – to initiate a response. Hagopian and Jennett (2008) suggested including prompting as a way to help the individual to comply with the steps of the exposure hierarchy, which may be especially important when the individual is displaying highly intense anxiety behaviors or not approximating the approach response. Half of the intervention studies (50%) that target fear or anxiety in individuals with ID involved assisting the individual to approach or engage with the feared stimulus using verbal prompts (e.g., mother verbally prompting her child to speak to a stranger; Matson, 1981a, 1981b) or physical prompts (e.g., physically guiding an adolescent to enter a feared setting at school; Schmidt et al., 2013). To illustrate, Runyan et al. (1985) used a least-to-most prompting sequence to help three adults with ID complete the steps of the hierarchy to ride an escalator. The prompting sequence involved the trainer modeling the desired behavior (e.g., approaching to within 1.5 m of escalator), verbally prompting the adult to complete the behavior, physically prompting him/her if necessary (i.e., light physical contact, such as holding hand or touching elbow), and, finally, manually guiding the adult through the behavior if necessary (i.e., physically moving his/her body through the desired behavior, which was stopped if the adult said "no" or moved away). One note of caution is that prompting should be used to *teach* approach responses rather than *force* them, as this can lead to negative emotions and problem behavior (Hagopian, Lilly, & Davis, 2017).

Modeling. With modeling, a therapist, parent, or peer demonstrates engaging in the anxiety-provoking situation or engaging with the feared stimulus appropriately while the participant observes. This can include the model demonstrating a step in the hierarchy and/or receiving reinforcement for brave behavior. Half of the studies (50%) that treat fear or anxiety in individuals with ID involve modeling. For example, in treating fear of grocery shopping in Matson (1981a, 1981b), participants were shown how to perform shopping behaviors, with the therapist first walking through the steps of the task. While some studies employ in vivo or live modeling (as in Matson, 1981a, 1981b), other studies use video modeling. For example, in Luscre and Center's (1996) study treating dental fear in children with autism and ID, four typically developing children acted as the peer models in a video of a dental exam conducted in the dentist's office. As another example of video modeling, in Moskowitz et al. (2017), a child with ASD and ID who was afraid of "happy birthday" viewed several internet videos of his favorite Sesame Street characters singing happy birthday and blowing out candles on a birthday cake.

Noncontingent Reinforcement/Counterconditioning. This intervention involves providing the person with noncontingent access to preferred or "anti-anxiety" stimuli before or during exposure to the feared stimulus/situation in order to inhibit or compete with a fear response. Only 26% of the intervention studies that target fear or anxiety in individuals with ID used this approach. In illustration, upon presentation of the birthday cake accompanied by the "happy birthday" song (feared stimuli), Jon was immediately given his most highly preferred item, a Sesame Street pop-up toy, without him having to perform any action to receive the toy (Moskowitz et al., 2017). This antecedent-based strategy of pairing anxiety-producing stimuli with positive stimuli *before* or during the exposure stands in contrast to the consequence-based strategy of positive reinforcement contingent upon the person displaying a certain behavior or completing a certain task (e.g., reinforcing Jon only after he attempts to blow out the candle). Related to noncontingent reinforcement, some studies provided individuals with noncontingent access to stimuli to help the individual relax or provide a distraction, such as allowing individuals to hold a coin purse or small bottle to keep their hands still during dental examinations (Maguire et al., 1996). Although it is not clear whether stimuli such as purses or bottles were reinforcing and could therefore be classified as noncontingent reinforcement, we will include these studies that provided noncontingent access to distracting stimuli in this category of noncontingent reinforcement.

This strategy of noncontingent reinforcement can also be conceptualized as counterconditioning, in which the conditioned stimulus (CS) is repeatedly paired with an unconditioned stimulus (US) having a valence opposite from the original US that was used for acquisition. Thus, counterconditioning involves presenting an item or activity that produces a relaxed, positive state that is incompatible with anxiety, such as pairing exposure to mannequins (Waranch et al., 1981) and dogs (Erfanian and Miltenberger 1990) with playing checkers or cards. Similarly, Luscre and Center (1996) used anti-anxiety stimuli (e.g., hand-held mirror, Play-Doh, the song "Achy Breaky Heart") to help counter a fear response to dental examinations; these anti-anxiety stimuli were paired with presentation of dental procedures and were individualized for each child. Although counterconditioning has not proven more effective than standard exposure (repeated presentation of the CS in the absence of the US) in the treatment of fear/anxiety, it should be noted that, in Moskowitz et al. (2017), the participants' anxious behaviors did not decrease throughout the repeated exposures of the baseline sessions. Thus, we decided to pair the anxiety-provoking situation (the CS) with a highly preferred and otherwise inaccessible positive reinforcer (e.g., pairing birthday cake/song with Sesame Street toy), rather than just exposing the participants to the anxiety-provoking situation without any accompanying positive stimuli.

Incorporating the Individual's Perseverative Interests. Given the level of anxiety that the targeted contexts appeared to provoke in three children with ASD and ID, Moskowitz et al. (2017) paired the anxiety-provoking situation with stimuli/activities that were not only reinforcing but taking it one step further by incorporating perseverative items or activities that were *the most* highly salient/preferred in an attempt to override or counteract the aversiveness of the anxiety-provoking contexts. "Perseverative interest" (also called "circumscribed interest" or "special interest" or "obsessive interest") refers to an object, activity, or topic with which the individual is intensely preoccupied. Studies have shown that using obsessions as reinforcers are more effective at reducing problem behaviors than other reinforcers such as edible reinforcers (Charlop-Christy & Haymes, 1996; Charlop, Kurtz, & Casey, 1990). It is possible that this could be because of the anxiety-reducing properties of a person's idiosyncratic perseverative, stimulatory, or obsessive reinforcers, in contrast to other tangible or social reinforcers, although – once again – there is a lack of research in relation to anxiety. To illustrate, in Moskowitz et al. (2017), Sam's parents used his primary perseverative interest (Dr. Seuss) as the reinforcer to pair with riding in the car (counterconditioning). Although Sam's favorite book, *The Sneetches* by Dr. Seuss, was an item he owned before intervention and could access at home, audio recordings of this book and other Dr. Seuss books were created at the start of intervention; Sam was only allowed to listen to these recordings during car rides. Of note, although incorporation of children's "special interests" is a part of treatment protocols for youth with ASD (e.g., Reaven et al., 2012), with the exception of Moskowitz et al. (2017) and Neil et al. (2017), these treatments have only included youth with "high-functioning" ASD (without ID).

Increasing Predictability **(e.g., safety signal, visual schedule, social story, priming).** Research has demonstrated that problem behavior is lower in predictable than in unpredictable conditions for individuals with ID (e.g., Flannery & Horner, 1994). Although Flannery and Horner (1994) did not conceptualize the reduction in problem behavior as being due to a reduction in "anxiety" per se, startle studies suggest that the ability to predict aversive events attenuates anxious responses (Grillon, 2008). However, only 12% of studies that targeted fear or anxiety in individuals with ID used some strategy designed to increase predictability.

One way to increase predictability is to provide an advanced warning to signal upcoming transitions or changes in routine or aversive events, which can provide the individual with time to prepare for the upcoming aversive situation or can signal how much time is left until an aversive activity will be over. These advanced warnings can be verbal and/or visual, including countdowns (e.g., "You have 10 seconds left until the blood test is finished… 10, 9, 8, 7, 6, 5, 4, 3, 2, 1, it's finished"), picture prompts (Mace, Shapiro & Mace, 1998), visual schedules (Dettmer, Simpson, Myles, & Ganz, 2000), and visual representations of time such as a time timer (displaying section of red indicating the given amount of time, with the red disappearing as the time runs out) (Dettmer et al., 2000). Although almost all of these studies that used advanced warnings did not mention fear or anxiety and only mentioned "aversive" events such as transitions, transition-/change-related anxiety is increasingly recognized as anxiety in individuals with ASD (e.g., Kerns et al., 2014, 2016), many of whom have co-occurring ID. To illustrate the use of advanced warnings to treat a traditional anxiety disorder, after three unsuccessful trials of attempting to treat a needle phobia with stimulus fading plus DRA, Wolff and Symons (2013) added a "safety signal" (i.e., a timer) to convey that the trial was of a finite duration and show when the trial would end. The addition of the safety signal corresponded with a change in the participant's behavior (increased compliance, reduced agitation) (Wolff & Symons, 2013). Further, even after the safety signal was later removed, the participant continued to keep his arm on the table for the duration of the needle exposure, suggesting he may have learned that all trials, with or without the safety signal, were of a limited duration.

In addition to advanced warnings such as timers and visual schedules, Social Stories (Gray & Garand, 1993) are individualized narratives that visually depict the sequence of events involved in a routine or situation and describe appropriate behavior relevant to the situation, which may decrease unpredictability and provide a model for non-anxious or coping behavior. For example, for a boy with ASD and ID who exhibited separation anxiety, Ben's Social Story normalized anxiety, explained that anxiety has a function or purpose, explained the concept of habituation (e.g., "I will see that my anxiety goes down after a while, even when Mommy and Daddy are not home"), and provided Ben with replacement behaviors that he could do instead of crying, screaming, and trying to run after them (i.e., coping self-talk such as "This is just my anxiety talking," relaxation techniques such as deep breathing, and fun activities such as playing or watching a movie) (Moskowitz et al., 2017). Although Social Stories have been shown to decrease levels of problem behavior in individuals with ASD and ID (Kokina & Kern, 2010), with the exception of Moskowitz et al. (2017), no other Social Stories studies have targeted or mentioned fear or anxiety specifically. A recent meta-analysis of Social Stories for students with ASD revealed that Social Stories are more effective when addressing inappropriate behaviors than when teaching social skills and when participants had lower cognitive ability rather than high or average intelligence (Kokina & Kern, 2010), which suggests that Social Stories could be helpful in treating anxiety or anxiety-related behaviors in individuals with ID (particularly anxiety that is related to unpredictability), although more research is necessary to support this.

Finally, another way to increase predictability by manipulating antecedent events is through priming, in which an individual previews future events so that they become more predictable. The use of video also allows for priming with children who are nonverbal or limited in their ability to comprehend verbal descriptions. Similar to advanced warnings, visual schedules, and Social Stories, the research on priming (Wilde, Koegel, & Koegel, 1992) and video priming (Schreibman, Whalen, & Stahmer, 2000) has shown that it decreases problem behavior associated with transitions or an upcoming unpredictable situation, but no studies except for Moskowitz et al. (2017) have mentioned treating fear or anxiety using priming.

In illustration, Sam, who had a fear of left/right turns, first watched videos of cars making turns on YouTube before exposure to turns while riding in the car (Moskowitz et al., 2017).

Extinction/Blocking. Escape extinction refers to not allowing the person to escape the anxiety-provoking situation or stop performing the given step in the hierarchy in response to problem behaviors or anxious/avoidance behaviors (so that these behaviors no longer produce reinforcement in the form of escape). This generally involves physically guiding the individual to engage in a step in the hierarchy (preventing him from escaping) or, in some cases, even blocking the individual from escaping. Only 12% of the intervention studies that target fear or anxiety in individuals with ID involved extinction or blocking. For example, in Rapp, Vollmer, and Hovanetz (2005), two therapists blocked or prevented Amy's attempts to elope from the swimming pool or flop to the ground by prompting her to sit in a rolling chair. In other cases, extinction does not involve response blocking but simply involves continuing with the anxiety-provoking situation regardless of the child's problem behavior or anxious behavior. To illustrate, in Moskowitz et al. (2017), when Sam was riding in the car with his parents during baseline, his parents often altered their driving route in response to his anxious behavior (e.g., crying "no left!" or "drive straight!") to avoid his feared stimulus of left/right turns, thus negatively reinforcing his anxious behavior. Thus, one component of intervention involved extinction, in which Sam's anxious behavior was no longer reinforced by escape through terminating the task (driving back home) or altering the driving route to accommodate Sam's anxiety (i.e., driving straight instead of turning right/left).

Providing Choices/Control. Allowing individuals to choose activities and reinforcers has been shown to increase task engagement while minimizing escape-motivated problem behaviors (Shogren, Faggella-Luby, Jik Bae, and Wehmeyer, 2004). Only 6% of the intervention studies that target fear or anxiety in individuals with ID involved providing individuals with ID with increased control or choices. To illustrate increased control, Maguire et al. (1996) asked clients with ID when they were ready to start at the beginning of each dental session and after each break. Similarly, to illustrate providing choices, in Moskowitz et al. (2017), Sam was given the opportunity to choose several preferred items (e.g., balls, books) to pack in a "car bag" that he could bring with him while riding in the car. In addition, whenever possible, Sam's parents attempted to provide him with a choice of the location to which they would drive. Although the existing research on providing choices/control to individuals with ID only mentions escape-motivated problem behaviors and (with the exception of Moskowitz et al. (2017)) does not mention fear or anxiety, researchers in the field of anxiety disorders have suggested that a sense of unpredictability and uncontrollability is at the heart of anxiety and that the development of coping responses that impart a sense of control can buffer anxiety (Barlow, 2000). Given that problem behavior can possibly be a way of exerting control over one's life (Wehmeyer, 1999), providing opportunities for choice-making may provide individuals with ID – especially those who are anxious – with an increased sense of control over their environments (Dattilo & Rusch, 1985), possibly reducing problem behavior that functions to escape/avoid anxiety, although this hypothesis remains to be tested.

Cognitive Behavioral Therapy (CBT). CBT typically includes not only graduated exposure to feared situations and stimuli (see above) but also psychoeducation (providing corrective information about fear and anxiety), cognitive restructuring (identifying maladaptive thoughts and teaching coping-focused thinking), somatic management (targeting autonomic arousal and physiological responses; e.g., relaxation), and relapse prevention (Albano & Kendall, 2002). CBT is widely considered to be the most effective evidence-based treatment for both neurotypical children and adults with anxiety disorders and has also demonstrated efficacy in reducing anxiety in youth with "high-functioning" ASD who do not have ID (Kerns, Renno, Storch, Kendall, & Wood, 2017). However, there is limited research on CBT to treat anxiety in individuals with ID. Only 21% of the studies that treat fear or anxiety in individuals with ID involved CBT. For the studies that used CBT to treat anxiety in children with borderline IQ or developmental delays, modifications included increased structure and emphasis on visuals, presenting concepts more concretely (e.g., robot versus ragdoll for relaxation), more practice opportunities to use concepts and skills, increased parental involvement, incorporation of the child's special interests, and, in some cases, a reduced focus on thoughts (Neil et al., 2017; Suveg et al., 2006). Although a systematic review of CBT for anxiety or depression in adults with ID (Unwin et al., 2016) did not identify any controlled trials for anxiety-focused CBT, they found equivocal evidence for the effectiveness of CBT in reducing anxiety in the three anxiety-focused CBT studies they located (Lindsay, 1999; Douglass et al., 2007; Marwood & Hewitt, 2012), with the latter two studies including participants with learning disabilities rather than ID. Of note, few studies in the review used standardized, objective criteria relating to a diagnosis or specific level of symptomatology. Further, the studies reviewed included only those with mild-moderate ID or learning disabilities and had inclusion criteria relating to verbal ability, which makes sense in light of the cognitive and verbal demands of cognitive restructuring.

Cognitive Restructuring. Cognitive restructuring (a component of CBT) involves identifying anxious thoughts or "self-talk"; challenging or disputing the accuracy, likelihood, or usefulness of those anxious thoughts; and replacing anxious self-talk with more accurate, probable, or productive thoughts that facilitate coping with anxiety-provoking situations rather than avoiding them (i.e., teaching coping self-talk). For example, in a case study of CBT for a man with ASD, ID, and social phobia, the man's negative automatic thoughts were identified, and the evidence for and against his thoughts was used to challenge his assumptions and develop more realistic alternative thoughts (Wright, 2013). When individuals have a cognitive deficit or are impaired in their ability to understand and/or express language, the cognitive components of CBT (i.e., cognitive restructuring, psychoeducation) are often downplayed, de-emphasized, simplified, modified, adapted to the individual's level, or most typically excluded altogether so that the intervention is more "behavioral" (e.g., exposure, reinforcement) rather than "cognitive behavioral." To illustrate, Moskowitz et al. (2017) used Social Stories as a way to provide psychoeducation (e.g., normalizing anxiety, explaining habituation) and a simplified form of cognitive restructuring in a visual modality that could be more easily comprehended. Although Moskowitz et al. (2017) did not address identifying or disputing anxious thoughts, which would have been too cognitively and verbally challenging, for Ben, cognitive restructuring took the form of prompting coping self-talk through the use of a Social Story. Coping self-talk is often used before exposures to help reduce anticipatory anxiety and/or during exposures to encourage brave/calm behavior rather than avoidance/anxious behavior. For example, Neil et al. (2017) taught a boy with ASD and ID to use coping statements (e.g., "I can do it, I'm not going to let OCD beat me!") to help himself refrain from ritualizing by ripping and sniffing. Similarly, in Moskowitz et al. (2017), Ben's Social Story provided him with coping statements that he could tell himself when he was feeling anxious, such as "I am going to fight my anxiety and I will win!" that appeared to help calm him when his parents left. However, it is unclear what role – if any – the cognitive elements played in the reduction of anxiety in these multicomponent treatments and whether the same results would have been achieved with the behavioral components (e.g., counterconditioning) alone. Thus, more research is needed on the use of the cognitive components of CBT (cognitive restructuring and psychoeducation) with individuals with ID.

Relaxation Training. Relaxation typically involves deep breathing and/or progressive muscle relaxation (PMR; Jacobson, 1938). Only 24% of the intervention studies that target fear or anxiety in individuals with ID involved relaxation. Although relaxation is often part of a wider CBT program for individuals with ID, Bouvet and Coulet (2016) examined the effect of relaxation on its own; they conducted an RCT comparing adults with mild to moderate ID who received PMR to those in a waitlist control group and found that relaxation significantly reduced state anxiety. Although some research (Luiselli, 1980) suggested that relaxation did not appear to be more effective than non-relaxation activities for individuals with ID, this could be because the individual was required to concentrate on internal states of muscle tension and relaxation, which may be conceptually challenging for individuals with ID. Lindsay, Baty, Michie, and Richardson (1989) found that individuals with moderate and severe ID who were randomly allocated to behavioral relaxation training (which focused on modeling/prompting of relaxed positions rather than subjective states of tension and relaxation) showed lower levels of rated anxiety than those who were allocated to abbreviated progressive muscle relaxation, suggesting the importance of focusing on observable behaviors to a greater degree than internal states in this population.

Acceptance-Based Interventions. The "third wave" CBT or acceptance-based interventions such as acceptance and commitment therapy (ACT), dialectical behavior therapy (DBT), and mindfulness-based cognitive therapy (MBCT) retain elements of traditional CBT (i.e., emphasizing the connection between thoughts, feelings, and behavior) but focus less on challenging thoughts and more on radical acceptance of those thoughts and oneself. Only 8% of studies that target fear or anxiety in individuals with ID used acceptance-based interventions.

MBCT combines elements of cognitive therapy with breathing meditations and yoga exercises to help increase the individual's awareness of the present moment and bodily sensations without attempting to get rid of those sensations. MBCT relies less on verbal exchange than traditional CBT, which could make it a good fit for individuals with ID. Idusohan-Moizer et al. (2015) consecutively ran two MBCT groups for adults with ID, incorporating metaphors and analogies from ACT (Hayes et al., 1999), modified exercises on developing self-compassion (Neff, 2003) and meditation on the Soles of Feet (Singh et al., 2003), and found participants reported significant decreases in anxiety post-group compared to pre-group. However, the findings suggested that MBCT appears to only be suitable for those with borderline and mild ID who are able to read and write and may not be suitable for those with moderate or severe ID (Idusohan-Moizer et al., 2015).

DBT incorporates skills training (mindfulness, interpersonal effectiveness, emotion regulation, distress tolerance), contingency management, exposure, and cognitive restructuring (Linehan, 2014). Results of a systematic review by McNair, Woodrow, and Hare (2017) suggested that DBT and DBT skills groups can be adapted for individuals with ID, although more high-quality studies are needed to demonstrate efficacy and only one of the studies they reviewed assessed anxiety specifically (Hall et al. (2013), who found a decrease in anxiety following DBT, though that was in individuals with learning disabilities rather than ID). Florez and Bethay (2017) also described a case study of adapted DBT (e.g., briefer sessions of skills training, more concrete images, simpler language, all skills taught through multiple experiential exercises) for a woman with mild ID and GAD; although they reported a decrease in the frequency of challenging behavior, they did not assess or report on anxiety or fear.

Summary and Future Directions

Chambless and Hollon (1998) classified a treatment as *possibly efficacious* if it has been shown to be beneficial to at least three participants in a single-case experimental study or one RCT, whereas they required at least two RCTs or at least three replications in carefully controlled single-case experiments by two or more independent research groups before classifying a treatment as *efficacious*. Other reviews (Lydon, Healy, Callaghan, Mulhern, and Holloway, 2015; Rosen et al., 2016) have used the Chambless criteria to determine the efficacy of interventions to treat anxiety in individuals with ASD and/or ID.

Using the Chambless and Hollon criteria to classify the studies reviewed in Tables 44.1 and 44.2 (excluding the case studies and pre-post designs), in vivo exposure, contingent reinforcement, hierarchy/shaping/stimulus fading, prompting, modeling, noncontingent reinforcement, increasing predictability, and relaxation can be classified as *efficacious* interventions for treating anxiety in ID; extinction/blocking, providing choices, incorporating perseverative interests, and CBT as *possibly efficacious* interventions; and acceptance-based interventions (MBCT, DBT) as interventions that *do not have enough evidence* to determine their efficacy. These classifications must be viewed with caution, given that evaluating the quality or methodological rigor of these studies (e.g., reliable and valid outcome assessment measures; see Chambless & Hollon, 1998) was beyond the scope of this chapter. Not evaluating the methodological quality of each study means that the number of interventions classified as efficacious and possibly efficacious treatments might be an overestimate (e.g., noncontingent reinforcement might have ≤3 replications in single-case experiments by ≥2 independent research groups, but these single-case studies might not necessarily meet all of the quality indicators outlined by Chambless & Hollon, 1998). Interested readers can refer to the paper by Jennett and Hagopian (2008) that reviews the quality of interventions used to treat phobic avoidance in individuals with ID as well as the paper by Rosen et al. (2016) that reviews the quality of interventions used for those with ASD and ID (not including those with ID who do not have ASD).

Overall, consistent with Jennett and Hagopian (2008) and Rosen et al. (2016), behavioral interventions as a whole can be considered efficacious for the treatment of phobic avoidance, fear, and anxiety (though not necessarily as a treatment for *DSM-5* anxiety disorders) in individuals with ID, with **exposure and reinforcement being the most commonly used and well-established interventions**. However, it is important to note that almost all of the studies we reviewed included multicomponent interventions, which makes it challenging to determine the unique contribution of exposure or reinforcement or any of the individual intervention components and clarify which components are necessary or sufficient for change. Thus, future research should aim to identify the active elements of multicomponent intervention packages. That said, most of the interventions reviewed in this chapter have been demonstrated in previous research to be effective in reducing fear or anxiety in neurotypical individuals with anxiety disorders who do not have ID. Further, the use of multicomponent rather than single-component interventions is currently considered to be the best practice for individuals with ID and developmental disabilities (e.g., Carr et al. 1999).

Another important point to consider is that one quarter of the reviewed studies provided noncontingent access to preferred or anti-anxiety stimuli, which would be considered distraction in the CBT literature. Foa and Kozak (1986) posited that distraction interferes with the activation of fear by disrupting the match between aspects of the stimulus setting and the fear structure. Nevertheless, anecdotally, our clinical observations suggested that pairing the anxiety-provoking stimuli with equally potent or even more powerful perseverative stimuli served to counteract the fearful/anxious responses for children with ID in a way that exposure to the feared stimulus alone (without pairing it with antianxiety stimuli) may not have been able to accomplish or at least may not have been able to accomplish as quickly (Moskowitz et al., 2017). Future research must be conducted to examine this hypothesis in individuals with ID, perhaps comparing an “exposure paired with anti-anxiety stimuli” (i.e., counterconditioning) to an “exposure-alone” condition. Although using anti-anxiety stimuli would be considered distraction by some researchers and would be contraindicated for individuals with anxiety disorders who have intact intellectual functioning, this may not be as true for individuals with ID, who may not habituate as quickly or lack the cognitive capacity to understand that their feared outcomes did not come true and that their anxiety may eventually habituate if they remain in the feared situation for long enough without them having to do anything at all to escape or avoid or terminate it.

Finally, as some researchers have noted (Chok et al., 2010; Lydon et al., 2015; Moskowitz et al., 2017), most intervention studies have focused only on the behavioral component of fear/anxiety (overt, observable behaviors) and have neglected the other two components – cognitive and physiological – most likely due to the difficulty measuring cognitions and (to a lesser extent) physiological arousal in those with ID. Moskowitz et al. (2017) found that the more sessions the children with ASD and ID participated in (regardless of whether baseline or intervention), the lower their heart rate (HR) dropped in the presence of the feared stimulus. However, respiratory sinus arrhythmia (RSA) was significantly higher (reflecting increased ability to self-soothe) in intervention than in baseline. This suggests that although it may be possible to reduce HR (and potentially anxiety) in individuals with ASD and ID solely through repeated exposures to feared stimuli, it is possible that the other intervention components (e.g., positive reinforcement, pairing the anxiety-provoking situation with anti-anxiety stimuli, increasing predictability) may have promoted coping, reflected in improvements in RSA as well as substantial behavioral improvements. This hypothesis remains to be explored and highlights the need for more studies that examine physiological arousal when treating anxiety in those with ID. Further, no studies formally assessed changes in the cognitive component of anxiety as a result of intervention for those with ID, suggesting the need to determine better ways to assess thoughts and feelings in this population.

As a result of only measuring overt fear/avoidance behaviors, it is easier to identify specific phobia in those with ID than the other anxiety disorders. As such, almost all of the studies we reviewed in this chapter treated SP or non-*DSM* "phobic avoidance" in individuals with ID, with only 6% of studies treating GAD or SoP, 3% of studies treating SM and OCD, and 0% of studies treating SAD or PD in those with ID. It is therefore much less clear how to assess or treat anxiety disorders that involve thoughts of anxiety (anticipation of future threat) and/or worry (apprehensive expectation) in individuals with ID, an important direction for future research.

Finally, although one might assume that the function of all anxious or fearful behavior in individuals with ID is to escape or avoid anxiety-provoking stimuli/situations, other possible functions must be examined (obtaining reassurance or comfort [i.e., attention], access to soothing or comforting tangible object/activity) and conceptualized as anxiety – when appropriate – rather than only as escape-maintained or attention-maintained problem behavior. Only a few studies have discussed functions other than escape in relation to anxiety, such as access to attention (Moskowitz et al., 2017; Neil et al., 2017). There is a need for more research into individualized interventions tailored to individuals with ID based on the function of their anxious behavior, the specific antecedents that evoke the anxious behavior, and the setting events that increase the likelihood of the anxious behavior (e.g., unpredictability, uncontrollability).

Concluding Comment

Affective states such as fear and anxiety and comorbid anxiety disorders were rarely discussed or acknowledged in individuals with ID until relatively recently, in part because of the difficulty assessing anxiety in this population. In the studies we reviewed, descriptions of the presenting problems varied across treatment studies (e.g., "specific phobia" versus "dental fear" versus "swimming pool avoidance," though all may be phobias) as did the names of treatments used (e.g., "exposure" versus "systematic desensitization" versus "contact desensitization," all seeming to describe the same or similar interventions), with little use of standardized diagnostic measures, outcome measures, or treatment protocols, all of which are important directions for future research. Nevertheless, taken together, the results of our review suggest that behavioral interventions – particularly exposure and reinforcement – can substantially reduce anxious and/or avoidant behaviors and increase approach responses in individuals with ID. Future research must examine whether and how to modify cognitive strategies to adapt to the needs of individuals with ID as well as how to affect components of anxiety other than behaviors (i.e., cognitions, emotions, physiological arousal) in this population.

References

Albano, A. M., & Kendall, P. C. (2002). Cognitive behavioural therapy for children and adolescents with anxiety disorders: Clinical research advances. *International Review of Psychiatry., 14*(2), 129–134.

APA. (2013). *Diagnostic and statistical manual of mental disorders* (5th ed.). Washington, DC: American Psychiatric Association.

Bailey, D. B., Raspa, M., Olmstead, M., & Holiday, D. B. (2008). Co-occurring conditions associated with FMR1 gene variations: Findings from a national parent survey. *American Journal of Medical Genetics, 146A*(16), 2060–2069.

Bandura, A. (1988). Self-efficacy conception of anxiety. *Anxiety, Stress, & Coping, 1*, 77–98.

Barlow, D. H. (2000). Unraveling the mysteries of anxiety and its disorders from the perspective of emotion theory. *American Psychologist, 55*, 1247–1263.

Bouvet, C., & Coulet, A. (2016). Relaxation therapy and anxiety, self-esteem, and emotional regulation among adults with intellectual disabilities: A randomized controlled trial. *Journal of Intellectual Disabilities, 20*(3), 228–240.

Brown, E. C., Aman, M. G., & Lecavalier, L. (2004). Empirical classification of behavioral and psychiatric problems in children and adolescents with mental retardation. *American Journal on Mental Retardation, 109*(6), 445–455.

Burgio, L. D., Willis, K., & Burgio, K. L. (1986). Operantly based treatment procedure for stair avoidance by a severely mentally retarded adult. *American Journal of Mental Deficiency, 91*, 308–311.

Carr, E. G., Horner, R. H., Turnbull, A. P., Marquis, J. G., Magito McLaughlin, D., McAtee, M., et al. (1999). *Positive behavior support for people with developmental disabilities: A research synthesis*. Washington, DC: American Association on Mental Retardation.

Chambless, D. L., & Hollon, S. D. (1998). Defining empirically supported therapies. *Journal of Consulting and Clinical Psychology, 66*, 7–18.

Charlop, M. H., Kurtz, P. F., & Casey, F. (1990). Using aberrant behaviors as reinforcers for autistic children. *Journal of Applied Behavior Analysis, 23*, 163–181.

Charlop-Christy, M. H., & Haymes, L. K. (1996). Using obsessions as reinforces with and without mild reductive procedures to decrease inappropriate behaviors of children with autism. *Journal of Autism and Development Disorders, 26*, 527–546.

Charlot, L., Deutsch, C., Hunt, A., Fletcher, K., & McLivane, W. (2007). Validation of the mood and anxiety semi-structured (MASS) interview for patients with intellectual disabilities. *Journal of Intellectual Disability Research, 51*(10), 821–834.

Chok, J. T., Demanche, J., Kennedy, A., & Studer, L. (2010). Utilizing physiological measures to facilitate phobia treatment with individuals with autism and intellectual disability: A case study. *Behavioral Interventions, 11*, 325–337.

Conyers, C., Miltenberger, R. G., Peterson, B., Gubin, A., Jurgens, M., Selders, A., … Barenz, R. (2004). An evaluation of in vivo desensitization and video modeling to increase compliance with dental procedures in persons with mental retardation. *Journal of Applied Behavior Analysis, 37*, 233–238.

Cooper, S. A., Smiley, E., Morrison, J., Williamson, A., & Allan, L. (2007). Mental ill-health in adults with intellectual disabilities: Prevalence and associated factors. *British Journal of Psychiatry, 190*, 27–35.

Cooray, S. E., Andrews, T., Bailey, N. M., Devapriam, J., McLaren, J. L., Purandare, K. N., … Wijeratne, A. (2016). Anxiety disorders. In R. J. Fletcher, J. Barnhlil, & S. Cooper (Eds.), *Diagnostic manual—Intellectual Disability: A textbook of diagnosis of mental disorders in persons with intellectual disability* (2nd ed., pp. 303–328). Kingston, NY: NADD Press.

Cordeiro, L., Ballinger, E., Hagerman, R., & Hassl, D. (2011). Clinical assessment of DSM-IV anxiety disorders in fragile X syndrome: Prevalence and characterization. *Journal of Neurodevelopmental Disorders, 3*, 57–67.

Dattilo, J., & Rusch, F. R. (1985). Effects of choice on leisure participation for persons with severe handicaps. *Journal of the Association for Persons with Severe Handicaps, 10*, 194.

Davis, T. E., Kurtz, P. F., Gardner, A. W., & Carman, N. B. (2007). Behavioral treatment for specific phobias with a child demonstrating severe problem behavior and developmental delays. *Research in Developmental Disabilities, 28*(6), 546–558.

Deb, S., Thomas, M., & Bright, C. (2001). Mental disorder in adults with intellectual disability. 1:Prevalence of functional psychiatric illness among a community-based population aged between 16 and 64 years. *Journal of Intellectual Disability Research, 45*(6), 495–505.

Dekker, M. C., Koot, H. M., van der Ende, J., & Verhulst, F. C. (2002). Emotional and behavioral problems in children and adolescents with and without intellectual disability. *The Journal of Child Psychology and Psychiatry, 43*, 1087–1098.

Dettmer, S., Simpson, R. L., Myles, B. S., & Ganz, J. B. (2000). The use of visual supports to facilitate transitions of students with autism. *Focus on Autism & Other Developmental Disabilities, 15*(3), 163.

Dixon, M. S., & Gunary, R. M. (1986). Fear of dogs: Group treatment of people with mental handicaps. *British Journal of Learning Disabilities, 14*(1), 6–9.

Douglass, S., Palmer, K., & O'Connor, C. (2007). Experiences of running an anxiety management group for people with a learning disability using behavioural intervention. *British Journal of Learning Disabilities, 35*(4), 245–252.

Erfanian, N., & Miltenberger, R. G. (1990). Contact desensitization in the treatment of dog phobias in persons who have mental retardation. *Behavioral Residential Treatment, 5*, 55–60.

Flannery, K., & Horner, R. (1994). The relationship between predictability and problem behavior for students with severe disabilities. *Journal of Behavioral Education, 4*, 157–176.

Florez, I. A., & Bethay, J. S. (2017). Using adapted dialectical behavioral therapy to treat challenging behaviors, emotional dysregulation, and generalized anxiety disorder in an individual with mild intellectual disability. *Clinical Case Studies, 16*(3), 200–215.

Foa, E. B., & Kozak, M. J. (1986). Emotional processing of fear: Exposure to corrective information. *Psychological Bulletin, 99*, 20–35.

Gillott, A., & Standen, P. J. (2007). Levels of anxiety and sources of stress in adults with autism. *Journal of Intellectual Disabilities, 11*(4), 359–370.

Gray, C. A., & Garand, J. D. (1993). Social stories: Improving responses of students with autism with accurate social information. *Focus on Autistic Behavior, 8*, 1–10.

Grillon, C. (2008). Models and mechanisms of anxiety: Evidence from startle studies. *Psychopharmacology, 199*, 421–437.

Groden, J., Cautela, J., Prince, S., & Berryman, J. (1994). The impact of stress and anxiety on individuals with autism and developmental disabilities. In E. Schopler & G. B. Mesibov (Eds.), *Behavioral issues in autism* (pp. 178–190). New York, NY: Plenum Press.

Hagopian, L., & Jennett, H. (2014). Behavioral assessment and treatment for anxiety for those with autism spectrum disorder. In T. Davis, S. White, & T. Ollendick (Eds.), *Handbook of autism and anxiety*. Cham: Springer.

Hagopian, L. P., & Jennett, H. K. (2008). Behavioral assessment and treatment of anxiety in individuals with intellectual disabilities and autism. *Journal of Developmental and Physical Disabilities, 20*, 467–483.

Hagopian, L. P., Wilson, D. M., & Wilder, D. A. (2001). Assessment and treatment of problem behavior maintained by escape from attention and access to tangible items. *Journal of Applied Behavior Analysis, 34*(2), 229–232.

Hagopian, L. P., Crockett, J. L., & Keeney, K. M. (2001). Multicomponent treatment for blood-injury-injection phobia in a young man with mental retardation.

Hagopian, L. P., Lilly, M., & Davis, T. E. (2017). Behavioral treatments for anxiety in minimally verbal children with ASD. In C. M. Kerns, P. Renno, E. A. Storch, P. C. Kendall, & J. J. Wood (Eds.), *Anxiety in children and adolescents with autism spectrum disorder* (pp. 193–210). San Diego, CA: Elsevier.

Hall, L., Bork, N., Craven, S., & Woodrow, C. (2013). People with learning disabilities experiences of dialectical behaviour therapy skills group: A thematic analysis. *Clinical Psychology & People with Learning Disabilities, 11*, 7–11.

Hayes, S. C., Strosahl, K. D., & Wilson, K. G. (1999). *Acceptance and commitment therapy: An experiential approach to behaviour change*. New York: Guildford.

Helverschou, S. B., & Martinsen, H. (2011). Anxiety in people diagnosed with autism and intellectual disability: Recognition and phenomenology. *Research in Autism Spectrum Disorders, 5*, 377–387.

Idusohan-Moizer, H., Sawicka, A., Dendle, J., & Albany, M. (2015). Mindfulness-based cognitive therapy for adults with intellectual disabilities: An evaluation of the effectiveness of mindfulness in reducing symptoms of depression and anxiety. *Journal of Intellectual Disability Research, 59 Part, 59*(2), 93–104.

Jacobson, E. (1938). *Progressive relaxation*. Chicago: University of Chicago Press.

Jennett, H., & Hagopian, L. P. (2008). Identifying empirically supported treatments for phobic avoidance in individuals with intellectual disabilities. *Behavior Therapy, 39*, 151–161.

Kendall, P. C., Robin, J. A., Hedtke, K. A., Suveg, C., Flannery-Schroeder, E., & Gosch, E. (2005). Considering CBT with anxious youth? Think exposures. *Cognitive and Behavioral Practice, 12*, 136–150.

Kerns, C. M., Kendall, P. C., Berry, L., Souders, M. C., Franklin, M. E., Schultz, R. T., … Herrington, J. (2014). Traditional and atypical presentations of anxiety in youth with autism spectrum disorder. *Journal of Autism and Developmental Disorders, 44*, 2851.

Kerns, C. M., Renno, P., Storch, E. A., Kendall, P. C., & Wood, J. J. (2017). *Anxiety in children and adolescents with autism spectrum disorder*. San Diego: Elsevier.

Kerns, C. M., Rump, K., Worley, J., Kratz, H., McVey, A., Herrington, J., & Miller, J. (2016). The differential diagnosis of anxiety disorders in cognitively-able youth with autism. *Cognitive and Behavioral Practice, 23*(4), 530–547.

Khreim, I., & Mikkelsen, E. (1997). Anxiety disorder in adults with mental retardation. *Psychiatric Annals, 27*, 175–181.

Koegel, R. L., Openden, D., & Koegel, L. K. (2004). A systematic desensitization paradigm to treat hypersensitivity to auditory stimuli in children with autism in family contexts. *Research & Practice for Persons with Severe Disabilities, 29*, 122–134.

Kokina, A., & Kern, L. (2010). Social story interventions for students with autism spectrum disorders: A meta-analysis. *Journal of Autism & Developmental Disorders, 40*, 812–826.

Leyfer, O. T., Woodruff-Borden, J., Klein-Tasman, B. P., Fricke, J. S., & Mervis, C. B. (2008). Prevalence of psychiatric disorders in 4–16-year-olds with Williams syndrome. *Am J Med Genet Part B, 141B*(6), 615–622.

Lindsay, W. R. (1999). Cognitive therapy. *The Psychologist, 12*, 238–241.

Lindsay, W. R., & Baty, F. J. (1986). Behavioural relaxation training: Explorations with adults who are mentally handicapped. *British Journal of Learning Disabilities, 14*(4), 160–162.

Lindsay, W. R., & Baty, F. J. (1989). Group relaxation training with adults who are mentally handicapped. *Behavioural Psychotherapy, 17*, 43–51.

Lindsay, W. R., Baty, F. J., Michie, A. M., & Richardson, I. (1989). A comparison of anxiety treatments with adults who have moderate and severe mental retardation. *Research in Developmental Disabilities, 10*, 129–140.

Lindsay, W. R., Michie, A. M., Baty, F. J., & McKenzie, K. (1988). Dog phobia in people with mental handicaps: Anxiety management training and exposure treatments. *Mental Handicap Research, 1*(1), 39–48.

Linehan, M. M. (2014). *DBT® skills training manual*. New York, NY: Guilford Press.

Love, S. R., Matson, J. L., & West, D. (1990). Mothers as effective therapists for autistic children's phobias. *Journal of Applied Behavior Analysis, 23*, 379–385.

Luiselli, J. (1980). Relaxation training with the developmentally disabled: A reappraisal. *Behavioral Research in Severe Developmental Disabilities,* 1*,* 19 l-2 13.

Luscre, D. M., & Center, D. B. (1996). Procedures for reducing dental fear in children with autism. *Journal of Autism and Developmental Disorders, 26*, 547–556.

Lydon, S., Healy, O., Callaghan, O. O., Mulhern, T., & Holloway, J. (2015). A systematic review of the treatment of fears and phobias among children with autism spectrum disorders. *Review Journal of Autism and Developmental Disorders, 2*, 141–154.

Mace, A. B., Shapiro, E. S., & Mace, F. C. (1998). Effects of warning stimuli for reinforcer withdrawal and task onset on self-injury. *Journal of Applied Behavior Analysis, 31*, 679.

Maguire, K. B., Lange, B., Scherling, M., & Grow, R. (1996). The use of rehearsal and positive reinforcement in the dental treatment of uncooperative patients with mental retardation. *Journal of Developmental and Physical Disabilities, 8*, 167–177.

Marwood, H., & Hewitt, O. (2012). Evaluating an anxiety group for people with learning disabilities using mixed methodology. *British Journal of Learning Disabilities, 41*, 150.

Matson, J. L. (1981a). Assessment and treatment of clinical fears in mentally retarded children. *Journal of Applied Behavior Analysis, 14*(3), 287–294.

Matson, J. L. (1981b). A controlled study of phobias in mentally retarded adults. *Behavior Research and Therapy, 19*(2), 101–107.

Matson, J. L. (1982). Treating obsessive-compulsive behavior in mentally retarded adults. *Behavior Modification, 6*, 551–567.

Matson, J. L., Belva, B. C., Hattier, M. A., & Matson, M. L. (2012). Scaling methods to measure psychopathology in persons with intellectual disabilities. *Research in Developmental Disabilities, 33*(2), 549–562.

Matson, J. L., & Dempsey, T. (2009). The nature and treatment of compulsions, obsessions, and rituals in people with developmental disabilities. *Research in Developmental Disabilities, 30*(3), 603–611.

McNair, L., Woodrow, C., & Hare, D. (2017). Dialectical behaviour therapy [DBT] with people with intellectual disabilities: A systematic review and narrative analysis. *Journal of Applied Research in Intellectual Disabilities, 30*, 787–804.

Moskowitz, L. J., Mulder, E., Walsh, C. E., McLaughlin, D., Zarcone, J. R., Proudfit, G., & Carr, E. G. (2013). A multimethod assessment of anxiety and problem behavior in children with autism spectrum disorders and intellectual disability. *American Journal on Intellectual and Developmental Disabilities, 118*(6), 419–434.

Moskowitz, L. J., Walsh, C. E., Mulder, E., McLaughlin, D., Hajcak, G., Carr, E. G., & Zarcone, J. R. (2017). Intervention for anxiety and problem behavior in children with autism spectrum disorder and intellectual disability. *Journal of Autism and Developmental Disorders, 47*, 3930–3948.

Neff, K. D. (2003). The development and validation of a scale to measure self compassion. *Self and Identity, 2*, 223–250.

Neil, N., Vause, T., Jaksic, H., & Feldman, M. (2017). Effects of group functional behavior based cognitive-behavioral therapy for obsessive-compulsive behavior in a youth with autism spectrum disorder. *Child & Family Behavior Therapy, 39*(3), 179–190.

Rapp, J. T., Vollmer, T. R., & Hovanetz, A. (2005). Evaluation and treatment of swimming pool avoidance exhibited by an adolescent girl with autism. *Behavior Therapy, 36*, 101–105.

Reardon, T. C., Gray, K. M., & Melvin, G. A. (2015). Anxiety disorders in children and adolescents with intellectual disability: Prevalence and assessment. *Research in Developmental Disabilities, 36*, 175–190.

Reaven, J., Blakeley-Smith, A., Culhane-Shelburne, K., & Hepburn, S. (2012). Group cognitive behavior therapy for children with high-functioning autism spectrum disorders and anxiety: A randomized trial. *Journal of Child Psychology and Psychiatry and Allied Disciplines, 53*, 410–419.

Reid, K. A., Smiley, E., & Cooper, S. A. (2011). Prevalence and associations of anxiety disorders in adults with intellectual disabilities. *Journal of Intellectual Disability Research, 55*, 172–181.

Reiss, S., Levitan, G. W., & Szyszko, J. (1982). Emotional disturbance and mental retardation: Diagnostic overshadowing. *American Journal of Mental Deficiency, 86*, 567–574.

Ricciardi, J. N., Luiselli, J. K., & Camare, M. (2006). Shaping approach responses as intervention for specific phobia in a child with autism. *Journal of Applied Behavior Analysis, 39*(4), 445–448.

Rosen, T. E., Connell, J. E., & Kerns, C. M. (2016). A review of behavioral interventions for anxiety-related behaviors in lower-functioning individuals with autism. *Behavioral Interventions, 31*, 120–143.

Royston, R., Howlin, P., Waite, J., & Oliver, C. (2017). Anxiety disorders in Williams syndrome contrasted with intellectual disability and the general population: A systematic review and meta-analysis. *Journal of Autism and Developmental Disorders, 47*(12), 3765–3777.

Runyan, M. C., Stevens, D. H., & Reeves, R. (1985). Reduction of avoidance behavior of institutionalized mentally retarded adults through contact desensitization. *American Journal of Mental Deficiency, 90*, 222–225.

Schmidt, J. D., Luiselli, J. K., Rue, H., & Whalley, K. (2013). Graduated exposure and positive reinforcement to overcome setting and activity avoidance in an adolescent with autism. *Behavior Modification, 37*, 128–142.

Schreibman, L., Whalen, C., & Stahmer, A. C. (2000). The use of video priming to reduce disruptive transition behavior in children with autism. *Journal of Positive Behavior Interventions, 2*, 3–11.

Shabani, D. B., & Fisher, W. W. (2006). Stimulus fading and differential reinforcement for the treatment of needle phobia in a youth with autism. *Journal of Applied Behavior Analysis, 39*, 449–452.

Shogren, K. A., Faggella-Luby, M. N., Jik Bae, S., & Wehmeyer, M. L. (2004). The effect of choice-making as an intervention for problem behavior: A meta analysis. *Journal of Positive Behavior Interventions, 6*, 228–237.

Singh, N., Wahler, P., Adkins, A., & Myers, S. (2003). Soles of the feet: A mindfulness based self-control intervention for aggression by an individual with mild mental retardation and mental illness. *Research in Developmental Disabilities, 24*, 158–169.

Sukhodolsky, D. G., Scahill, L., Gadow, K. D., Arnold, L. E., Aman, M. G., McDougle, C. J., … Vitiello, B. (2008). Parent rated anxiety symptoms in children with pervasive developmental disorders: Frequency and association with core autism symptoms and cognitive functioning. *Journal of Abnormal Clinical Abnormal Child Psychology, 36*, 117.

Suveg, S., Comer, J. S., Furr, J. M., & Kendal, P. C. (2006). Adapting manualized CBT for a cognitively delayed child with multiple anxiety disorders. *Clinical Case Studies, 5*, 488.

Unwin, G., Tsimopoulou, I., Kroese, B. S., & Azmi, S. (2016). The effectiveness of cognitive behavioural therapy (CBT) programmes for anxiety or depression in adults with intellectual disabilities: A review of the literature. *Research in Developmental Disabilities, 51-52*, 60–75.

Waranch, H. R., Iwata, B. A., Wohl, M. K., & Nidiffer, E. D. (1981). Treatment of a retarded adult's mannequin phobia through in vivo desensitization and shaping approach responses. *Journal of Behavior Therapy and Experimental Psychiatry, 12*, 359–362.

Wehmeyer, M. L. (1999). A functional model of self-determination: Describing development and implementing instruction. *Focus on Autism and Other Developmental Disabilities, 14*, 53–61.

Wilde, L., Koegel, L. K., & Koegel, R. L. (1992). *Increasing success in school through priming*. Santa Barbara: University of California.

Witwer, A. N., & Lecavalier, L. J. (2010). Validity of comorbid psychiatric disorders in youngsters with autism spectrum disorders. *Journal of Developmental and Physical Disabilities, 22*(4), 367–380.

Wolff, J. J., & Symons, F. J. (2013). An evaluation of multi-component exposure treatment of needle phobia in an adult with autism and intellectual disability. *Journal of Applied Research in Intellectual Disabilities, 26*, 344–348.

Wright, K. P. (2013). Cognitive behavioral therapy for anxiety in a man with autism spectrum disorder, intellectual disability, and social phobia. *Advances in Mental Health and Intellectual Disabilities, 7*(5), 284–292.

45

Depression

Gemma L. Unwin, Shoumitro Deb, and John Rose

Definition of Depression, Incidence and Prevalence

Depression refers to a range of mental health problems characterised by low mood, absence of positive affect, loss of interest and enjoyment, and a range of concomitant behavioural, physical, emotional and cognitive symptoms, and results in significant behavioural and functional disability. The course of depression can be recurrent or chronic but in some cases complete remission is possible. In people with ID, depression tends to be more severe and enduring than in the general population (Collishaw, Maughan, & Pickles, 2004), perhaps due to increased vulnerability or poorer management. Depression commonly co-occurs with anxiety in people with ID and intervention studies with combined samples are common.

The DSM-V (APA, 2013) has separated out depressive disorders from bipolar and related disorders and includes a new diagnostic category of 'disruptive mood dysregulation disorder' alongside major depressive disorder, persistent depressive disorder (dysthymia), premenstrual dysphoric disorder, substance/medication-induced depressive disorder, and depressive disorder due to another medical condition. Disruptive mood dysregulation disorder may become the focus of future research in ID. It was developed to address the over-diagnosis of bipolar disorder in children, however, both children and adults with ID could meet the diagnostic criteria, but to date, no studies have examined its clinical utility in ID (Charlot et al., 2017). The new version of the International Classification of Diseases, currently in development (ICD-11 Draft/Beta version; World Health Organization, 2017) has single episode and recurrent depressive disorder, dysthymic disorder, and mixed depressive and anxiety disorder as diagnostic categories. The DM-ID-2 provides an adapted framework for diagnosis of mental health problems in people with ID (Fletcher, Barnhill, & Cooper, 2017). It makes limited adaptations to the standard diagnostic criteria for depression but provides detailed behavioural descriptions of symptoms which can be used with informants to improve the reliability of their report and therefore diagnosis (Chap. 12; Charlot et al., 2017). Whilst diagnostic categories may be helpful for epidemiological, research, and clinical purposes, their clinical validity in understanding complex

---

G. L. Unwin (✉) · J. Rose
Centre for Applied Psychology,
School of Psychology, The University of Birmingham,
Edgbaston, Birmingham, UK
e-mail: G.L.Unwin@bham.ac.uk;
J.L.Rose@bham.ac.uk

S. Deb
Division of Brain Sciences, Department of Medicine,
Imperial College London, Centre for Psychiatry,
London, UK
e-mail: S.Deb@imperial.ac.uk

J. L. Matson (ed.), *Handbook of Intellectual Disabilities*, Autism and Child Psychopathology Series, https://doi.org/10.1007/978-3-030-20843-1_45

mental health problems has been questioned (Stenfert Kroese, Dewhurst, & Holmes, 2001). In the UK, the British Psychological Society promotes the use of formulation to move away from a disease model of mental illness to understand conditions as a response to psychosocial factors such as trauma, loss or inequality (Jahoda, Stenfert Kroese, & Pert, 2017).

Clinical Presentation and Diagnosis

Box 45.1 gives an overview of the features of clinical depression.

Box 45.1: Clinical Features of Depressive Disorder

General Features

- Severe, persistent low mood that is pervasive across all situations and unresponsive to circumstances, even those that would normally lift a person's mood.
- Can be precipitated by a life event.
- A marked loss of interest in all activities ('anhedonia').
- Feelings of excessive tiredness.
- Loss of energy and initiative.

Biological Features

- Sleep disturbance (usually insomnia in the form of early morning awakening, however, in atypical cases excessive sleep can be a feature, early insomnia and interrupted sleep may also be present).
- Changes in appetite (usually decrease but increase in some atypical cases).
- Significant weight loss not due to dieting or any other physical illness (increase in weight in atypical cases).
- Psychomotor retardation or agitation.
- Decreased libido.
- Diurnal variation in mood (mood being worse in the morning).

Cognitive Features

- Loss of self-esteem or confidence.
- Lack of purpose in life (life is not worth living, there is no future).
- Feelings of guilt or self-reproach.
- Suicidal thoughts or behaviour.
- Difficulty with concentration.
- Marked slow thought processes.
- Ruminative thoughts (pre-occupation with depressive thoughts).
- Speech is slow and lacks content.

Appearance

- Depressed look (may be crying).
- Restless and agitated.
- Poor eye contact.
- Evidence of self-neglect (or sometimes self-harm).
- Stooped posture.
- Slow and sometimes awkward movements.

Psychotic Features (in Severe Cases)

- Auditory hallucinations (often derogatory and in second person) or delusions (nihilistic).

Those with milder ID may present with depressive symptoms similar to those of the general population and be able to recognise and articulate their emotional state. Standard diagnostic criteria such as DSM-V (APA, 2013) or ICD-10 (World Health Organisation, 1992) may therefore be applied in some cases. However, those with moderate or severe ID are less able to express their internal states so clinicians will be more reliant on observable features and informant-report. Diagnosis is more commonly based on agitation, aggression, self-injury, appearance of sadness, and changes in sleep or appetite, however, these are not reliable behavioural equivalents of depression, as discussed in more detail later. Thus, diagnosis is more complicated than for the general population, and may in part, be reliant on the observational and reporting skills of informants such as paid carers and family members as well as the assessment skills of the clinician.

Rose et al. (2013) found that 34% of a sample of people with ID who were referred for an anger management group had high levels of depression as measured by the Glasgow Depression Inventory (Cuthill, Espie, & Cooper, 2003). They also asked the people with ID themselves and the staff who supported them to complete a range of other measures that were then used to predict self-reported anger in the people with ID. The staff relied on a number of indicators which reflected the outwardly expressed challenging behaviours of individuals to explain expressed anger. The scores people with ID achieved on measures relating to their own emotional state, including their depression scores, best reflected their own assessment of expressed anger. This finding suggests, perhaps not surprisingly, that staff were using mainly external cues to understand the emotional state of people with ID. When the people with ID themselves report their own internal state, information provided by them is often unknown, not used or may be disregarded by staff in relation to the person with ID's emotional condition. Previous research has suggested that staff measures do not correlate well with the self-report of individuals with ID in this area (Bramston & Fogarty, 2000) however, there are some behavioural correlates of depression including: sad mood, crying, and anhedonia (Hurley, 2008).

As staff are likely to facilitate a referral to health services to support people with ID their relative lack of ability to identify depression may partially explain why people with ID are more likely to be identified as having difficulties with externalising symptoms such as challenging behaviour and anger rather than depression. For example, Hurley (2008) found that depressed mood was rarely cited as the primary complaint for people with ID and diagnosed depression, rather it was behavioural disturbances which prompted referral. Similarly, Mileviciute and Hartley (2015) found that informants reported fewer depressive symptoms than people with ID reported themselves. This suggests that a much more detailed assessment process is required that includes an assessment from the individuals' personal perspective, where possible, to get a true insight into the levels of depression in this population. Furthermore, awareness raising in carers about mental health problems beyond behavioural features may be required. The Diagnostic Manual-Intellectual Disability (DM-ID-2, Chap. 12, Charlot et al., 2017) therefore provides examples of how people with ID may demonstrate each depressive symptom in an attempt to reduce diagnostic errors related to reliance on informant reports.

In the field of ID 'diagnostic overshadowing' has been implicated whereby excessive focus is given to the ID rather than underlying mental health problems, resulting in under diagnosis (Reiss & Szyszanko, 1993). This may be further compounded by 'behavioural overshadowing' in which presenting behavioural problems may be indicative of underlying mental health problems, such as depression, but serve to mask the condition with treating clinicians (especially non-specialists) giving excessive attention to the behaviour rather than seeking to identify underlying mood disorder (Hemmings, Deb, Chaplin, Hardy, & Mukherjee, 2013). Whilst behavioural problems may be a common and atypical manifestation of depression in people with ID, they should not be considered as behavioural equivalents as they are non-specific – they occur at high rates in all mental health problems in people with ID. Aggression could be considered as a "final common pathway for distress" for people with ID who have limited other opportunities to demonstrate this (Charlot et al., 2017).

Prevalence

Depression is a common disorder among the general population and even more common among people with ID, however, prevalence is currently unknown due to the aforementioned problems with diagnosis along with methodological issues in existing prevalence studies such as differences in sampling and diagnostic procedures. Emotional problems, such as depression, are the most common psychiatric diagnoses for people with ID

(Azam, Sinai, & Hassiotis, 2009; Reid, Smiley, & Cooper, 2011; Richards et al., 2001; Smiley, 2005; Tsiouris, Kim, Brown, & Cohen, 2011). One of the few population-based studies reports a prevalence rate of 4.1% for unipolar depression, (based on clinical diagnosis) among 1025 service-users with ID aged 16 year or over in the Glasgow Health Board area in Scotland (Cooper, Smiley, Morrison, Williamson, & Allan, 2007). When using DSM-IV (American Psychiatric Association, 1994) criteria, 3.6% were diagnosed with affective disorder and the rate increased to 5.7% when using DC-LD (Royal College of Psychiatrists, 2001) adapted criteria. It could be anticipated that reported prevalence will increase with the addition of the new diagnostic category 'disruptive mood dysregulation disorder' and associated adapted criteria in the DM-ID-2 (Charlot et al., 2017). More recently, a large-scale survey reported that 14% of 4069 community-based adults with ID receiving specialist services in New York had a documented DSM-IV or DSM-IV-TR diagnosis of depression (Tsiouris et al., 2011).

Risk Factors for Depression

Risk factors for depression in the non-ID, general population include: (a) negative affectivity as a temperamental factor, including borderline personality disorder; (b) environmental factors including childhood adverse experiences; (c) genetic and physiological factors (for example, two to four fold increase of depression among first degree relatives of those who have suffered from major depressive disorder); (d) other biological factors such as presence of chronic medical and other disabling conditions and substance use (American Psychiatric Association, 2013). As with the general population, a range of biological, psychological and social factors are also implicated in the development of depression in people with ID (Irvine & Beail, 2016; see Fig. 45.1). A biopsychosocial model may account for the increased prevalence of depression in people with ID who may have more physical predispositions, higher rates of physical illness, exposure to psychological stressors, limited capacity for adaptive coping, and poorer social environments. Psychosocial factors for people with ID play a significant role as they face inequities such as social disadvantage, poor self-esteem, stigmatisation and poor social support.

Research has sought to identify risk factors for depression among people with ID, most commonly a range of demographic variables, life events and psychiatric diagnoses/mental health conditions. The following presents a brief overview of the research findings pertaining to the most commonly studied risk factors. Some research results have been inconsistent and determining a definitive risk profile remains elusive, not least because of the aforementioned challenges in diagnosing depression.

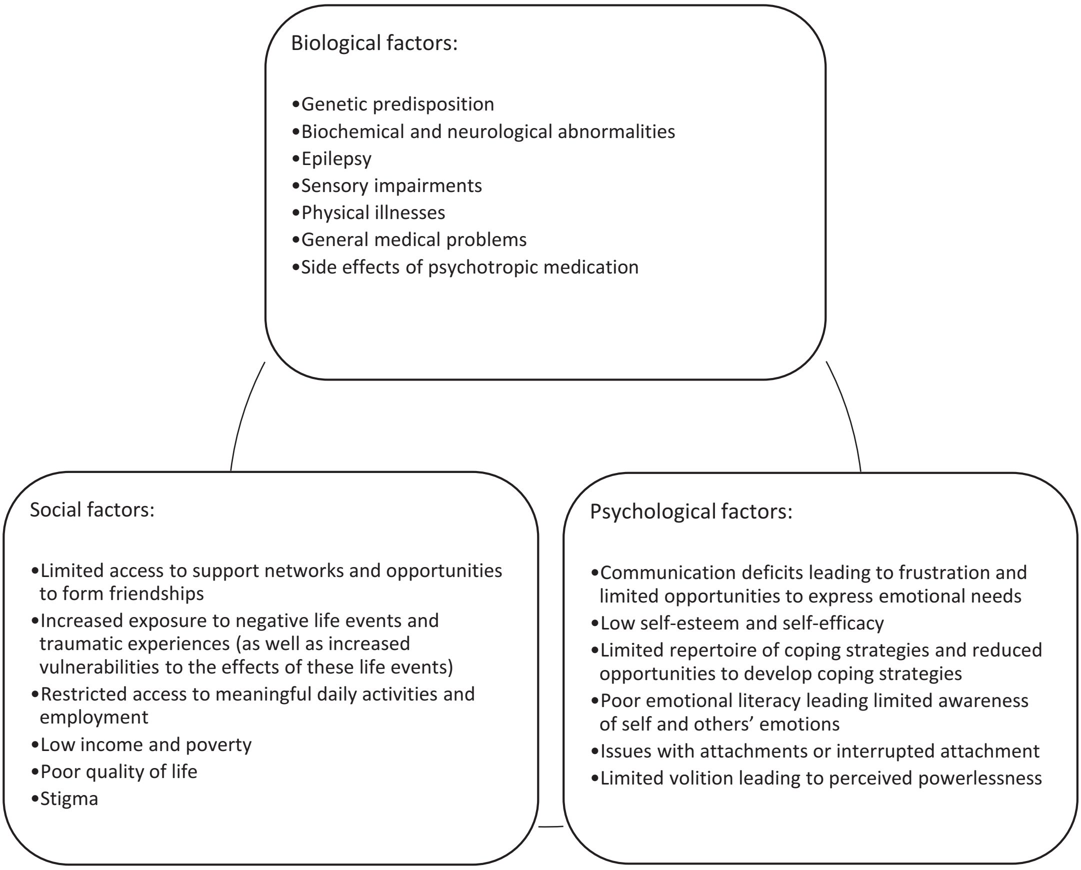
Gender: Diagnosis of depression is more common in females with ID (Bhaumik, Tyrer, McGrother, & Ganghadaran, 2008).

Age: Depression is more common in older people (over 65 years) with ID (Cooper, 1997) with a positive correlation between depressive symptoms and age (Hermans & Evenhuis, 2013).

Severity of ID: No association between depression and severity of ID has been reported (Cooper et al., 2007).

Genetic syndromes: There are certain genetic syndromes that cause ID in which there is an increased rate of depression (Levitas et al., 2016). These include Tuberous sclerosis, 22 q11 deletion syndrome, Neurofibromatosis, Prader-Willi syndrome, untreated Phenylketonuria, Myotonic dystrophy type 1 and Rubenstein Taybi syndrome (Charlot et al., 2016; Deb, Unwin, Rojahn, & Cooper, 2018),. A review did not find any evidence of increased prevalence of depression among people with Down syndrome (Walker, Dosen, Buitelaar, & Janzing, 2011).

Neurological abnormalities: Low levels of neurotransmitters, particularly serotonin in the brain has been implicated in depression among the non-ID general population and the underlying brain damage in people with ID may make them more vulnerable to these neurochemical changes but the empirical evidence to support this hypothesis is not currently available (Hassiotis, Stueber, Thomas, & Charlot, 2014).



Biological factors:

- Genetic predisposition
- Biochemical and neurological abnormalities
- Epilepsy
- Sensory impairments
- Physical illnesses
- General medical problems
- Side effects of psychotropic medication

Social factors:

- Limited access to support networks and opportunities to form friendships
- Increased exposure to negative life events and traumatic experiences (as well as increased vulnerabilities to the effects of these life events)
- Restricted access to meaningful daily activities and employment
- Low income and poverty
- Poor quality of life
- Stigma

Psychological factors:

- Communication deficits leading to frustration and limited opportunities to express emotional needs
- Low self-esteem and self-efficacy
- Limited repertoire of coping strategies and reduced opportunities to develop coping strategies
- Poor emotional literacy leading limited awareness of self and others' emotions
- Issues with attachments or interrupted attachment
- Limited volition leading to perceived powerlessness

Fig. 45.1 Risk factors for depression in people with ID

Epilepsy: The relationship between epilepsy and mental illness in adults with ID is complex (see review by Deb, 2007b) but minor psychiatric symptoms of depression and anxiety seem to be more commonly associated with epilepsy.

Autism spectrum conditions: Similar rates of depression for those with and without autism have been reported (Melville et al., 2008). However, other research has reported that the onset or increase in behaviours such as hoarding, repetitive touching or tapping may be presenting features of depression (Kannabiran & McCarthy, 2009).

Dementia: Onset of dementia is associated with signs of depression such as low mood and disturbed sleep (Strydom et al., 2010).

Medical conditions: A number of chronic medical conditions such as hypothyroidism (common in people with Down syndrome), dental problems, acid reflux, drug induced Parkinsonism, sensory problems (vision and hearing), all of which are common in people with ID, are likely to be associated with a higher rate of depression (Kwok & Cheung, 2007).

Life Events: Exposure to adverse life events such as bereavement or moving home increase the risk of depression (Hulbert-Williams & Hastings, 2008). Furthermore, total number of negative life events has been shown to be associated with depression in both children (Hatton & Emerson, 2004) and adults (Cooper et al., 2007; Esbensen & Benson, 2006) with ID.

Social factors: Other social factors including poor diet, lack of physical exercise, and lack of appropriate social, occupational and vocational opportunities make people with ID vulnerable to developing depressive symptoms (Hassiotis et al., 2014) whereas positive social support seems to mitigate against depressive symptoms (McGillivray & McCabe, 2007).

Psychological factors: Low self-esteem, loneliness and negative automatic thoughts have been shown to be associated with depressive symptoms in adults with mild to moderate ID (Lunsky, 2004).

Medication for Depression

The management of depression includes comprehensive assessment and diagnosis followed by treatment by competent practitioners in either a psychological or pharmacological approach, or a combination of these. Psychological therapies have become first line interventions for the treatment of depression in the general population and UK-based guidelines on the treatment of depression in people with ID (NICE, 2016). However, antidepressants are commonly prescribed to people with ID, often in the absence of a diagnosis of depression, and in the absence of robust evidence of their effectiveness as people with ID are often excluded from medication trials. Doan, Lennox, Taylor-Gomez, and Ware (2013) found that 20% of community-based adults with ID residing in Brisbane, Australia were taking antidepressant medication with 30% of those prescribed psychotropic medication not having a psychiatric diagnosis. Newer generation antidepressants (selective serotonin reuptake inhibitors; SSRIs) are prescribed more often than old generation antidepressants. A study of medication use among a clinical sample of 100 adults with ID and aggressive behaviour found that 10% were prescribed citalopram, 6% fluoxetine and 7% paroxetine (Deb, Unwin, & Deb, 2015). Previously, Holden and Gittlesen (Holden & Gitlesen, 2004) found that 8.5% of 300 adults with ID who were in receipt of ID services were prescribed SSRIs and 2.4% other antidepressants. Of those taking SSRIs, only 40% had a diagnosis of depression and none of those taking other antidepressants had a diagnosis of depression.

Prescribing guidelines for people with ID recommend starting low and going slow (NICE, , 2016; Deb, 2016; Bhaumik, Branford, Barrett, et al., 2015; Sullivan et al., 2018; Rush & Frances, 2000). It is recommended to use the lowest effective dose, taking account of side effects and difficulties the person may have in reporting them, whilst avoiding sub-therapeutic doses which may not be sufficient to treat the mental health problem. There is limited evidence of the effectiveness of antidepressants among people with ID, as highlighted below, and therefore guidelines are based upon evidence derived from the general population as it is asserted that they have a similar effectiveness and side effect profile (Bhaumik et al). SSRIs are recommended as first line medication-based treatment for depression in people with ID (NICE; Bhaumik et al). This group of medications work by increasing the levels of serotonin in the brain. The reason SSRIs are the first choice antidepressant is because there is good evidence that they are as effective as other classes of antidepressants and they have a better risk-benefit balance with regards to their adverse effects as compared with the other classes of antidepressant medications. They are also less dangerous in overdose as compared with older generation of antidepressants (Cleare et al., 2015).

Tricyclic antidepressants (TCAs) are an older class of antidepressant medication compared with SSRIs. They work by increasing serotonin and noradrenaline levels in the brain. Monoamine oxidase inhibitors (MAOIs) are again an older class of antidepressants compared with SSRIs. They work by increasing the levels of serotonin and noradrenaline in the brain. However the action of MAOIs is not limited to the brain. They also act in the liver and gut. When a person taking a MAOI eats food containing tyramine (a substance that can affect blood vessels), due to the action of the MAOI in the gut, the tyramine is not broken down. This leads to high levels of tyramine in the body which can enter the blood stream and cause a 'hypertensive crisis'. This is a dangerous adverse effect of MAOIs and therefore tyramine-containing foods need to be avoided. Due to the serious adverse effects of MAOIs, they are rarely used in people with ID. There are several other antidepressants that do not fall into the categories of SSRIs, TCAs or MAOIs. These include mirtazepine, venlafaxine, agomelatine and duloxetine.

These antidepressants are often used before trying TCAs or MAOIs (NICE, 2009). In certain patients depression can be very difficult to treat. It may be necessary to 'augment' the antidepressant therapy by adding another medication. This should only be done in consultation with prescribing guidelines. An antidepressant can be augmented with lithium, an antipsychotic (e.g. aripiprazole, olanzapine, quetiapine or risperidone) or another antidepressant such as mirtazepine.

A systematic review located no controlled studies of medication for depression among adults with mild to moderate ID (Koslowski et al., 2016). Another systematic review of interventions for adults with mild ID (a broad definition used to include studies where 50% + had mild ID) and mental ill-health found only two studies of two different antidepressants (Osugo & Cooper, 2016). Bhaumik, Collacott, Gandhi, Duggirala, and Wildgust (1995) conducted a retrospective case notes review to evaluate the effects of antidepressants among 71 adults with ID (60% with mild ID) with an ICD-9 diagnosis of depression. They used retrospective ratings of clinical global improvement (CGI) based on descriptions in clinic notes. Mean CGIs were 2.09 for those prescribed SSRIs and 1.96 for those prescribed tricyclic antidepressants indicating that both groups were 'much improved' over the course of the 3 year review period. However, 6.4% discontinued SSRIs and 9.5% discontinued tricyclics due to lack of efficacy and 9.6% and 26% discontinued due to adverse effects respectively. The same group carried out another case notes review to examine the effectiveness of fluoxetine and paroxetine among 56 adults with ID with similar results to those of the SSRI group in the earlier study (Bhaumik & Wildgust, 1996). Of those discontinuing paroxetine, 41.6% experienced withdrawal symptoms whereas none of those discontinuing fluoxetine reported withdrawal symptoms. The results of these studies need to be interpreted with caution as the CGI is subjective and retrospective, being based on evidence provided in case notes, there were no control groups, and administration of the medications were not standardised.

An additional long-term, open label study of citalopram in 20 inpatients with ID and symptoms aligning to affective, motivational, motor, and vital domains of depression followed participants up for six to 12 months (Verhoeven, Veendrik-Meekes, Jacobs, van den Berg, & Tuinier, 2001). A moderate to marked improvement on the CGI was reported for 12 participants. No response was observed for the other eight participants. One participant without history of epilepsy developed seizures and another developed a delirious state; no other side effects were reported. Again, this study does not have a control group and subjective outcome measures were used. Furthermore, reporting of results is incomplete – there are no data on biweekly monitoring of frequency and intensity of target symptoms.

Owing to the lack of studies of antidepressants for depression, it may be useful to examine the literature on antidepressants for other indications, such as behavioural problems, especially given the aforementioned challenges with making diagnoses and with reference to information on tolerability. Sohanpal et al. (2007) conducted a systematic review of studies evaluating antidepressants for behavioural problems such as aggression, self-injurious behaviour and property destruction. They found one cross over, randomised controlled trial (RCT), seven prospective uncontrolled studies and two retrospective studies. The RCT evaluated the tricyclic antidepressant clomipramine and the rest investigated SSRIs. The studies tended to use small sample sizes, non-validated outcome measures, were of short duration with limited follow-up periods, and used antidepressants as add on therapy. The authors summarise that response to SSRIs was varied – some studies showed favourable results, others showed negative effects and some reported mixed outcomes with improvement in self-injurious behaviour or aggression in less than 50% of participants across the studies. More favourable results were found in those with underlying anxiety or obsessive compulsive disorder.

Authors have expressed concern over adverse effects related to antidepressant use and the results of the limited studies support this. One adverse effect of antidepressants, of particular concern for people with ID owing to the increased prevalence of epilepsy (Berney & Deb, 2012; Deb, 2007a), is the potential to increase seizure frequency or reduce the threshold at which seizures occur in people with epilepsy. Another is the potential to increase anxiety or agitation, which may already be elevated among people with ID who also have difficulties expressing their discomfort verbally, however, this usually settles after a few weeks of treatment. Other adverse effects of SSRIs include gastrointestinal adverse effects (e.g. nausea, vomiting, indigestion, abdominal pain, diarrhoea, constipation, reduced appetite and weight loss), possible increased thoughts or self-harm and suicidal thoughts and acts when initiating treatment, sexual dysfunction, difficulty sleeping, urinary retention (being unable to pass urine), sweating, tremor, rash, low sodium levels in the blood (hyponatraemia), and serotonin syndrome (a rare but serious adverse effect). Adverse effects of TCAs include effects on the heart (such as abnormal heart rhythm), low blood pressure on standing which could increase risk of falling, 'antimuscarinic' adverse effects (e.g. dry mouth, blurred vision, constipation, and urinary retention), anxiety, dizziness, agitation, confusion, sleep problems, 'pins and needles', hallucinations, delusions and mania, breast enlargement, changes in appetite and weight, changes in blood sugar (glucose) levels, nausea and vomiting, and taste disturbance. Withdrawal of antidepressants can also result in unwanted effects such as headache, anxiety, dizziness, sleep disturbance, sweating and flu like symptoms. For this reason, it is recommended that medication should be withdrawn slowly and gradually and the introduction of medication should be rationalised in the first place. Any cognitive and functional impairments, as a result of starting or withdrawing medication is an additional concern for people who already have such impairments.

Meins (1996, cited in Verhoeven et al., 2001) observed that, owing to atypical symptomatology, mis-diagnosis and under-diagnosis, depression in people with ID may be treated with antipsychotics rather than antidepressants. A further retrospective case notes review comparing various antipsychotics with no antipsychotics was identified in the review by Osugo and Cooper (2016; Shedlack et al., 2005) in which 37% of 72 participants had major depression (with or without psychosis). For those with major depression, Global Assessment of Function (GAF) ratings increased by 9 points in the no antipsychotic group but showed little change in the atypical antipsychotic group. The opposite was reported for those with major depression with psychosis. However, the same methodological weaknesses persist in this study (use of subjective outcome measures, unstandardised medication procedures including different doses and concomitant medication, limited information on blinding provided, etc.) and no information is provided on side effects. There is therefore no evidence to support the use of other classes of medication in the treatment of depression in ID and this practice is not recommended.

In the UK, a national project has been initiated to stop the overuse of medication of people with ID (Stopping over medication of people with a learning disability, autism or both; STOMP). It is in reaction to the high levels of inappropriate psychotropic medication prescription to these vulnerable groups.

Psychological Interventions for Depression

It was previously asserted that, owing to cognitive deficits, people with ID could not engage in psychological therapy. This assumption has been challenged and there are now a greater range of psychological therapies available to people with ID. Cognitive behavioural therapy (CBT) remains the most commonly studied (e.g. see Beail, 2016) and is recommended for the treatment of depressive disorders (American Psychiatric Association, APA, 2010; National Institute for Health and Clinical Excellence, NICE, 2009). However, the application of CBT for depression among people with ID is not considered in these guidelines which focus on the general population. In the UK, CBT adapted for people with ID is recommended for the treatment of depression or subthreshold depressive symptoms in people with milder ID (NICE, 2016). The evidence base for CBT's effectiveness with this population is discussed in more detail later in this chapter.

The evidence of effectiveness for other forms of psychological therapy, such as psychotherapy and 'new wave' therapies remains more limited, however, trials are increasingly being planned and funded. In line with this, modified psychological interventions are being developed, appropriate for different communication styles, necessity for prompting, simplification, and breaking down of tasks etc. In developing the UK-based guideline for the treatment of mental health problems among people with ID, NICE recommend that psychological interventions are adapted and trialed to ensure they are accessible to people with ID (NICE, 2016). Whilst adapted therapeutic protocols may be suitable for people with mild ID, it is not clear whether such protocolised interventions are suited to people with more severe intellectual impairments (Campbell, Robertson, & Jahoda, 2014) and people with severe ID, or without capacity to consent, are still often excluded from trials due to ethical issues. Making large modifications to interventions to make them more suitable for people with ID limits the applicability of the evidence base derived from the general population (Leyin, 2011) and may compromise fidelity to the approach. Therefore, trials with people with ID which also consider fidelity are required to make more evidence-based psychological interventions available. More research with people with more severe ID is warranted to investigate whether psychological interventions can be sufficiently adapted. For example, whilst certain aspects of CBT may not be accessible for some people with ID, some of the behavioural aspects such as activity scheduling are relevant to all. Practice-based evidence may be especially helpful here to resolve some of the ethical issues in involving people who lack capacity in clinical trials. The following presents an overview of the evidence for the effectiveness of the most common forms of psychological interventions for depression.

Behavioural Activation

Behavioural activation has been shown to be effective in treating depression in the general population (Ekers, Richards, McMillan, Bland, & Gilbody, 2011; Richards, Ekers, McMillan, et al., 2016). In behavioural activation, the focus is on behaviour change, rather than affecting behavioural change through modifying thinking patterns (as with cognitive therapy). In this way, it may be more accessible for people with cognitive impairments as it is less cognitively demanding and an RCT has shown promising results among people with ID (Jahoda et al., 2017). The authors compared an adapted, manualised behavioural activation intervention with guided self-help (mainly psychoeducational) among 161 adults with mild-moderate ID and clinical depression. In behavioural activation, clients are facilitated in increasing their levels of activity and helped to address barriers (e.g. emotional or organisational) in engaging in activities. In guided self-help, clients are provided with self-help materials and some contact with therapists to help them to use those materials outside of clinic settings. The authors report for both groups significant reductions in depression scores and improvements in mental health and wellbeing measures at the end of treatment (four months after randomisations) that were maintained at 12-month follow-up with moderate to large effect sizes. Neither intervention was superior to the other and no serious adverse effects were reported. The authors reflect on the limitation of no treatment as usual arm in the study, but this was not possible as the trial funders insisted on an active treatment comparison arm.

This trial represents a significant advancement in the field as it is the largest trial of individual psychological therapy among people with ID. Its high quality design makes an important contribution to the evidence base and reinforces the idea that large-scale trials of psychological interventions among people with ID are possible. The interventions were provided by community nurses and allied health professionals working with people with ID who received 1–2 days training in the intervention indicating that psychological interventions can be successfully delivered by professional groups other than psychologists who are already working with people with ID and who receive some training. Furthermore, aspects of this type of intervention may be appropriate for those with more severe ID, something which future research could seek to investigate.

Cognitive Behavioural Therapy (CBT)

CBT is a collaborative talking therapy in which the client and therapist work together to identify relationships between thoughts, feelings and behaviours and attempt to moderate patterns in these systems to produce changes. A systematic review and meta-analysis by Koslowski et al. (2016) located four controlled studies that had evaluated CBT for depressive symptoms among people with mild and moderate ID, three of which used samples of adults with depressive symptoms only (Hassiotis et al., 2013; McCabe, McGillivray, & Newton, 2006; McGillivray, McCabe, & Kershaw, 2008) and the fourth was concerned with anger management (Willner et al., 2013). The meta-analysis of these four studies showed a moderate, but non-significant effect size of CBT on depressive symptoms, but with high levels of heterogeneity between studies. Similarly, an earlier meta-analysis by Vereenooghe and Langdon (2013) included the same three studies of CBT for depression and found a moderate to large, but non-significant effect size. They also comment on the high levels of between-study variance as each study employed different study designs.

Owing to the limited and varying evidence, Unwin, Tsimopoulou, Azmi, and Stenfert Kroese (2016) undertook a broad scoping review of CBT for anxiety or depression among adults, which included studies of any design and took a narrative approach to analysis. Unwin et al. located two additional studies which evaluated CBT for depression (Lindsay, 1999; McGillivray & Kershaw, 2015) and three additional studies (one of which resulted in two separate papers) which included mixed clinical presentations (including mood and emotional disorders such as anxiety and anger; Lindsay et al., 2015; Stenfert Kroese et al., 2014; Pert et al., 2013; Ghafoori, Ratanasiripong, & Holladay, 2010) Tables 45.1 and 45.2 present summaries of the eight studies.

The three RCTs including people with ID and depression report varying results with the trial by Hassiotis et al. (2013) reporting no overall treatment effect whereas the other two report significant treatment effects that were maintained at three month follow-up. Unwin et al. (2016) suggest that these discrepancies may be due to methodological differences, especially as Hassiotis et al. used a trans-diagnostic approach which may have diluted the effect of CBT: the authors found a trend towards treatment effect for those with clinical depression, but not for those with anxiety disorder. More recently, an RCT of computerised CBT for adults with mild and moderate ID and anxiety or depression has been published with the opposite results: significant treatment effects were observed on the primary outcome measure of anxiety, but not on depression (Cooney, Jackman, Coyle, & O'Reilly, 2017). The authors suggest that computerised CBT may not be a good match for people with depressive symptoms but also highlight that few participants presented with depression compared to anxiety symptoms resulting in low levels of depression across the sample and therefore loss of statistical power.

Unwin et al. (2016) considered qualitative studies in their review and suggest that results from these studies are more consistent and reflect a positive perception of CBT among clients and carers: 'participants especially valued the space and time to discuss their thoughts and feelings, the validation of their problems, and the formation or development of empathetic relationships with therapists and, in the case of group therapy, other group members' (pg. 69). CBT is a collaborative therapy and other research has shown that clients with ID adopt a collaborative rather than passive role in the therapeutic process, contributing to the analytic process and dialogues (Jahoda et al., 2009). Furthermore, in line with research with the general population, the importance of a supportive therapeutic relationship was emphasised in these studies. Therapists, working with people with ID, also view the therapeutic relationship as fundamental to their work, influential on outcome, and more important than theoretical

Table 45.1 Summary of studies of CBT for depression

First author (year)	Study design	Participants	Mental health condition	Intervention	Measures	Main findings
Lindsay (1999)	Quantitative Case series Pre, post and follow-up assessments	$N = 5$ (referred between 1990–1996) Age: not mentioned ID: not mentioned Country: USA	Depression	Individual adapted Beck's Cognitive Therapy (simplified form) Delivered by therapist	– BDI or ZDS – reported frequency or intensity of problematic cognitions	– BDI or ZDS reduced from around 50% of total score to around 25% (no statistical analyses due to small sample size) – Decrease remained at 6-month follow-up
McCabe (2006)	Quantitative RCT (quasi-independent) Pre, post and follow-up assessments ($n = 18$ for follow-up)	EG $N = 34$ (including 15 from CG) Mean age: 34 16 M/18F CG $N = 15$ Mean age: 40 6 M/9F ID: mild-moderate Country: Australia	Depression (at least minimal, as measured by the BDI-II, clinical and depressive symptoms)	EG Group CBT 5 weekly, 2-hour sessions Delivered by therapist CG Waitlist, received intervention after follow-up for EG complete	– BDI-II – SCS – ATQ-R – Rosenberg Self-esteem Scale (RSES)	– Significant difference between the groups at the three time points on the combined outcome measures ($p < 0.001$). - When the measures were considered separately, BDI-II ($p < 0.001$), SCS ($p < 0.001$), and ATQ-R ($p < 0.001$) reached significance. – No difference in RSES – Significant reduction in BDI-II scores and frequency of negative automatic thoughts, and increase in SCS scores (indicating that the participants viewed themselves more positively) from pre-test to post-test within the EG (p values not given) - Significant improvements remained at 3-month follow-up
McGillivray (2008)	Quantitative RCT (cluster) Pre, post and follow-up assessments ($n = 45$ for follow-up)	EG $N = 20$ Mean age: 38 13 M/7F CG $N = 27$ 7Mean age: 31 19 M/8 F ID: mild (IQ 50–70) Country: Australia	Depression (as measured by the BDI-II; 49% minimally depressed, 17% mildly depressed, 15% moderately depressed, 19% severely depressed	EG Manualised group ($n = 5$–6) CBT 12 weekly, 2-hour sessions Delivered by agency staff ($N = 13$) CG Waiting list, received the intervention after follow-up for EG complete	– BDI-II – ATQ-R – SCS – The Social Readjustment Rating Scale (SRRS; 10 items only)	– Significant group by time (pre-test to post-test) interaction effect on the combined outcome measures ($p < 0.001$). Significant main effect for time ($p < 0.001$), not group - Significant group by time interaction effect for BDI-II ($p = 0.000$), ATQ-R ($p = 0.021$) and SCS ($p = 0.015$) – No difference in SRRS – Significant decrease in BDI-II ($p < 0.01$) and ATQ-R ($p < 0.01$) scores within EG from pre-test to post-test – Positive effects on BDI-II and ATQ-R comparable across groups (once CG also received intervention) over time (pre-test, post-test, and follow-up) – Changes were maintained at 3-month follow-up

(continued)

Table 45.1 (continued)

First author (year)	Study design	Participants	Mental health condition	Intervention	Measures	Main findings
McGillivray (2015)	Quantitative Controlled trial Pre, post and follow-up assessments	CBTG $N = 23$ CTG $N = 23$ BTG $N = 24$ 42 M/28F Mean age: 36 ID: mild Country: Australia	$N = 24$ minimally depressed, $n = 28$ mildly depressed, $n = 13$ moderately depressed, $n = 5$ severely depressed according to BDI-II cut-offs	Manualised group CBT, CT or BT All 12 weekly, 1.5 hour sessions Groups of 7–8 All delivered by same therapist	– BDI-II – ATQ-R (10 most relevant items only)	– Significant main effect of time but not group on BDI-II ($p < .01$and $p = 0.98$ respectively) and ATQ-R ($p < 0.01$ and $p = 0.13$ respectively). – No significant interaction effect on BDI-II ($p = 0.61$) indicating that effect of time did not differ between groups – Significant interaction effect on ATQ-R ($p < 0.01$). Individual tests revealed a significant reduction in the CBT group pre- to post-test which was maintained at follow-up. No significant reductions for the CT or BT groups. – At post-intervention, 53%, 47%, and 71% indicated recovery (to no or minimal depression) from at least mild depression in the CBT, CT and BT groups respectively. – The proportion of recovered individuals was 53%, 53% and 47% at follow-up.

Reprinted from Research in Developmental Disabilities, 51–52, Unwin G, Tsimopoulou I, Stenfert Kroese B & Azmi S, Effectiveness of Cognitive Behavioural Therapy (CBT) programmes for anxiety or depression in adults with intellectual disabilities: A review of the literature, Pages 60–75, Copyright (2016), with permission from Elsevier

ID = level of intellectual disabilities; IPA = Interpretative phenomenological analysis; M = males; F = females; EG = Experimental Group; CG = Control Group; IPA = Interpretative Phenomenological Analysis; CBT = Cognitive Behavioural Therapy; CT = Cognitive Therapy; BT = Behavioural Therapy; TAU = Treatment As Usual; RCT = Randomised Controlled Trial

Scales: GAS-ID = Glasgow Anxiety Scale-for people with an Intellectual Disability; GDS = Glasgow Depression Scale; BAI = Beck Anxiety Inventory; ZUS = Zung Anxiety Scale; QOLS = Quality of Life Scale; HoNOS-LD = Health of the Nation Outcome Scale – Learning Disability Version; BDI = Beck Depression Inventory; ZDS = Zung Depression Scale; SCS = Social Comparison Scale; BSI = Brief Symptom Inventory; ATQ-R = Automatic Thoughts Questionnaire – Revised

Table 45.2 Summary of studies of CBT for mixed clinical presentations

First author (year)	Study design	Participants	Mental health condition	Intervention	Measures	Main findings
Ghafoori (2010)	Quantitative One-group Pre, post and follow-up assessments	$N = 8$ Mean age: 20 2 M/6F ID: mild-borderline Country: USA	Mood difficulties (depression, anxiety and anger)	Group trans-diagnostic CBT 9 weekly, 90-minute sessions Delivered by therapist	– Symptom Checklist 90-Revised (SCL-90)	– Significant decrease in global severity index ($p < 0.05$), and anxiety ($p < 0.05$), depression ($p < 0.05$), somatisation ($p < 0.05$), obsessive-compulsive ($p < 0.05$), interpersonal sensitivity ($p = 0.003$) and psychoticism ($p < 0.05$) subscale scores from pre-test to post-test all with large effect sizes (Cohen's d > 0.85) – No change in hostility, phobic anxiety and paranoid ideation – Significant decreases maintained at follow-up
Hassiotis (2013)	Mixed methods RCT Pre, post and follow-up assessments (content analysis for qualitative data)	EG $N = 16$ Interviews: $N = 13$ Mean age: 34 5 M/11F CG $N = 16$ Mean age: 38 7 M/9F ID: mild-moderate Country: UK	Mood disorder or symptoms of depression and/or anxiety (72% with depression with/out anxiety and 29% with anxiety only)	EG Manualised trans-diagnostic individual CBT 16 weekly, 1-h sessions Delivered by therapist Plus TAU CG TAU	– BDI-Youth – BAI-Youth – Manchester Short Assessment Of Quality of Life (MANSA) – Client Satisfaction Questionnaire (CSQ-8) – Client Service Receipt Inventory, adapted for the study (CSRI) – Interview schedule comprised 6 open ended questions	Quantitative: – No significant differences in interaction or main effects for the two groups – Participants in the intervention group presented a non-significant reduction only in depression scores (no matter if they also had anxiety or not) – CSQ-8 scores indicated the participants were highly satisfied with services – Participants were unable to complete the MANSA due to its complexity Qualitative: −8 gave examples where they had used CBT techniques including reports of improved confidence and increased calmness −7 felt that CBT had a positive impact on mood but did not resolve all their difficulties

(continued)

Table 45.2 (continued)

First author (year)	Study design	Participants	Mental health condition	Intervention	Measures	Main findings
						−6 would have liked the sessions to be free-floating rather than task orientated −7 participants were openly positive about their therapist −8 would recommend therapy to others – The role of support worker was viewed in a positive way −8 would have liked further contact after sessions −1 viewed homework tasks as persecutory by 1
Lindsay (2015)	Quantitative Matched controlled trial (quasi experimental) pre, post and follow-up assessments	EG $N = 12$ Mean age: 28.9 6 M/6F IQ: 62.4 CG (retrospective waitlist control) $N = 12$ Mean age: 33.1 6 M/6F IQ: 63.9 ID: mild Country: UK	Diagnosis of emotional disorder (anxiety $n = 8$, depression $n = 6$, mixed anxiety & depression $n = 7$, pathological jealousy $n = 1$, interpersonal conflict $n = 2$)	EG Manualised trans-diagnostic individual CBT Mean 10.75 (range: 8–14) sessions Delivered by therapist CG Waitlist, TAU (not including CBT)	– BSI (Anxiety, Depression and Global Severity Index) – GAS-ID (EG only) – GDS (EG only; self- and informant- report)	– Significant decrease in BSI Global Severity in EG compared with CG ($p = 0.001$) – Marginally significant decrease in BSI Anxiety in EG compared to CG ($p = 0.07$) – No treatment effect for BSI Depression – Within EG change in all BSI measures maintained at 6-month follow-up – Significant decreases in GAS-ID ($p < 0.01$) and GDS (self- and informant- report, $p < 0.01$ and $p < 0.05$ respectively) within EG pre- to post-test, changes maintained at 6-month follow-up.

Stenfert Kroese (2014)	Qualitative Interviews pre and after the first phase of therapy (thematic analysis)	Staff First interview *N* = 11 3 M/8F Support workers = 5, community nurses = 4, social worker = 1, supported employment worker = 1 Second interview *N* = 9 3 M/6F People with ID *N* = 15 (see 11b)	N/A	Individual CBT 9 sessions between interviews (first phase of CBT: Focused on engagement and socialising the client into the therapeutic model) Delivered by therapist	– Semi-structured interviews with paid support or professional key workers of people with ID who were participating in CBT	1st interview: – Most felt it was beneficial for the service user to talk to an outsider – Some felt that CBT offered nothing new beyond what had already been tried – The majority had poor understanding of CBT and had negative expectations of its effectiveness Second interview: –9 had observed positive outcomes including improved mental health (e.g. more relaxed, more sociable, more confident, improved self-esteem) and improved cognitive skills/processing (e.g. better insight into problems and better problem solving skills). All of these participants identified at least 1 aspect of the therapist's approach that had benefitted (e.g. good liaison, teaching appropriate coping strategies, giving emotional support and normalising the problem) –1 felt that the therapist functioned as a threat rather than a positive influence –1 felt that the changes were due to medication –1 felt that CBT was not helpful – A number said that service users were easier to talk to – Most expressed doubt that changes would be permanent and felt that long-term sessions or 'top-ups' would be required

(continued)

Table 45.2 (continued)

First author (year)	Study design	Participants	Mental health condition	Intervention	Measures	Main findings
Pert (2013)	Qualitative 2 interviews in the initial stage of therapy (IPA)	N = 15 (22 recruited to take part in therapy) Mean age: 39 8 M/7F ID: borderline-mild (IQ 55–79) Country: UK	Referred for anxiety (4), depression (4), anger (4), Mixed presentation (3)	Individual CBT 10 weekly/ fortnightly, one-hour sessions Delivered by therapist	– Semi-structured interview with client participant – Interview 1 between fourth and fifth session; interview 2 between ninth and tenth session)	(transcripts from the two interviews were analysed together) – All participants found talking in sessions about problems helpful in itself, including a sense of relief and immediate benefit – Participants identified positive changes that they attributed to therapy, referring to improved self-perceptions, feeling more confident, understanding and expressing problems better, getting more out of life, being more sociable, improved relationships – Many valued having an identified person (who was not a carer) with whom they could talk – A number valued the opportunity to talk without their carers in a confidential and private manner – Talking was difficult at first – Participants appreciated the thorough approach of therapists and that their problems were being taken seriously—this made them feel valued and their problems validated – Participants benefitted from an approachable and empathetic therapeutic style – Participants expressed cautious optimism about changes which may be fragile and short-lived, and many felt they required longer-term input

Reprinted from Research in Developmental Disabilities, 51–52, Unwin G, Tsimopoulou I, Stenfert Kroese B & Azmi S, Effectiveness of Cognitive Behavioural Therapy (CBT) programmes for anxiety or depression in adults with intellectual disabilities: A review of the literature, Pages 60–75, Copyright (2016), with permission from Elsevier

ID = level of intellectual disabilities; IPA = Interpretative phenomenological analysis; M = males; F = females; EG = Experimental Group; CG = Control Group; IPA = Interpretative Phenomenological Analysis; CBT = Cognitive Behavioural Therapy; CT = Cognitive Therapy; BT = Behavioural Therapy; TAU = Treatment As Usual; RCT = Randomised Controlled Trial

Scales: GAS-ID = Glasgow Anxiety Scale-for people with an Intellectual Disability; GDS = Glasgow Depression Scale; BAI = Beck Anxiety Inventory; ZUS = Zung Anxiety Scale; QOLS = Quality of Life Scale; HoNOS-LD = Health of the Nation Outcome Scale – Learning Disability Version; BDI = Beck Depression Inventory; ZDS = Zung Depression Scale; SCS = Social Comparison Scale; BSI = Brief Symptom Inventory; ATQ-R = Automatic Thoughts Questionnaire – Revised approaches (Jones, 2014). Therapeutic relationships may be especially important for people with ID due to more limited opportunities to develop positive relationships, however, research has not yet investigated the association between quality of relationship and outcome. In the study by Hassiotis et al. (2013), CBT was delivered by CBT therapists who were not specialists in ID, and Unwin et al. (2016) suggest that this lack of experience of working and engaging with people with ID may have affected the relationship and subsequently the outcome.

The review authors comment on the methodological limitations of the studies including small sample sizes, few control groups, short-term follow-ups, and lack of treatment fidelity assessment (Unwin et al., 2016). Furthermore, issues in the choice of outcome measure persist (such as non-validated measures or those designed for the general population) and Unwin et al. recommend that researchers should move beyond symptom measurement to use more holistic, personally-meaningful measures which reflect the client's aims for therapy. Attempting to detect changes in symptoms as a result of psychological therapy may be compounded by the aims of therapy which often seeks to improve emotional literacy (understanding, identification and expression of emotions) as a first goal of therapy. Thus, self-report of symptoms could improve and therefore result in an increase in scores on symptom measures.

There is emerging evidence to suggest that CBT for depression among people with mild and moderate ID is feasible, well tolerated, and effective at reducing symptoms of depression, including negative automatic thoughts. There is currently no evidence for people with severe ID and CBT may not be suited due to language impairments and high support needs (Sturmey, 2004). Other psychological approaches may be more appropriate such as behavioural activation.

Psychoanalysis, Psychoanalytic and Psychodynamic Psychotherapy

Psychodynamic therapy for people with ID remains one of the least well investigated, but long-established psychological therapies (Flynn, 2012). The development of adapted manuals of psychotherapy for people with ID needed for trials has been slow (Shepherd & Beail, 2017). However, in the general population, there is evidence of efficacy for depression (Shedler, 2010). A systematic review found 13 papers which evaluated the effectiveness of psychoanalytic and psychodynamic psychotherapy for adults with ID; no papers were located for psychoanalysis (Shepherd & Beail, 2017). Two papers included samples with mixed clinical presentations that included depression (Bichard, Sinason, & Usiskin, 1996; Carlsson, 2000). Both of these studies are case series of individual therapy and include 4 and 11 participants with mild-moderate ID but it is not clear how many participants had a diagnosis of depression. Neither study reports on depressive symptoms, but improvements were observed in tests of specific psychoanalytical concepts such as increased integration and decreased primitive defensive functioning.

More recently, Statham & Beail (2018) investigated client views of the accessibility, acceptability and effectiveness of psychodynamic psychotherapy through interviews with 10 participants with mild ID and difficulties including anger, trauma, low mood and interpersonal difficulties. As with similar research on CBT, participants appreciated being listened to but were unhappy about therapy ending and struggled to generalise what they learnt in therapy to other settings and with others. Some participants were surprised by the content of sessions and some felt anxious when travelling to sessions. However, participants were able to identify ways in which therapy had helped.

'New Wave' Psychological Interventions

Mindfulness and Acceptance-Based Therapies

Mindfulness approaches aim to help people focus on the present moment, including what is happening in mind, body and the world around them. It "relies more on reducing experiential avoidance by enabling individuals to become aware of and normalise emotions and bodily sensations" (pg. 95), rather than challenging thoughts, and beliefs, and this process relies less on verbal exchange between therapist and client which may be more suited to people with problems with communication (Idusohan-Moizer, Sawicka, Dendle, & Albany, 2015). Two structured, mindfulness-based intervention packages have been developed: mindfulness-based stress reduction (Kabat-Zinn, 1990) and mindfulness-based cognitive therapy (Segal, Williams, & Teasdale, 2002). There is evidence to suggest that mindfulness is useful for a range of psychological conditions among the general population, especially recurrent depressive episodes (Piet & Hougaard, 2011), but mindfulness has not been widely used with people with ID (Chapman et al., 2013). A review found evidence of improvements in aggression for people with ID, but did not locate any studies specifically on depression (Chapman et al., 2013). Subsequently, in a small study, Idusohan-Moizer and colleagues (2015) adapted mindfulness-based cognitive therapy for people with ID and incorporated aspects of acceptance and compassion-focused therapy. They evaluated this therapy among 15 participants with recurrent episodes of anxiety or depression and borderline to moderate ID. None of the participants had depression in isolation and only four had depression and anxiety; the majority had anxiety. Depressive symptoms and compassion towards the self significantly improved after the intervention and these effects were maintained at 6-week follow-up; however, there was no control group in this study. Other research has focused on mindfulness for other problems in adults with ID with limited, but positive and promising results (see Pert, 2017 for a review).

Solution-Focused Therapy

Solution-focused brief therapy is a short-term, client-directed and goal-focused approach, originally developed by de Shazer and colleagues (De Shazer, 1985). Central is the idea that clients should be supported to reach their goals by developing solutions rather than analysing problems (de Shazer et al., 1986). Researchers have recognised the potential of solution-focused therapy for people with ID due to its focus on empowerment, strengths and competencies (Carrick & Randle-Phillips, 2018; Roeden, Bannink, Maaskant, & Curfs, 2009). Furthermore, through solution-focused coaching or consultation, there is the potential to work with family and care staff on their competencies to achieve their goals in working with/caring for a person with ID (Roeden, Maaskant, Bannink, & Curfs, 2012). A meta-analysis found support for solution-focused brief therapy for depression among the general population (Gingerich & Peterson, 2013). More recently, a systematic review of solution-focused approaches for people with ID found 12 papers, of which 5 included participants referred for treatment of depression (Carrick & Randle-Phillips, 2018: Stoddart, McDonnell, Temple, & Mustata, 2001: 4/16 participants; Roeden, Maaskant, Bannink, & Curfs, 2011: 1/10 participants; Roeden, Maaskant, & Curfs, 2014: 2/38 participants; Lloyd & Dallos, 2006, 2008: 3/7 participants). The authors conclude that there is preliminary evidence for effectiveness, primarily from case studies. None of the reviewed studies used measures of depression and, owing to the small proportion of participants referred for depression in each study, it is hard to extrapolate the results.

Dialectical Behavioural Therapy

Dialectical behaviour therapy (DBT) was developed for people with borderline personality disorder and high-risk suicidal behaviour (Linehan, 1993) and has subsequently been adapted for use with older adults with depression (Lynch, Morse, Mendelson, & Robins, 2003). It consists of weekly, individual sessions, group skills training and access to telephone coaching (Linehan). In the field of ID, interest has grown in terms of its use for people with poor impulse control or difficulty with emotion regulation, however, few studies implement all three aspects of traditional DBT (Crossland, Hewitt, & Walden, 2017) and none have evaluated DBT specifically for depression. A brief outline of group DBT skills training among seven people with ID does report decreases in depression measures, but very little information is provided (Hall, Bork, Craven, & Woodrow, 2013). Another study reports significant decreases in depression scores following DBT among 18 participants with ID and difficulties relating to emotion regulation, managing crises and interpersonal effectiveness (McNair, 2014).

Other Interventions

A variety of other interventions are used to treat depression. Among the general population, exercise interventions have been recognised for their antidepressant effect. Given the health inequalities experienced by people with ID, exercise interventions may be especially helpful in improving both physical and mental health. A small-scale RCT evaluated a 12-week exercise intervention among 27 participants with mild-moderate ID with promising results (Carraro & Gobbi, 2014). The authors report a significant reduction in depressive symptoms in the exercise group compared with the control group (who received painting classes). However, the participants were not diagnosed with depressive disorder and there was no long-term follow-up. Other treatments for depression include bright light therapy, particularly or seasonal affective disorder. Hermans, Soerokromo, and Evenhuis (2017) evaluated bright light therapy among 14 adults with moderate, severe or profound ID and depressive symptoms. They report some positive effects and suggest that bright light therapy was well tolerated by participants with no adverse effects; however, there are practical issues in using this type of therapy which require the participant to remain still for a period of time at the same time each day (preferably upon getting up).

Electroconvulsive therapy (ECT) is rarely used for people with ID but it is still available as a treatment option for severe depression (Osugo & Cooper, 2016) despite concern over adverse effects, the invasive nature of this procedure, and problems gaining fully informed consent (for example, assurance that people with ID can weigh up the risk: benefit profile of the intervention and issues with acquiescence). Osugo and Cooper did not locate any studies involving 20 or more participants (and at least 50% with mild ID) that evaluated ECT in their review. Despite this, ECT is still recommended for severe psychotic depression or serious suicide risk in people with ID, after unsuccessful medication trials (Bhaumik et al., 2015). Another review of ECT for people with ID located 72 case reports, of which almost 58% had an affective disorder (Collins, Halder, & Chaudhry, 2012). The authors conclude that ECT had few adverse effects; however, there are problems with positive reporting bias in case studies. Short-term adverse effects of ECT include headache, aching muscles and risk of the anaesthetic. Longer-term risks include problems with memory (Barnes, 2010) which is especially problematic for people with existing cognitive impairments. Other uncommon treatments include repeated transcranial magnetic stimulation, transcranial direct current stimulation and vagus nerve stimulation but they are not recommended as first line interventions (even in the general population), are invasive procedures, and are not evidence-based in ID.

The herbal remedy ‘St. John’s Wort’ is purported to be useful in low mood, anxiety and premenstrual stress. Despite being available over-the-counter, it can have serious interactions with other medications. This includes interactions with antidepressants, strong painkillers and the oral contraceptive pill. It can also reduce the effectiveness of some anti-epileptic drugs, digoxin and warfarin (Werneke, 2010).

Conclusions

Depression in people with ID is likely to be more common than in the general population but is currently underdiagnosed. It is therefore likely to be inappropriately treated as treatment may focus on behaviours that challenge. This is also reflected in the evidence base which is much more limited than for behaviours that challenge; however, some studies which focus on treatment of behaviour may also include measures of depression and therefore can be used to supplement the evidence. For example, a CBT-based anger management intervention showed positive effects on depressive symptoms (Willner et al., 2013). However, other research on trans-diagnostic CBT have mixed results. Future research should ensure that adequate resources are afforded to assessment and detection of depression to facilitate intervention studies.

There are a range of interventions available for depression in people with ID, however, evidence pertaining to their effectiveness remains limited. Much evidence comes from practice-based research (for example, case studies) which provides important, ecologically valid evidence about effectiveness, but more large-scale trials are required to establish the efficacy of both psychological and biological interventions. Furthermore, systematic, qualitative evaluation of the process of providing interventions is warranted to ascertain acceptability of interventions from the perspectives of those who receive, those who provide and those around the person. These studies can also investigate the components of interventions, especially psychological interventions, in an attempt to determine mechanisms of action. Further research should also compare credible treatment options, including psychological interventions and medication.

Currently, CBT is the most evidence-based intervention option for those with mild or moderate ID, however, interest in other therapeutic modalities is growing and subsequently the evidence base will continue to expand. There is limited evidence to support the use of antidepressants in people with ID, unless evidence is extrapolated from the general population. The evidence for people with ID raises concern over adverse effects, especially given the poorer physical health of people with ID and problems in reporting adverse effects. As with the general population, it is appropriate that minimally invasive treatment options are prioritised.

Almost all the research in the field is with people with mild or moderate ID. There is scant research which involves people with severe ID, most likely owing to the ethical complexities of involving adults who may not have capacity to consent in intervention trials. This is especially problematic in relation to psychological interventions as further adaptation of therapies may be required to make them suited to people with more limited cognitive and verbal abilities. Indeed, people with moderate or severe ID may need a more tailored, one-to-one approach to psychological therapy which is not easy to evaluate in trials which require consistency in intervention. Furthermore, assessing symptoms of depression in intervention studies are susceptible to the same issues as diagnosis of depression in people who are unable to self-report on internal emotional states. As a result, treatment options outside of antidepressants are very limited for those with severe or profound ID. The only study in the present chapter to include people with severe ID evaluated bright light therapy. This is an initial study to assess acceptability rather than effectiveness but it is representative of the diversification of interventions being made available and the strive for equity in healthcare for people with ID. Improving the involvement of people with moderate and severe ID in research remains a challenge to be met by researchers.

References

American Psychiatric Association (APA). (1994). *Diagnostic and statistical manual of mental disorder* (4th ed.). Arlington, VA: American Psychiatric Publishing.

American Psychiatric Association (2010). *Practice guideline for the treatment of patients with major depressive disorder*. Retrieved April 16, 2018, from www.psychiatryonline.org.

American Psychiatric Association (APA). (2013). *Diagnostic and statistical manual of mental disorder* (5th ed.). Arlington, VA: American Psychiatric Publishing.

Azam, K., Sinai, A., & Hassiotis, A. (2009). Mental ill-health in adults with learning disabilities. *Psychiatry, 8*(10), 376–381. https://doi.org/10.1192/bjp.bp.106.022483

Barnes, R. (2010). *Information on ECT*. London: Royal College of Psychiatrists.

Beail, N. (2016). *Psychological therapies for people who have intellectual disabilities*. Leicester: Bristish Psychological Society.

Berney, T., & Deb, S. (2012). Epilepsy in learning disability. In S. Shorvon, R. Guerrini, M. Cook, & S. Lahtoo (Eds.), *Oxford textbook of epilepsy and epileptic seizures: Oxford textbooks in clinical neurology* (pp. 195–199). Oxford: Oxford University Press.

Bhaumik, S., Branford, D., Barrett, M., et al. (2015). *Frith guidelines for prescribing in intellectual disability*. Wiley Press.

Bhaumik, S., Collacott, R. A., Gandhi, D., Duggirala, C., & Wildgust, H. J. (1995). A naturalistic study in the use of antidepressants in adults with learning disabilities and affective disorders. *Human Psychopharmacology, 10*, 283–288.

Bhaumik, S., Tyrer, F. C., McGrother, C., & Ganghadaran, S. K. (2008). Psychiatric service use and psychiatric disorders in adults with intellectual disability. *Journal of Intellectual Disability Research, 52*(2), 986–995.

Bhaumik, S., & Wildgust, H. J. (1996). Treatment outcomes including withdrawal phenomenon with fluoxetine and paroxetine in patients with learning disabilities suffering from affective disorders. *Human Psychopharmacology: Clinical and Experimental, 11*, 337–338.

Bichard, S., Sinason, V., & Usiskin, J. (1996). Measuring the change in mentally retarded clients in long-term psychoanalytic psychotherapy. The draw-a-person test. *NADD Newsletter, 13*, 6–11.

Bramston, P., & Fogarty, G. (2000). The assessment of emotional distress experienced by people with an intellectual disability: A study of different methodologies. *Research in Developmental Disabilities, 21*(6), 487–500.

Campbell, M., Robertson, A., & Jahoda, A. (2014). Psychological therapies for people with intellectual disabilities: Comments on a matrix of evidence for interventions in challenging behaviour. *Journal of Intellectual Disability Research, 58*(2), 172–188. https://doi.org/10.1111/j.13652788.2012.01646.x

Carlsson, B. (2000). Psychoanalytic psychotherapy with intellectually disabled adults –evaluation by using projective tests: A collaboration project between a psychiatric clinic and the services for mentally handicapped. *NADD Bulletin, 3*(5). Article 3.

Carraro, A., & Gobbi, E. (2014). Exercise intervention to reduce depressive symptoms in adults with intellectual disabilities. *Perceptual & Motor Skills, 119*, 1–5.

Carrick, H., & Randle-Phillips, C. (2018). Solution-focused approaches in the context of people with intellectual disabilities: A critical review. *Journal of Mental Health. Research in Intellectual Disabilities, 11*(1), 30–53. https://doi.org/10.1080/19315864.2017.1390711

Chapman, M. J., Hare, D. J., Caton, S., Donalds, D., McInnis, E., & Mitchell, D. (2013). The use of mindfulness with people with intellectual disabilities: A systematic review and narrative analysis. *Mindfulness, 4*, 179.

Charlot, L. R., Bensons, B. A., Fox, S., Tasse, M., Hassiotis, A., & Pary, R. J. (2016). Depressive disorders. In R. J. Fletcher, J. Barnhill, & S.-A. Cooper (Eds.), *Diagnostic manual-intellectual disability: A textbook of diagnosis of mental disorders in persons with intellectual disability: 2nd edition (DM-ID2)* (pp. 265–302). New York: NADD Press.

Charlot, L. R., Tasse, M., Fox, S., Pary, R. J., Benson, B. A., & Hassiotis, A. (2017). Chapter 12: Depressive disorders. In R. J. Fletcher, J. Barnhill, & S.-A. Cooper (Eds.), *Diagnostic manual–intellectual disability (DM-ID-2): A textbook of diagnosis of mental disorders in persons with intellectual disability*. Kingston, NY: National Association for the Dually Diagnosed (NADD).

Cleare, A., Pariante, C. M., Young, A. H., Anderson, I. M., Christmas, D., Cowen, P. J., et al. (2015). Evidence-based guidelines for treating depressive disorders with antidepressants: A revision of the 2008 British Association for Psychopharmacology guidelines. *Journal of Psychopharmacology, 29*(5), 459–525.

Collins, J., Halder, N., & Chaudhry, N. (2012). Use of ECT in patients with an intellectual disability: Review. *Psychiatric Bulletin, 36*, 55–60.

Collishaw, S., Maughan, B., & Pickles, A. (2004). Affective problems in adults with mild learning disability: The roles of social disadvantage and ill health. *British Journal of Psychiatry, 185*, 350–351.

Cooney, P., Jackman, C., Coyle, D., & O'Reilly, G. (2017). Computerised cognitive-behavioural therapy for adults with intellectual disability: Randomised controlled trial. *British Journal of Psychiatry, 211*, 95–102. https://doi.org/10.1192/bjp.bp.117.198630

Cooper, S. (1997). Epidemiology of psychiatric disorders in elderly compared with younger adults with learning disabilities. *British Journal of Psychiatry, 170*(4), 375–380. https://doi.org/10.1192/bjp.170.4.375

Cooper, S. A., Smiley, E., Morrison, J., Williamson, A., & Allan, L. (2007). Mental ill-health in adults with intellectual disabilities: Prevalence and associated factors. *British Journal of Psychiatry, 190*, 27–35.

Crossland, T., Hewitt, O., & Walden, S. (2017). Outcomes and experiences of an adapted dialectical behaviour therapy skills training group for people with intellectual disabilities. *British Journal of Learning Disabilities, 45*, 208–216. https://doi.org/10.1111/bld.12194

Cuthill, F. M., Espie, C. A., & Cooper, S.-A. (2003). Development and psychometric properties of the Glasgow depression scale for people with a learning disability: Individual and carer supplement versions. *British Journal of Psychiatry, 182*, 347–353.

De Shazer, S. (1985). *Keys to solution in brief therapy*. New York, NY: Norton.

De Shazer, S., Berg, I. K., Lipchik, E., Nunnally, E., Molnar, A., Gingerich, W., & Weiner Davis, M. (1986). Brief therapy: Focused solution development. *Family Process, 25*(2), 207–221. https://doi.org/10.1111/j.1545-5300.1986.00207.x

Deb, S. (2007a). Epilepsy in people with mental retardation. In J. W. Jacobson & J. A. Maulick (Eds.), *Handbook of mental retardation and developmental disabilities* (pp. 81–96). New York: Kluwer Academic Publishers.

Deb, S. (2007b). Mental health and epilepsy among adults with intellectual disabilities. In N. Bouras & G. Holt (Eds.), *Psychiatric and behavioural disorders in intellectual and developmental disabilities* (pp. 238–251). Cambridge: Cambridge University Press.

Deb, S. (2016). Psychopharmacology. In N. N. Singh (Ed.), *Handbook of evidence-based practices in intellectual and developmental disabilities, evidence-based practices in behavioral health* (pp. 347–381). Cham, Switzerland: Springer International Publishing.

Deb, S., Unwin, G., & Deb, T. (2015). Trajectory of psychotropic medication use and factors influencing antipsychotic medication prescribing among adults with intellectual disabilities who show aggressive behaviour. *Journal of Intellectual Disability Research, 59*(1), 11–25.

Deb, S., Unwin, G. L., Rojahn, J., & Cooper, S.-A. (2018). Problem behaviours. In M. O. Bertelli, S. Deb, K. Munir, A. Hassiotis, & L. Salvador-Carulla (Eds.), *Textbook of psychiatry for intellectual disability and autism*. Cham: Springer International Publishing. Switzerland & World Psychiatric Association, Geneva, Switzerland (in press).

Doan, T. N., Lennox, N. G., Taylor-Gomez, M., & Ware, R. S. (2013). Medication use among Australian adults with intellectual disability in primary healthcare settings: A cross-sectional study. *Journal of Intellectual and Developmental Disability, 38*(2), 177–181. https://doi.org/10.3109/13668250.2013.778968

Ekers, D., Richards, D., McMillan, D., Bland, J. M., & Gilbody, S. (2011). Behavioural activation delivered by the non-specialist: Phase II randomised controlled trial. *British Journal of Psychiatry, 198*, 66–72.

Esbensen, A. J., & Benson, B. A. (2006). A prospective analysis of life events, problem behaviors and depression in adults with intellectual disability. *Journal of Intellectual Disability Research, 50*, 248–258.

Fletcher, R. J., Barnhill, J., & Cooper, S.-A. (2017). *Diagnostic manual–intellectual disability (DM-ID-2): A textbook of diagnosis of mental disorders in persons with intellectual disability*. Kingston, NY: National Association for the Dually Diagnosed (NADD).

Flynn, A. (2012). Fact or faith? *Current Opinion in Psychiatry, 25*, 342–347. https://doi.org/10.1097/YCO.0b013e328355e196

Ghafoori, B., Ratanasiripong, P., & Holladay, C. (2010). Cognitive behavioral group therapy for mood management in individuals with intellectual disabilities: A pilot study. *Journal of Mental Health Research in Intellectual Disabilities, 3*(1), 1–15. https://doi.org/10.1080/19315860903326079

Gingerich, W., & Peterson, L. (2013). Effectiveness of solution-focused brief therapy: A systematic qualitative review of controlled outcome studies. *Research on Social Work Practice, 23*(3), 266–283.

Hall, L., Bork, N., Craven, S., & Woodrow, C. (2013). People with learning disabilities experiences of a dialectical behaviour therapy skills group: A thematic analysis. *Clinical Psychology & People with Learning Disabilities, 11*, 7–11.

Hassiotis, A., Serfaty, M., Azam, K., Strydom, A., Blizard, R., Romeo, R., et al. (2013). Manualised individual cognitive behavioural therapy for mood disorders in people with mild to moderate intellectual disability: A feasibility randomised controlled trial. *Journal of Affective Disorders, 151*, 186–195.

Hassiotis, A., Stueber, K., Thomas, B., & Charlot, L. (2014). Mood and anxiety disorders. In E. Tsakanikas & J. McCarthy (Eds.), *Handbook of psychopathology in intellectual disability: Research, practice, and policy, autism and child psychopathology series* (pp. 161–175). New York: Springer.

Hatton, C., & Emerson, E. (2004). The relationship between life events and psychopathology amongst children with intellectual disabilities. *Journal of Applied Research in Intellectual Disabilities, 17*, 109–117.

Hemmings, C., Deb, S., Chaplin, E., Hardy, S., & Mukherjee, R. (2013). Review of research for people with ID and mental health problems: A view from the United Kingdom. *Journal of Mental Health Research in Intellectual Disabilities, 6*(2), 127–158.

Hermans, H., & Evenhuis, H. M. (2013). Factors associated with depression and anxiety in older adults with intellectual disabilities: Results of the healthy ageing and intellectual disabilities study. *International Journal of Geriatric Psychiatry, 28*(7), 691–699.

Hermans, H., Soerokromo, N., & Evenhuis, H. (2017). The applicability of bright light therapy in adults with moderate, severe or profound intellectual disabilities: A brief report. *Journal of Intellectual Disability Research, 61*, 618–623. https://doi.org/10.1111/jir.12365

Holden, B., & Gitlesen, J. P. (2004). The association between severity of intellectual disability and psychiatric symptomology. *Journal of Intellectual Disability Research, 48*(6), 556–562.

Hulbert-Williams, L., & Hastings, R. P. (2008). Life events as a risk factor for psychological problems in individuals with intellectual disabilities: A critical review. *Journal of Intellectual Disability Research, 52*(2), 883–895.

Hurley, A. D. (2008). Depression in adults with intellectual disability: Symptoms and challenging behaviour. *Journal of Intellectual Disability Research, 52*(11), 905–916.

Idusohan-Moizer, H., Sawicka, A., Dendle, J., & Albany, M. (2015). Mindfulness based cognitive therapy for adults with intellectual disabilities: An evaluation of the effectiveness of mindfulness in reducing symptoms of depression and anxiety. *Journal of Intellectual Disability Research, 59*(2), 93–104. https://doi.org/10.1111/jir.12082

Irvine, M., & Beail, N. (2016). Chapter 1: Identifying and meeting the emotional and mental health needs of people who have intellectual disabilities through psychological therapies. In N. Beail (Ed.), *Psychological therapies for people who have intellectual disabilities*. Leicester: British Psychological Society.

Jahoda, A., Selkirk, M., Trower, P., Pert, C., Kroese, B. S., Dagnan, D., & Burford, B. (2009). The balance of power in therapeutic interactions with individuals who have intellectual disabilities. *British Journal of Clinical Psychology, 48*(1), 63–77. https://doi.org/10.1348/014466508X360746

Jahoda, A., Stenfert Kroese, B., & Pert, C. (2017). *Cognitive behaviour therapy for people with intellectual disabilities –thinking creatively*. London: Palgrave Macmillan.

Jones, R. A. (2014). Therapeutic relationships with individuals with learning disabilities: A qualitative study of the counselling psychologists' experience. *British Journal of Learning Disabilities, 42*(3), 193–203. https://doi.org/10.1111/bld.12028

Kabat-Zinn, J. (1990). *Full catastrophe living: Using the wisdom of your body and mind to face stress, pain and illness*. New York: Delacorte.

Kannabiran, M., & McCarthy, J. (2009). The mental health needs of people with autism spectrum disorders. *Psychiatry, 8*, 398–401.

Koslowski, N., Klein, K., Arnold., K., Kösters, M., Schützwohl, M., Salize, H. J. & Puschner, B. (2016). Effectiveness of interventions for adults with mild to moderate intellectual disabilities and mental health problems: Systematic review and meta-analysis. British Journal of Psychiatry, 209: 469–474.

Kwok, H., & Cheung, P. (2007). Co-morbidity of psychiatric disorder and medical illness in people with intellectual disabilities. *Current Opinion in Psychiatry, 20*(5), 443–449.

Levitas, A., Finucane, B., Simon, E. W., Schuster, M., Kates, W. R., Olszewski, A. K., … Danker, N. (2016). Behavioral phenotypes of neurodevelopmental disorders. In R. J. Fletcher, J. Barnhill, & S.-A. Cooper (Eds.), *Diagnostic manual-intellectual disability: A textbook of diagnosis of mental disorders in persons with intellectual disability: 2nd edition (DM-ID2)* (pp. 35–74). New York: NADD Press.

Leyin, A. (2011). Improving access to psychological therapies for people with learning disabilities. *Tizard Learning Disability Review, 16*, 29–37.

Lindsay, W. (1999). Cognitive therapy. *The Psychologist, 12*, 238–241.

Lindsay, W. R., Tinsley, S., Beail, N., Hastings, R. P., Jahoda, A., Taylor, J. L., & Hatton, C. (2015). A preliminary controlled trial of a trans-diagnostic programme for cognitive behaviour therapy with adults with intellectual disability. *Journal of Intellectual Disability Research, 59*(4), 360–369. https://doi.org/10.1111/jir.12145

Linehan, M. (1993). Dialectical behavior therapy for treatment of borderline personality disorder: Implications for the treatment of substance abuse. *NIDA Research Monograph, 137*, 201–201.

Lloyd, H., & Dallos, R. (2006). Solution-focused brief therapy with families who have a child with intellectual disabilities: A description of the content of initial sessions and the processes. *Clinical Child Psychology and Psychiatry, 11*, 367–386. https://doi.org/10.1177/1359104506064982

Lloyd, H., & Dallos, R. (2008). First session solution-focused brief therapy with families who have a child with severe intellectual disabilities: Mothers' experiences and views. *Journal of Family Therapy, 30*, 5–28. https://doi.org/10.1111/j.1467-6427.2008.00413.x

Lunsky, Y. (2004). Suicidality in a clinical and community sample of adults with mental retardation. *Research in Developmental Disabilities, 25*, 231–243.

Lynch, T. R., Morse, J. Q., Mendelson, T., & Robins, C. J. (2003). Dialectical behavior therapy for depressed older adults: A randomized pilot study. *The American Journal of Geriatric Psychiatry, 11*(1), 33–45.

McCabe, M. P., McGillivray, J. A., & Newton, D. C. (2006). Effectiveness of treatment programmes for depression among adults with mild/moderate intellectual disability. *Journal of Intellectual Disability Research, 50*, 239–247.

McGillivray, J. A., & Kershaw, M. (2015). Do we need both cognitive and behavioural components in interventions for depressed mood in people with mild intellectual disability? *Journal of Intellectual Disability Research, 59*, 105–115.

McGillivray, J. A., & McCabe, M. P. (2007). Early detection of depression and associated risk factors in adults with mild/moderate intellectual disability. *Research in Developmental Disabilities, 28*, 59–70.

McGillivray, J. A., McCabe, M. P., & Kershaw, M. M. (2008). Depression in people with intellectual disability: An evaluation of a staff-administered treatment program. *Research in Developmental Disabilities, 29*, 524–536.

McNair, L. C. (2014). *Dialectical behaviour therapy for adults with intellectual disabilities*, PhD Theses. Retrieved May 1, 2018, from https://www.escholar.manchester.ac.uk.

Meins, W. (1996). Psychopharmacological treatment of mentally retarded adults with depressive disorders, cited in Verhoeven W. M. A., Veendrik-Meekes M. J., Jacobs G. A. J., van den Berg Y. W. M. M. & Tuinier B. S. (2001). Citalopram in mentally retarded patients with depression: a long-term clinical investigation. *European Psychiatry, 16*, 104–8.

Melville, C. A., Cooper, S. A., Morrison, J., Smiley, E., Allan, L., Jackson, A., … Mantry, D. (2008). The prevalence and incidence of mental ill-health in adults with autism and intellectual disabilities. *Journal of Autism and Developmental Disorders, 38*, 1676–1688.

Mileviciute, I., & Hartley, S. L. (2015). Self-reported versus informant-reported depressive symptoms in adults with mild intellectual disability. *Journal of Intellectual Disability Research, 59*(2), 158–169.

National Institute of Clinical and Health Excellence (NICE). (2016). *Mental health problems in people with learning disabilities: Prevention, assessment and management: NICE guideline*. London: Author.

NICE (2009). *Depression: The treatment and management of depression in adults (update)*. Retrieved April 1, 2018, from http://www.nice.org.uk/CG90.

Osugo, M., & Cooper, S.-A. (2016). Interventions for adults with mild intellectual disabilities and mental ill-health: A systematic review. *Journal of Intellectual Disability Research, 60*(6), 615–622.

Pert, C. (2017). Chapter 9: Mindfulness and third wave therapies. In A. Jahoda, B. S. Kroese, & C. Pert (Eds.), *Cognitive behaviour therapy for people with intellectual disabilities–thinking creatively*. London: Palgrave McMillan.

Pert, C., Jahoda, A., Stenfert Kroese, B., Trower, P., Dagnan, D., & Selkirk, M. (2013). Cognitive behavioural therapy from the perspective of clients with mild intellectual disabilities: A qualitative investigation of process issues. *Journal of Intellectual*

Disability Research, 57(4), 359–369. https://doi.org/10.1111/j.1365-2788.2012.01546.x

Piet, J., & Hougaard, E. (2011). The effect of mindfulness based cognitive therapy for prevention of relapse in recurrent major depressive disorder: A systematic review and meta-analysis. *Clinical Psychology Review, 31*, 1032–1040.

Reid, K. A., Smiley, E., & Cooper, S. A. (2011). Prevalence and associations of anxiety disorders in adults with intellectual disabilities. *Journal of Intellectual Disability Research, 55*(2), 172–181. https://doi.org/10.1111/j.1365-2788.2010.01360.x

Reiss, S., & Szyszanko, J. (1993). Diagnostic overshadowing and professional experience with mentally retarded person. *American Journal of Mental Deficiency, 47*, 415–421.

Richards, D. A., Ekers, D., McMillan, D., et al. (2016). Cost and outcome of behavioural activation versus cognitive behavioural therapy for depression (COBRA): A randomised, controlled, non-inferiority trial. *Lancet, 388*, 871–880.

Richards, M., Maughan, B., Hardy, R., Hall, I., Strydom, A., & Wadsworth, M. (2001). Long-term affective disorder in people with mild learning disability. *British Journal of Psychiatry, 179*(6), 523–527.

Roeden, J. M., Bannink, F. P., Maaskant, M. A., & Curfs, L. M. G. (2009). Solution-focused brief therapy with persons with intellectual disabilities. *Journal of Policy and Practice in Intellectual Disabilities, 6*, 253–259. https://doi.org/10.1111/j.1741-1130.2009.00226.x

Roeden, J. M., Maaskant, M. A., Bannink, F. P., & Curfs, L. M. G. (2011). Solution-focused brief therapy with people with mild intellectual disabilities: A case series. *Journal of Policy and Practice in Intellectual Disabilities, 8*, 247–255. https://doi.org/10.1111/j.1741-1130.2011.00317.x

Roeden, J. M., Maaskant, M. A., Bannink, F. P., & Curfs, L. M. G. (2012). Solution-focused coaching of staff of people with severe and moderate intellectual disabilities: A case series. *Journal of Policy and Practice in Intellectual Disabilities, 9*, 185–194. https://doi.org/10.1111/j.17411130.2012.00352.x

Roeden, J. M., Maaskant, M. A., & Curfs, L. M. G. (2014). Effectiveness of solution-focused coaching of staff of people with intellectual disabilities: A controlled study. *Journal of Systemic Therapies, 33*, 16–34. https://doi.org/10.1521/jsyt.2014.33.2.16

Rose, J., Willner, P., Shead, J., Jahoda, A., Gillespie, D., Townson, J., … Mac Mahon, P. (2013). Different factors influence self-reports and third-party reports of anger by adults with intellectual disabilities. *Journal of Applied Research in Intellectual Disabilities, 26*(5), 410–419.

Royal College of Psychiatrists, Cooper, S.-A., & Council for Exceptional Children. (2001). *DC-LD: [diagnostic criteria for psychiatric disorders for use with adults with learning disabilities/mental retardation]*. London: Gaskell.

Rush, A. J., & Frances, A. (2000). Expert consensus guideline series: Treatment of psychiatric and behavioral problems in mental retardation. *American Journal of Mental Retardation, 105*(3), 159–228.

Segal, Z. V., Williams, J. M. G., & Teasdale, J. D. (2002). *Mindfulness based cognitive therapy for depression: A new approach to preventing relapse*. New York: Guilford.

Shedlack, K. J., Hennen, J., Magee, C., & Cheron, D. M. (2005). Assessing the utility of atypical antipsychotic medication in adults with mild mental retardation and comorbid psychiatric disorders. *Journal of Clinical Psychiatry, 66*, 52–62.

Shedler, J. (2010). The efficacy of psychodynamic psychotherapy. *American Psychologist, 65*, 98–109.

Shepherd, C., & Beail, N. (2017). A systematic review of the effectiveness of psychoanalysis, psychoanalytic and psychodynamic psychotherapy with adults with intellectual and developmental disabilities: Progress and challenges. *Psychoanalytic Psychotherapy, 31*(1), 94–117. https://doi.org/10.1080/02668734.2017.1286610

Smiley, E. (2005). Epidemiology of mental health problems in adults with learning disability: An update. *Advances in Psychiatric Treatment, 11*(3), 214–222. https://doi.org/10.1192/apt.11.3.214

Sohanpal, S., Deb, S., Thomas, C., Soni, R., Lenotre, L., & Unwin, G. (2007). The effectiveness of antidepressant medication in the management of behavioural problems in adults with intellectual disabilities: A systematic review. *Journal of Intellectual Disability Research, 51*(10), 750–765.

Statham, V. & Beail, N. (2018). The views of service user on the accessibility, acceptability, and effectiveness of psychodynamic psychotherapy. *International Journal of Developmental Disabilities, 64*(3), 175–183.

Stenfert Kroese, B., Dewhurst, D., & Holmes, G. (2001). Diagnosis and drugs: Help or hindrance when people with learning disabilities have psychological problems? *British Journal of Learning Disabilities, 29*(1), 26–33.

Stenfert Kroese, B., Jahoda, A., Pert, C., Trower, P., Dagnan, D., & Selkirk, M. (2014). Staff expectations and views of cognitive behaviour therapy (CBT) for adults with intellectual disabilities. *Journal of Applied Research in Intellectual Disabilities, 27*(2), 145–153. https://doi.org/10.1111/jar.12059

Stoddart, K. P., McDonnell, J., Temple, V., & Mustata, A. (2001). Is brief better? A modified brief solution-focused therapy approach for adults with a developmental delay. *Journal of Systemic Therapies, 20*, 24–40. https://doi.org/10.1521/jsyt.20.2.24.23039

Strydom, A., Shooshtari, S., Lee, L., Raykar, V., Torr, J., Tsiouris, J., et al. (2010). Dementia in older adults with intellectual disabilities— Epidemiology, presentation, and diagnosis. *Journal of Policy and Practice in Intellectual Disability, 7*(2), 96–110.

Sturmey, P. (2004). Cognitive therapy with people with intellectual disabilities: A selective review and critique. *Clinical Psychology and Psychotherapy, 11*, 222–232.

Sullivan, W. F., Diepstra, H., Heng, J., Ally, S., Bradley, E., Casson, I., & Hennen, B. (2018). Primary care of adults with intellectual and developmental disabilities: 2018 Canadian consensus guidelines. *Canadian Family Physician, 64*(4), 254–279.

Tsiouris, J. A., Kim, S. Y., Brown, W. T., & Cohen, I. L. (2011). Association of aggressive behaviours with psychiatric disorders, age, sex and degree of intellectual disability: A large-scale survey. *Journal of Intellectual Disability Research, 55*(7), 636–649. https://doi.org/10.1111/j.1365-2788.2011.01418.x

Unwin, G. L., Tsimopoulou, I., Azmi, S., & Stenfert Kroese, B. (2016). Effectiveness of cognitive Behavioural therapy (CBT) programmes for anxiety or depression in adults with intellectual disabilities: A review of the literature. *Research in Developmental Disabilities, 51-52*, 60–75.

Vereenooghe, L., & Langdon, P. E. (2013). Psychological therapies for people with intellectual disabilities: A systematic review and meta-analysis. *Research in Developmental Disabilities, 34*(11), 4085–4102.

Verhoeven, W. M. A., Veendrik-Meekes, M. J., Jacobs, G. A. J., van den Berg, Y. W. M. M., & Tuinier, B. S. (2001). Citalopram in mentally retarded patients with depression: A long-term clinical investigation. *European Psychiatry, 16*, 104–108.

Walker, J. C., Dosen, A., Buitelaar, J. K., & Janzing, J. G. E. (2011). Depression in down syndrome: A review of the literature. *Research in Developmental Disabilities, 32*(5), 1432–1440.

Werneke, U. (2010). *Complementary and alternative medicines 1*. London: Royal College of Psychiatrists.

Willner, P., Rose, J., Jahoda, A., Stenfert Kroese, B., Felce, D., MacMahon, P., … Hood, K. (2013). A cluster randomised controlled trial of a manualised cognitive behavioural anger management intervention delivered by supervised lay therapists to people with intellectual disabilities. *Health Technology Assessment, 17*(21), 1–173. https://doi.org/10.3310/hta17210

World Health Organization. (1992). *The ICD-10 classification of mental and behavioural disorders: Clinical descriptions and diagnostic guidelines*. Geneva: World Health Organization.

World Health Organization. (2017). The ICD-11 Beta version, classification of mental and behavioural disorders: Clinical descriptions and diagnostic guidelines. Geneva: World Health Organization. Available www.icd.who.int, accessed 02/05/2018.

46

Psychosis

Arlene Mannion, Keeley White, Emily Porter,
Julia Louw, Bernadette Kirkpatrick,
Shawn P. Gilroy, and Geraldine Leader

Introduction

Psychosis is a topic of importance in research in individuals with intellectual disabilities (ID). The focus of this chapter is to review the literature on definition, incidence, and prevalence of psychosis in individuals with ID. Importance of studying psychosis in ID, difficulties of studying psychosis in ID, assessment of psychosis in ID, and risk factors of psychosis in ID will be discussed. Treatment of psychosis in ID will be presented, including pharmacological and behavioral treatments, and recommendations for treatment will be given. Following this, future directions for research will be outlined.

Definitions of Psychosis

Psychosis is a set of symptoms related to mental health issues that occurs in a range of psychotic disorders, to include schizophrenia spectrum disorder, bipolar disorder, posttraumatic stress disorder, and major depressive disorder (Doherty & Owen, 2014). Several diagnostic guidelines and assessments exist that can be used to categorize and summarize the dimensional representations of psychosis (Bakken & Høidal, 2014; Cooper & Bailey, 2001; Schützwohl et al., 2016; van Os & Tamminga, 2007). Generally, psychosis is defined as an experience that affects a person's concept of reality, wherein their perception of reality differs considerably from those around them (Krishnan, Kraus, & Keefe, 2011; Sass & Parnas, 2003). During a psychotic occurrence, the person may experience either partial or total loss of contact with reality, and symptoms can range from hallucinations, delusions, disorganized confused thinking and speech, abnormal motor behavior to catatonia (Bakken & Høidal, 2014; Canitano & Pallagrosi, 2017; Spauwen, Krabbendam, Lieb, Wittchen, & Van Os, 2006; Yung, Nelson, Thompson, & Wood, 2010). Positive symptoms of psychosis refer to abnormal, disturbing thoughts, perceptions, and behaviors, such as hallucinations (seeing, hearing, tasting, smelling, and feeling things that are not real) and delusions (fixed false beliefs). Negative symptoms refer to disruption to normal emotions or behavior, for example, apathy and lack of emotional reactivity (Doherty & Owen, 2014). Psychosis differs considerably from person to person however the psychotic experience is very real to that person and it is this lack of awareness of reality that can be very distressing and frightening.

A. Mannion · K. White · E. Porter · J. Louw
B. Kirkpatrick · S. P. Gilroy · G. Leader (✉)
Irish Centre for Autism and Neurodevelopmental Research, School of Psychology, National University of Ireland, Galway, Ireland
e-mail: geraldine.leader@nuigalway.ie

J. L. Matson (ed.), *Handbook of Intellectual Disabilities*, Autism and Child Psychopathology Series, https://doi.org/10.1007/978-3-030-20843-1_46

The literature often refers to psychosis as a psychotic episode (Cooper et al., 2007; McCarthy & Barbot, 2016; Tandon et al., 2013). An episode is defined by the duration of time in which the person has developed symptoms that meet the symptomatic criteria of a specific mental health disorder (Tandon et al., 2013). Episodes can differ in accordance with the type of mental health disorder and can include several symptoms, or a quantified severity or frequency of symptoms (Bakken & Høidal, 2014). They are further differentiated into a single (first) episode or recurrence or relapse of multiple episodes (Tandon et al., 2013).

Defining psychosis in a person with an intellectual disability (ID) or Autism Spectrum Disorder (ASD) is a complex process (Cooper et al., 2007). Quite often a psychotic episode can share core characteristics with repetitive patterns of behaviors, cognitive, social, communication and emotional deficits (Bakken & Høidal, 2014; Canitano & Pallagrosi, 2017; Schützwohl et al., 2016), and observable challenging behaviors which are possibly attributed to the person's ID or ASD rather than a psychotic disorder (Cooper et al., 2007; McCarthy & Barbot, 2016). Even though the core symptoms of psychosis can be difficult to differentiate with the characteristics attributed to varying degrees of ID (Carthy et al., 2010), ID is not a psychiatric disorder, despite being part of the psychiatric classification system (Deb, Matthews, Holt, & Bouras, 2001).

A person with moderate, severe, and profound ID or ASD may have restricted ability to communicate or self-report the symptoms of a psychotic episode (Bakken & Høidal, 2014), which may impede an accurate definition of psychosis. These factors have presented a challenge not only in the diagnosis of psychosis but have also contributed to the lack of explicit ID and ASD psychosis definitions presented within the literature. Researchers have recognized that heterogeneity in cognitive abilities and communication skills of those with an ID and ASD has made it difficult to standardize a definition of psychosis for this population (Canitano & Pallagrosi, 2017; Doherty & Owen, 2014).

Incidence and Prevalence of Psychosis

In general, determining how many people have experienced or are experiencing psychosis can be challenging, especially for individuals with intellectual disabilities, and therefore, estimates on incidence and prevalence can vary (Bagalman & Cornell, 2018; Cooper et al., 2007; Kildahl, Bakken, Holm, & Helverschou, 2017). Some of the challenges associated with quantifying the prevalence of psychosis in the population include changing diagnostic criteria over time (especially for schizophrenia and affective disorders), differing diagnostic tools and instruments (across time and geography), and problems in case finding and confirmation (Perälä et al., 2007). For those with ID, the challenges are even greater, and include lack of appropriate assessment tools, difficulties in separating symptoms of comorbid disorders, and barriers for individuals with ID to self-report their symptoms (Cooper et al., 2007; Hassiotis, Barron, & Hall, 2009; Kildahl et al., 2017). Despite these challenges, a range of studies have examined the prevalence and incidence of psychosis. This section will provide an overview of the key findings in this area.

Prevalence

The lifetime prevalence (LTP) of psychotic disorders in the general population is estimated at just above 3% (Perälä et al., 2007). As psychotic disorders can be severe and impairing, this LTP is substantial and indicates that these conditions likely have a notable impact on public health and well-being. The total LTP corresponds to an approximate LTP of 2.29% for non-affective psychotic disorders and 0.62% for affective psychotic disorders (Perälä et al., 2007). It is thought that psychosis is more prevalent in individuals with ID than in the general population (Cooper et al., 2007; Hassiotis et al., 2009; Holden & Gitlesen, 2004b; Smiley, 2005; Welch, Lawrie, Muir, & Johnstone, 2011). For example, in Cooper et al. (2007) in a study of more than 1000 adults with ID, the point prevalence of psychotic disorders in those with ID was found to range between 2.6% (95% CI = 1.8–3.8%) to 4.4% (95% CI = 3.2–5.8%), depending on the diagnostic criteria. Notably, these rates are significantly higher than those of the general population. In the UK general population, the 12-month prevalence was found to only be 0.4%, which is around 10 times less than the reported point prevalence of 2.6–4.4% (Cooper et al., 2007). Further, the prevalence of schizophrenia specifically in the general population is approximately 1%, while the prevalence in individuals with ID is reported around 3% (Hassiotis et al., 2009; Smiley, 2005).

The prevalence of psychotic disorders in individuals with ID likely varies based on the level of ID. It was reported that the point prevalence in adults with mild ID is around 5.8%, while it is around 3.5% in those with moderate to profound ID (Cooper et al., 2007; Hassiotis et al., 2009). This trend was also confirmed in Holden and Gitlesen (2004b), where it was found that psychosis was more prevalent in those with moderate ID than in those with severe or profound ID; while the difference in prevalence between those with severe ID and profound ID was minor.

In the general population, prevalence estimates have been found to vary based on a wide number of factors (McGrath, Sukanta, Chant, & Welham, 2008), and research has shown that psychosis and schizophrenia do not occur equally across all geographic and demographic groups (Kirkbride et al., 2012; Messias, Chen, & Eaton, 2007; Steel et al., 2014). For example, the prevalence of schizophrenia was found to differ across latitude and economic status, with developed countries having a median prevalence estimate of 3.3 per 1000, with less-developed countries having 2.6 per 1000 (McGrath et al., 2008). The prevalence was also associated with migration status (McGrath et al., 2008). However, there is still uncertainty in the exact factors that impact prevalence. For instance, in McGrath et al. (2008) the prevalence of schizophrenia was found not to statistically differ between males and females, between urban and rural environments, or over time; while in Messias et al. (2007), schizophrenia was found to be more prevalent among males, with males having a 30–40% higher lifetime risk of developing schizophrenia than females. Despite this, it is clear that schizophrenia affects males and females differently, with varying ages of onset and incidence.

Gender differences in the prevalence of psychotic disorders, along with the subtype of disorder, have also been indicated in individuals with ID, however results have not been consistent across studies and therefore more research is needed in this area (Axmon, Sandberg, & Ahlström, 2017). Overall, further investigation in this area is warranted, as individuals with ID may differ from the general population with regard to their interaction with societal factors (Axmon et al., 2017).

Incidence

Based on a recent international, multisite study, the crude incidence of psychotic disorders in the general population is estimated at 21.4 per 100,000 person-years (95% CI = 19.4–23.4) (Jongsma et al., 2018). Of all non-organic psychotic disorders, it was estimated that 78.7% of incident cases are non-affective psychotic disorders and 19.9% are affective (Jongsma et al., 2018). The average age at the first contact with a first diagnosed episode of a non-organic psychotic disorder, was 30.5 years. The first contact age is generally earlier in men (28) than in women (34) (Jongsma et al., 2018). Crude incidence rates of the first episode peaked at 61 per 100,000 person-years for men between the ages of 18 and 24, and decreased sharply after this age range. For women, the peak incidence occurred also at 18–24, however decreased more slowly afterwards with another, more minor, peak between ages 50 and 54 (Jongsma et al., 2018). Similar trends in age were also found in other studies (Kirkbride et al., 2012; Messias et al., 2007).

In adults with ID, the 2-year incidence of psychosis is 1.4% (95% CI = 0.6–2.6), and the first-episode incidence is 0.5% (95% CI = 0.1–1.3) (Cooper et al., 2007). The incidence ratio for (first-episode) psychosis in adults with ID is 10 (95% CI = 2.1–29.3) relative to the general population (Cooper et al., 2007; Hassiotis et al., 2009).

However, like prevalence estimates in the general population, the current literature on incidence estimates have shown significantly varying rates across groups (Jongsma et al., 2018; Kirkbride et al., 2012, 2017; McGrath et al., 2008; Messias et al., 2007). Specifically, incidence of psychotic disorders has been demonstrated to vary by sex (Jongsma et al., 2018; Kirkbride et al., 2012; Messias et al., 2007), age (Jongsma et al., 2018; Kirkbride et al., 2012; Messias et al., 2007), migration status and ethnicity (Bourque, vander Ven, & Malla, 2011; Jongsma et al., 2018; Kirkbride et al., 2012; McGrath et al., 2008), and geographical region (Jongsma et al., 2018; Kirkbride et al., 2012, 2017; McGrath et al., 2008; Messias et al., 2007). Geographical region variations include across countries and latitudes, along with local variations from urban to rural regions (McGrath et al., 2008). Socioeconomic factors may also impact incidence (Jongsma et al., 2018; Kirkbride et al., 2012).

Notably, individuals with schizophrenia have a two- to three-fold increased risk of death relative to the general population (McGrath et al., 2008). As approximately 7 individuals out of every 1000 may be affected by this condition (McGrath et al., 2008), and it co-occurs with ID three times more than in the general population (Welch et al., 2011), the burden on health is considerable. Overall, while it is clear that there is a degree of heterogeneity in the incidence of psychotic disorders globally, both in the general population and in those with ID (Axmon et al., 2017; Holden & Gitlesen, 2004b; Steel et al., 2014) further research is needed to examine the impact of the factors that may be responsible for this heterogeneity.

Importance of Studying Psychosis in Intellectual Disabilities

Advances in our understanding of psychosis in intellectual disabilities and comorbidity between behavioral profiles and psychiatric disorders may help to identify diagnostic overshadowing, that is, attributing the symptoms of mental ill-health to a person's intellectual disability behavioral profile (Canitano & Pallagrosi, 2017). Psychotic episodes may create a disorganized, frightening, isolated, and socially withdrawn environment (Bakken & Høidal, 2014), in which the person with ID and ASD may not have the cognitive, social, emotional, and communication capabilities to seek help. Research that focuses on observations of the nonverbal communications and suspected psychotic symptoms of individuals with ID may help to differentiate psychosis from the person's behavioral profile and thus help to improve the diagnostic system (Bakken & Høidal, 2014). In the absence of further research into psychosis and coexisting behaviors associated with ID, there is risk for individuals with ID to receive the incorrect mental health diagnosis (Bakken & Høidal, 2014), and subsequently an inappropriate ineffective treatment (Allen, 2008). Any treatment outcomes must be considered within the context of improved quality of life and daily functioning and must provide relief of physical and perceived emotional pain (Allen, 2008). Implications extend to the provision of mental health care for the person with ID or ASD, who often fall between services, particularly as they transition into adulthood (Selten, Lundberg, Rai, & Magnusson, 2015).

Research into psychosis and ID has important implications for psychiatric research. By developing new procedures to detect and identify psychosis we can advance our knowledge of the pathways from genotype to clinical phenotype, which are essential to inform new classification systems and to develop innovative therapeutic treatments (Doherty & Owen, 2014).

Recent research has suggested that prescription of psychotropic medication prescribed to adults with ID results in an overmedicated population with the prescription of antipsychotics drugs (Axmon, Kristensson, Ahlström, & Midlöv, 2017; McNamara et al., 2017; O'Dwyer et al., 2017; Schützwohl et al., 2016) and in some cases the drugs may be ineffective (McNamara et al., 2017; O'Dwyer et al., 2017) and cause adverse effects (O'Dwyer et al., 2017). Prescription of psychotropic medication needs to be reassessed to evaluate their overuse and effectiveness.

Difficulties of Diagnosis of Psychosis in Intellectual Disabilities

The identification of psychosis in individuals with intellectual disability (ID) presents considerable challenges. Some of these challenges consist of the limited number of appropriate assessment tools, the presence of dual diagnosis of ID and psychiatric disorders, the level and severity of ID and the difficulty in self-report of diagnostic and/or psychotic symptoms (Helverschou, Bakken, & Martinsen, 2011; Underwood, Kumari, & Peters, 2016). One of the cautionary aspects on which clinicians are particularly advised is to be careful and pay special attention to the diagnoses of schizophrenia in the presence of ID (Feinstein, Eliez, Blasey, & Reiss, 2002). The section below describes the complexity and difficulty in diagnosing psychosis in individuals with ID.

Diagnostic Systems and Assessment Tools

Several researchers have reported a higher prevalence of psychosis in adults with ID compared to the general population (Buckles, Luckasson, & Keefe, 2013; Cooper et al., 2007). However, existing diagnostic systems that measure the diagnoses in individuals with ID may not be fully compatible when making a psychiatric diagnosis in them (Watson, 2005). This may be related to the lack of one standardized criterion for psychiatric diagnosis across the whole spectrum of ID (Cohen-Kettenis & Pfäfflin, 2010). Therefore, typical diagnostic systems are not always useful for individuals with intellectual disabilities. Further, the heterogeneity in abilities and communication skills among adults with ID is varied. Some features might not always be apparent, or may be difficult to elicit, making the diagnosis difficult, particularly in those who have severe intellectual disability, and impaired communication (Wing, Gould, & Gillberg, 2011). This explains the reasons why the evidence-based knowledge on the assessment and diagnosis of psychosis problems in individuals with ID is still under-researched (Perry et al., 2018).

It has also been noted that a number of limitations with classification systems such as the *Diagnostic Manual-Intellectual Disability* or *Diagnostic Criteria for Psychiatric Disorders for Use with Adults with Learning Disabilities/Mental Retardation* still exist (Madhavan, 2018). In addition, factors such as gender differences have also been reported as potentially explanatory predictors in understanding psychiatric ill-health in the general population (Axmon, Sandberg, & Ahlström, 2017; Cohen-Kettenis & Pfäfflin, 2010). But given the social differences in how individuals with ID are treated in society with regard to accessibility, attitudes, and perceptions, this association between gender and psychiatric disorders in individuals with ID will not apply similarly as in the general population (Cohen-Kettenis & Pfäfflin, 2010). What is imperative in the development of assessment tools and systems is a detailed description of specific clinical features of psychiatric illness, with special reference to where these may differ from their presentation in the general population without disabilities. Moreover, a stronger emphasis on reports from support staff and caregivers is often required.

Some tools do aim at screening for psychiatric disorders in individuals with ASD and ID such as the Psychopathology in Autism Checklist (PAC; Helverschou, Bakken, & Martinsen, 2009). The PAC is a behavior checklist that is designed for completion by caregivers or family members, and therefore no general knowledge of psychiatric disorder is required. However, it is unclear whether or not such subjective knowledge influences final scores. Another instrument designed to identify psychiatric symptoms in individuals with ID is the Mini Psychiatric Assessment Schedules for Adults with Developmental Disabilities (Mini PAS-ADD; Moss & Hurley, 2014; Prosser et al., 2002). However, the Mini PAS-ADD may not be sensitive enough to detect symptoms in individuals with no or very limited speech.

The Presence of Dual Diagnosis

Similar deficits exist between developmental disabilities (such as ID and ASD) and psychosis. Therefore, psychosis and developmental disabilities have overlap (Matson, Mayville, Lott, Bielecki, & Logan, 2003). Intellectual disability and psychosis occur 300% more often than would be expected by chance (Cooper et al., 2007; Turner, 1989). Some note that the psychosis demonstrated by those with ID may be subtly different (Welch et al., 2011). In adulthood, negative symptomology of psychosis shares some similarities with autism spectrum disorder (Skokauskas & Gallagher, 2010). These populations warrant intervention to address several areas of negative symptoms, which are largely similar to those addressed as adaptive functioning deficits, such as deficits in communication, socialization, daily, adaptive living skills, and deficits in the management of behavior problems.

The manifestation of correlated symptoms of comorbid disorders including psychopathologies other than cognitive impairments has been recognized in individuals with ID (Kozlowski, Matson, Sipes, Hattier, & Bamburg, 2011). It is also evident in the literature that the presence of ID reduces the diagnostic significance of behaviors that normally constitute psychiatric symptoms (Holden & Gitlesen, 2004b). Research often discusses the terms psychosis and ID separately but makes no mention of their possible interrelationship (Allington-Smith, 2006; Clarke, 2006). It is important to review the extent of the correlation between comorbid disorders to address the difficulty in clear and reliable diagnosis of psychotic symptoms.

Similarities in the Etiology of Challenging Behavior and Psychiatric Disorder

Often, challenging behavior is referred to as the severe level of behavior problems among individuals with ID (Emerson et al., 2001). In particular, aggression towards others, extreme temper tantrums, screaming or shouting, and self-injury are examples of behaviors that may be challenging. A number of authors have suggested that such behavior problems may be indicators of psychiatric disorders in individuals with intellectual disability (Emerson et al., 2001; Moss & Hurley, 2014). Family members, support staff, and carers tend to excessively manage these behaviors that may result in social isolation and restricted opportunities for taking part in ordinary social and societal activities. This makes it very difficult to establish a dignified life situation for individuals with disabilities as endorsed by United Nations Convention of the Rights of People with Disabilities (United Nations General Assembly, 2006). Therefore, the emerging consensus may be accurate that individuals with severe to profound ID may be more prone to psychiatric illness, because they are unable to live normal lives (Munir, 2016). In spite of the parallels, the relation between behavior problems and psychiatric disorders is not entirely clear, given the mixed empirical results that indicate that perhaps studies adopted varied methodologies. What we know is that psychiatry refers to the study of the mind and treatment of disorders of the mind, including but not limited to mood, personality, development, and cognition. Psychosis on the other hand, is understood to be a symptom and essentially a flawed ability to perceive reality with the most commonly discussed symptoms being hallucinations and delusions but, this is not all necessarily experienced in a negative way. It is worth considering that in diagnosing psychiatric illness, it is important to differentiate which symptoms can be part of such an illness, and which can be explained by the intellectual disability.

Severity of Challenging Behaviors

In recent years, maladaptive and challenging behaviors are increasingly explained by psychiatric disorders, however, because of little or total lack of speech, the diagnosis relies predominantly on more tangible behaviors (Emerson et al., 2001). However, diagnosing psychiatric disorders in individuals with ID, particularly those with severe and profound ID, is more problematic. Individuals with milder levels of disabilities can use self-report measures and can be diagnosed using standard criteria with little modification, but for those with moderate and severe disability, diagnosis is challenging. The severity of behavior problems relates to signs and symptoms common in psychiatric illness such as social withdrawal, excessive agitation, lack of concentration, stereotyped movement disorders, abnormal sleep, and certain other behaviors (Kelly, 2013). These symptoms have been described as the expression of "diagnostic overshadowing," where the diagnostic symptoms are an underlying result from possible brain damage rather than symptoms of an illness (Mason & Scior, 2004).

The Level and Degree of Intellectual Disability

Studies show controversial evidence as to whether or not psychiatric illness is more common among individuals across the spectrum of mild-to-profound ID (Hove & Havik, 2010). Current research indicates that schizophrenia can be reliably diagnosed in adults with mild ID (Deb, Thomas, & Bright, 2001). For example, researchers made a comparison to the general population and there appears to be a higher rate of schizophrenia among adults who have a mild intellectual disability (Kessler, Chiu, Demler, & Walters, 2005). However, studies report the overall rate of psychiatric illness in adults with ID does not differ significantly from that of individuals without ID in the general population if diagnoses like behavioral disorder, personality disorders, ASD, and Attention-deficit/hyperactivity disorder (AD/HD) are excluded (McGough et al., 2005). But the diagnosis of schizophrenia in those with an IQ less than 45 may be difficult or impossible, because of the inability to do an in-depth verbal interview (Friedlander & Donnelly, 2004). In particular, the difficulty in diagnosing schizophrenia is of concern, specifically in those with a moderate or more severe intellectual disability (Deb, Thomas, & Bright, 2001). The diagnosis is based upon the presence of a number of complex subjective symptoms, delusions and thought broadcasting. The diagnosis of psychosis not otherwise specified (NOS) in individuals in particular is associated with moderate or severe ID (Friedlander & Donnelly, 2004).

While psychiatrists tend to use a cluster of symptoms or behaviors that relate with each other, also referred to as syndromic classification, some clinicians believe that the approach of behavioral classification that is more prevalent in the clinical psychology literature may be more appropriate for use in adults with severe intellectual disability (Drake et al., 2001). As a result, diagnoses of affective disorders are even possible in profound intellectual disability, due to observable behavioral elements of such disorders (Drake et al., 2001).

Difficulties in Self-Report

Generally, individuals with ID have significant limitations in verbal ability with increasing levels of the degree of ID. However, psychiatric assessment in ID often requires knowledge through verbal expression of the individual's previous functioning, and that can be a very complicated and time-consuming process in individuals with severe to profound ID. It involves investigating and identifying qualitative changes in symptoms related to the level of ID and interpreting atypical symptoms. In addition, investigating of conventional symptoms of psychiatric disorder is also crucial (Helverschou et al., 2011). Consequently, a certain level of communicative ability is needed to describe such symptoms.

Clinical reviews do provide valuable information about the identification, diagnosis and outcome of psychotic symptoms in individuals with low verbal abilities (Friedlander & Donnelly, 2004). In the absence of limited validated tools for assessment of psychosis in individuals with ID, it is noted that conventional assessment tools can be useful in such assessments. However, a full compensation for a lack of self-report is not always possible when relying on conventional assessment tools. It is possible though to partly compensate for lack of self-report by using multiple informers, a process sometimes referred to as triangulation (Yin, 2013). In this instance, it involves including family and professional caregivers as informers, instead of the challenging self-report of the person with ID. As such, a systematic assessment can be initiated with in-depth interviews with family members and caregivers, as well as direct observation by clinicians.

Some anxiety symptoms can be difficult to assess in nonverbal individuals, like excessive worry, unreasonable thoughts, and verbal reports of physiological signs (Bailey & Andrews, 2003). Therefore, the identification of psychiatric disorders in this group is reported to be difficult because of inherent communication and cognitive deficits. As such, giving symptoms a higher score in individuals with more severe ID in order to increase total scores is hardly justified, as there is no gold standard (Mungkhetklang, Crewther, Bavin, Goharpey, & Parsons, 2016). One possibility might be to improve the observational skills of caregivers. However, for behavior to be observed, it must have some strength and clarity related to the specific behavioral symptoms.

Many psychiatric symptoms are a subjective phenomena, for instance hearing voices, and not available to observers unless the individual with ID has the necessary verbal ability to report such experiences. As a result, it is not feasible to reliably diagnose in individuals who are nonverbal or those with limited communication skills. Where an individual's level of functioning and disorganized behavior make self-report impossible, no observer is able to report such symptoms. Treating observational data as interchangeable with self-report data would have led to a serious risk of symptoms being overlooked. Research within behavior analysis indicates that verbal behavior plays a key role in the development and maintenance of psychopathology, not only in anxiety and depression (Tone, Garn, & Pine, 2016).

It is clear that to address the challenges in the diagnoses of psychosis in individuals with ID, an accurate diagnosis will likely not be made from a one-time encounter by a professional. Instead, the diagnosis will rather evolve over time, because a key component of psychosis is based on internal experiences and their description. Thus, a comprehensive assessment and a clear description of the diagnostic criteria used are needed.

Risk Factors of Psychosis

Psychotic disorders are heterogeneous, and as such, a wide range of risk factors and protective factors have been investigated (Hassiotis et al., 2009; Heckers, 2009; Radua et al., 2018). As incidence rates vary significantly across geography and demographics, risk factors are also likely to vary across time and space (McGrath et al., 2008). While many potential risk factors for psychosis have been identified, for some factors the evidence is not conclusive to date. What follows is an examination of the potential risk factors for psychosis, and an indication of the level of evidence for each is provided. The risk factors are divided into four broad categories, based on those in Dean and Murray (2005) and in Heckers (2009): (1) baseline risks, (2) early-life risks (in utero and during birth), (3) childhood risks, and (4) risks that develop later in life and those that occur close to the incidence of psychosis. It is important to note that for individuals with ID, the risk factors have not been as thoroughly investigated as for the general population (Axmon et al., 2017; Cooper et al., 2007; Hassiotis et al., 2009; Smiley, 2005), and as such the risk factors are sometimes extrapolated from those of the general population (Hassiotis et al., 2009). Furthermore, many risk factors for psychosis are also risk factors for intellectual disabilities so it can be difficult to separate the impacts of each (Hassiotis et al., 2009).

Baseline Risks

The baseline risk relates to the level of risk that an individual may have due to their family history, genetics, and place and time of birth. For psychotic disorders, the risk topology for the general population is generally viewed as a complex interaction between environmental risk factors and susceptibility genes (Dean & Murray, 2005; Heckers, 2009; Radua et al., 2018; van Os, Rutten, & Poulton, 2008), and this view is maintained for individuals with intellectual disabilities (Hassiotis et al., 2009).

For schizophrenia, in particular, genes are necessary but are not the sole operator in developing the condition (Dean & Murray, 2005). However, genetics do play a substantial role in an individual's probability of developing schizophrenia (Heckers, 2009; Messias et al., 2007). For example, the risk of an individual presenting with a psychotic disorder increases several fold if a first-degree relative also has a history with a psychotic disorder (Heckers, 2009). Several genes are being investigated in terms of their link to psychosis (Hassiotis et al., 2009).

Baseline risk factors aside from genetics include gender, culture, residence type (urban versus rural), ethnicity, and immigration status (Dean & Murray, 2005; Heckers, 2009; Messias et al., 2007). In general, men are at increased risk for developing schizophrenia, with a 30–40% higher risk than women (Messias et al., 2007). More research is needed to identify whether this increased risk holds for individuals with ID, however current research does suggest that there are gender differences in this group as with the general population (Hassiotis et al., 2009).

Early-Life Risks (In Utero and During Birth)

Many risk factors that occur in utero or during birth have been well documented in the general population (Dean & Murray, 2005; Heckers, 2009) and have also been suggested for those with intellectual disabilities (Hassiotis et al., 2009). These risk factors include obstetric complications, such as intrauterine growth restriction (also called fetal growth retardation) or hypoxia, prenatal or postnatal infections, maternal malnutrition, and maternal stress (Dean & Murray, 2005; Heckers, 2009; Messias et al., 2007). These complications can impact the process of normal brain development (Heckers, 2009).

Childhood Risks

A number of risks that can occur during childhood have been identified in the general population, and may also impact individuals with ID. These include the broad categories of traumatic life events (TLEs) or adverse events or experiences, developmental abnormalities, and infections and immune responses.

In Khandaker et al. (2018) it was reported that exposure to infections can lead to an increased risk of developing non-affective psychoses (with a hazard ratio for an infection during the first year of life of 1.19, with a 95% confidence interval of 1.06–1.33). Other studies have linked meningitis to an increased risk (approximately 5×) of developing psychosis and schizophrenia (Messias et al., 2007), and shown that individuals with *Toxoplasmosis gondii* antibodies have a higher prevalence of schizophrenia (Messias et al., 2007). In general, it is thought that infections during early childhood may impact typical neurodevelopment (Khandaker et al., 2018).

Childhood developmental delays or abnormalities have also been associated with an increased risk in developing schizophrenia and psychosis (Hassiotis et al., 2009; Khandaker et al., 2018; Messias et al., 2007). The relation between psychosis and IQ has been investigated thoroughly, and studies suggest a linear association between premorbid IQ and schizophrenia (Khandaker et al., 2018). There may be a common risk that links low IQ and psychosis (for example, abnormal brain development), or the link may be causal (Hassiotis et al., 2009). Future research is needed in this area. As early childhood infections are associated with both risk of lower IQ and developing psychosis, these associations may also be causal (Khandaker et al., 2018).

Typically developing children who experience traumatic life events (TLEs) including abuse, trauma, or adverse experiences have increased risk of developing psychosis (Dean & Murray, 2005; Gibson, Alloy, & Ellman, 2016; Radua et al., 2018). Children who have experienced TLEs are at increased risk of psychotic disorders, and such TLEs have been shown to be one of the most robustly associated (environmental) risk factors for psychosis (Bendall, Alvarez-Jimenez, Nelson, & McGorry, 2013;Gibson et al., 2016 ; Varese et al., 2012). Specifically, the odds of developing a psychotic disorder or psychotic symptoms in individuals with a history of TLEs are between 2.78 and 11.50 (where the value varies based on TLE type and methodology) (Gibson et al., 2016; Varese et al., 2012). Notably, there has been a demonstrated dose–response relationship between TLEs and psychosis (Gibson et al., 2016). Similarly, life events have also been significantly associated with the development of schizophrenia in individuals with ID (Hassiotis et al., 2009; Tsakanikos, Bouras, Costello, & Holt, 2007).

Risks Later in Life

The risks discussed below include risks that occur later in life, and those that occur close to the transition into psychosis and during the symptomatic risk stage. In the general population, studies have shown that there is a robust link between sleep dysfunction and psychosis, but more investigation is needed to establish a causal association (Reeve, Sheaves, & Freeman, 2015). Exposure to drugs, especially marijuana, or drug abuse, are also suggested as risk factors (Dean & Murray, 2005; Hassiotis et al., 2009; Heckers, 2009) for the general population and for individuals with ID. A range of studies across the world have shown higher risk, ranging from 2 times up to 25 times higher for typically developing individuals using cannabis (Messias et al., 2007). However, this risk is difficult to study in individuals with ID since this population has low rates of usage (Cooper et al., 2007).

Smoking has been associated with a risk of psychosis in the general population and in individuals with ID, as has visual impairment (Cooper et al., 2007). However, in individuals with ID, not having comorbid epilepsy was found to correspond with psychosis; while in the general population the opposite appears to be true (Cooper et al., 2007).

The most significant risk factor for the general population, with the strongest level of evidence of association with psychosis, is the existence or emergence of sub-threshold psychotic symptoms (Heckers, 2009; Yung, Phillips, Yuen, & McGorry, 2004). Individuals already experiencing subthreshold symptoms, along with a family history of psychotic disorders, were categorized as being at "ultra-high-risk" (UHR) for developing schizophrenia and other psychotic disorders (Yung et al., 2004). It was found that 34.6% of the individuals at UHR developed obvious psychotic symptoms within 1 year (i.e., the transition rate to disease was 34.6%) and 39.4% developed psychosis eventually.

The identification of such high-risk individuals is of special interest, as this group may benefit in particular from early interventions that aim to prevent the onset of psychosis (Yung et al., 2004). Another recent review found that those individuals with prodromal symptoms were at greater risk for developing a psychotic disorder; however, less than 40% of those actually did (Fusar-Poli et al., 2013). For youths (under age 18) with intellectual disabilities, the early onset of psychotic symptoms led to a later diagnosis of schizophrenia or schizoaffective disorder in more than half of the individuals (Friedlander & Donnelly, 2004) suggesting transition rates for young people with ID may be higher than rates in the general population; but it should be noted that the sample size in this study was small. Overall, however, transition rates for those at high risk have been highly variable in the literature (Conrad et al., 2017) and more investigation of the role of negative symptoms (as opposed to solely positive symptoms) may provide useful clinical information (Fusar-Poli et al., 2013).

Assessment of Psychosis

As discussed above, an accurate diagnosis of psychotic disorders in individuals with ID is difficult partly because of the cognitive and language difficulties, making it difficult for the individual to describe psychotic symptoms

(Helverschou et al., 2011; Myrbakk & von Tetzchner, 2008). However, behavioral phenotypes have been used in identifying psychotic disorders among individuals with ID. For instance, disorganized behavior has been described as potentially being equivalent to disorganized thought or speech in individuals without ID (Bakken, Eilertsen, Smeby, & Martinsen, 2009; Bakken, Friis, Løvoll, Smeby, & Martinsen, 2007; Kildahl et al., 2017).

It has been recommended that clinicians gain knowledge about the person's previous functioning and modes of communication for those with ID referred for concerns of psychosis (Kildahl et al., 2017). This information in addition to behavioral observations will allow the clinical to investigate conventional symptoms of psychiatric disorders (Helverschou et al., 2011). Furthermore, combined knowledge of ID and psychosis/psychiatric disorders are invaluable to the assessment (Kildahl et al., 2017). Although general assessment tools are helpful, they warrant careful interpretation and inclusion of multiple informers (e.g., caregivers, family, or other professionals), as this may help to attain a more accurate diagnosis (Kildahl et al., 2017).

Generally, clinicians utilize a combination of structured diagnostic interview as well as assessments of positive and negative symptoms. For example, a structured diagnostic interview like the Mini-International Neuropsychiatric Interview (MINI; Sheehan et al., 1998) as well as the use of a structured clinical scale for assessment of positive and negative symptoms such as the Positive and Negative Syndrome Scale (PANSS) or Brief Psychiatric Rating Scale, which have been used in a case study (Kildahl et al., 2017).

Tools aimed at screening for and aiding in the diagnosis of psychotic disorders have been developed. These include the Assessment of Dual Diagnosis (ADD; Matson, 1997), the Diagnostic Assessment for the Severely Handicapped-II (DASH-II; Matson, 1995), and Psychopathology Instrument for Mentally Retarded Adults (PIMRA) (Matson, Kazdin, & Senatore, 1984), among others. The ADD is used for assessment of individuals with mild or moderate intellectual disabilities, while the DASH-II is used with individuals with severe or profound intellectual disabilities. The ADD is a parent/caregiver report measure which contains 79 items and measures comorbid psychopathology such as mania, depression, anxiety, post-traumatic stress disorder (PTSD), substance abuse, somatoform disorders, dementia, conduct disorder, pervasive developmental disorder, schizophrenia, personality disorders, eating disorders, and sexual disorders.

The DASH-II is an 84-item parent/caregiver report measure which assesses psychopathologies such as anxiety, depression, mania, pervasive developmental disorder/autism, schizophrenia, stereotypies, self-injury, elimination, eating, sleeping, sexual, organic, and impulse control. The PIMRA is a 56-item parent/caregiver report measure which assesses schizophrenia, affective disorder, psychosexual disorder, adjustment disorder, anxiety disorder, somatoform disorder, and personality disorder and inappropriate adjustment.

The Mini Psychiatric Assessment Schedule for Adults with Developmental Disability - Mini PAS-ADD (Prosser et al., 2002) and the PAS-ADD Checklist (Moss et al., 2002) have been used to assess psychotic disorders in individuals with ID. The PAS-ADD checklist consists of 27 items that can be used to help clinicians recognize mental health problems, including psychotic disorders. The Mini PAS-ADD is more elaborate requiring more extensive training in its administration and allows clinicians to gage the presence of psychosis. The Mini PAS-ADD comprises of 86 psychiatric symptoms and generates subscale scores on depression, anxiety, hypomania, obsessive-compulsive disorder, psychosis, unspecified disorder, and pervasive developmental disorders. Both of these measures have been shown to be reliable and valid measure for assessing psychiatric disorders in adults with ID (Moss et al., 2002; Prosser et al., 2002).

Recommendations for the assessment of psychosis in individuals include the following. First, screening tools should be used in combination with the International Statistical Classification of

Diseases and Related Health Problems (ICD-10; World Health Organization, 2004) and the Diagnostic and Statistical Manual of Mental Disorders (DSM-5; American Psychiatric Association, 2013). Second, a biopsychosocial approach should be taken, therefore a multidisciplinary team, including a psychiatrist, psychologist, and physician is recommended. Third, medical conditions should be assessed. It is important to assess for preexisting conditions that may predispose an individual to psychosis. Medication side effects and drug interactions need to be evaluated. It must be determined if certain symptoms are related to substance use. Assessing for medical conditions may include a complete physical, blood tests and brain scans. Certain medical conditions may display similar symptoms to psychosis including neurological diseases (e.g., Parkinson's disease, Huntington's disease), delirium, brain tumors or cysts, dementia/Alzheimer's disease, HIV and other infections that affect the brain, some types of epilepsy, stroke, hyponatremia, hepatic encephalopathy, uremia, hyperadrenalism, or Wilson's disease. Genetic disorders that may have a predisposition to psychosis should also be considered, including velocardiofacial syndrome, Prader–Willi syndrome, Turner's syndrome, Phenylketonuria, or Klinefelter's syndrome. It is possible that psychosis has a medical cause if hallucinations are olfactory, as this may be seizure related. Where hallucinations are tactile, it may be delirium or substance abuse/withdrawal. If hallucinations occur while going to sleep or while waking up, they may be hypnogogic or hypnopompic hallucinations. Fourth, if there is no medical cause for psychosis (such as seizures), the following should be assessed through interview with significant others: Background and life history, current living circumstances, environment, history of presenting symptoms, and changes in presentation of symptoms over time or in different places or situations. Fifth, the following needs to be ruled out: developmentally appropriate self-talk, imaginary friends, reports that are culturally normative (e.g., seeing relatives who have died) in isolation from other symptoms, and learned behavior that is adaptive to the environment. Sixth, the individual should be observed over time. Accurate diagnosis comes over time and with a multidisciplinary team. A major reason for the identification of a psychiatric disorder, or a thorough psychiatric assessment may increase the probability of prescribing effective treatment, both pharmacological and psychological.

Pharmacological and Psychological Treatment of Psychosis in ID

There exists limited empirical evidence on the treatment of psychotic disorders in adults with ID. As such, treatment approaches are often extrapolated from research in the general population and from clinical experience. Treatment for most individuals with comorbid mild ID and major mental illness is delivered predominantly by the mental health services (Patil, Keown, & Scott, 2013). However, interagency and/or intensive support approaches providing coordinated and integrated services have been associated with improved clinical outcomes in some (Lunsky, Bradley, Durbin, & Koegl, 2008; Lunsky, Gracey, Bradley, Koegl, & Durbin, 2011) but not all cases (Martin et al., 2005).

Much like assessment, treatment takes a biopsychosocial approach, including both pharmacological and psychological treatments, which is tailored to the individual's needs. Prior to initiating treatment, it is important to conduct a risk assessment to ensure the client's safety as well as the safety of others.

Pharmacological Treatment

Pharmacological treatments for psychotic disorders include both first and second generation antipsychotic medications. Particularly, antipsychotic drugs are the most frequently prescribed of the psychotic drugs among ID population (Matson & Mahan, 2010). In terms of psychosis, they are used to treat the positive symptoms such as hallucinations and delusions. See Table 46.1. for a list of first- and second-generation antipsychotic medications.

Table 46.1 List of common first-generation antipsychotics (FGAs) and second-generation antipsychotics (SGAs)

FGAs (Typical)	SGAs (Atypical)
Haldol (Haloperidol)	Clozaril (Clozapine)
Loxitane (Loxapine)	Risperdal (Risperidone)
Mellaril (Thioridazine)	Abilify (Aripiprazole)
Moban (Molindone)	Seroquel (Quetiapine)
Navane (Thiothixene)	Zyprexa (Olanzapine)
Prolixin (Fluphenazine)	
Serentil (Mesoridazine)	Invega (Paliperidone)
Stelazine (Trifluoperazine)	Fanapt (Iloperidone)
Thorazine (Chlorpromazine)	Zeldox (Ziprasidone)
Trilafon (Perphenazine) (Droperidol) (Pimozide) (Prochlorperazine) (Thioridazine)	Saphris (Asenapine)

First-Generation Antipsychotics (FGAs)

First-generation antipsychotics (FGAs) are also known as *typical antipsychotics*, dopamine antagonists, neuroleptics, and classic antipsychotics. These terms have evolved over the years due to historical and conceptual implications. Neuroleptics refers to the ability of a drug to cause "neurolepsis" which includes psychomotor slowing, emotional quieting, and affective indifference. These symptoms were initially a reliable sign of antipsychotic efficacy; however, we now know that these effects are not required for drugs to have therapeutic action and that the presence of these symptoms predicts low treatment adherence. Dopamine antagonists act on different regions such as mesolimbic, mesocortical, nigrostriatal, and tuberoinfundibular pathways.

Typical Antipsychotics is the most commonly used term in practice and is based on the view that SGAs have atypical properties (e.g., low risk of extrapyramidal symptoms [EPS]). Therefore, drugs that do not have atypical properties are referred to as typical antipsychotics. This original concept of atypicality risk (low EPS) changed to a broader definition that included efficacy for negative and cognitive symptoms in schizophrenia (Gründer, Hippius, & Carlsson, 2009). This definition was revised after the CATIE (Clinical Antipsychotic Trials Intervention Effectiveness) trial failed to confirm that SGAs are more effective than FGAs, with the exception of clozapine for treatment-resistant schizophrenia. In order to avoid confusions regarding effectiveness in schizophrenia, the World Psychiatric Association suggested the term first-generation antipsychotics.

FGAs are the primary treatment of schizophrenia and related psychotic disorders. The use of FGAs has declined, mainly due to the increase in prescriptions of second-generation antipsychotics. However, they remain a valuable option in the treatment of psychotic disorder as they are considered less expensive. FGAs are associated with higher risk of neurological side effects such as tardive dyskinesia, extrapyramidal side effects (EPSE), and dystonia. For instance, haloperidol is effective in reducing psychotic symptoms; however, they can produce unpleasant sides effects such as drowsiness and EPSE (particularly dystonia, parkinsonism, and akathisia) which can lead to discontinuation of medication and relapse.

Second-Generation Antipsychotics (SGAs)

Second-generation antipsychotics (SGAs), also known as *atypical antipsychotic*s. There is a lower risk of EPS associated with the use of clozapine (Leucht, Kissling, & Davis, 2009). Atypical antipsychotics were broadened to include efficacy against negative and cognitive symptoms, lack of prolactin elevation, and efficacy in treatment-resistant individuals. SGAs also block D2 receptors (like in FGAs) in addition to their ability to block 5HT2A receptors. Specifically, SGAs are known as dopamine–serotonin antagonists, due to their high affinity for 5HT2A receptors (Sadock & Sadock, 2009). Although SGAs have lower risk of neurological side effects, they have an increased risk of developing metabolic side effects such as hyperglycemia, weight gain, and dyslipidemia.

Clozapine was manufactured by Sandoz in 1959 and was the first of the SGAs to be produced. However, it was later discontinued and withdrawn in several countries following reports of severe blood reactions (i.e., neutropenia) and subsequent death of eight individuals (Crilly, 2007; Hippius, 1999). Clozapine was later reintroduced with a rigid monitoring protocol after several studies showed its efficacy and safety in treatment-resistant schizophrenia. Currently, it is used in the UK as second-line treatment for schizophrenia after a failed trial with two antipsychotic medications for an adequate time period (Farooq et al., 2011). Though, in some countries it is routinely used as first-line treatment (Tang et al., 2008; Wang & Li, 2012).

Antipsychotics as a Treatment of Challenging Behavior and Comorbid Psychopathology in ID Population

There is a high rate of prescription of psychotropic medication, including antipsychotics, among individuals with an ID (Deb, Unwin, & Deb, 2014; Scheifes, Egberts, Stolker, Nijman, & Heerdink, 2016). In fact, there are about 25–30% of all individuals with ID using services that regularly receive antipsychotics (Branford, 1994; Holden & Gitlesen, 2004a). This increases to 48% when you include challenging behaviors (Kiernan, Reeves, & Alborz, 1995). There are two main reasons for the use of antipsychotics among this population. First, antipsychotics are used to treat comorbid psychosis. Second, and perhaps more controversially, antipsychotics are used to manage challenging behaviors.

Studies have consistently reported that the rate of prescription of antipsychotics far exceed the expected prevalence of psychoses in individuals with ID (Perry et al., 2018; Robertson et al., 2000). For instance, of the 74% of the sample who were prescribed antipsychotics, 26% had documented evidence of psychotic features (e.g., bipolar, psychosis) whereas 48% did not (Perry et al., 2018). In a survey of 500 residents with ID, Robertson et al. (2000) reported significantly higher use of all psychotropics, especially typical antipsychotics, in those living in residential campuses (56%), versus those living in village communities (17%) or dispersed housing (27%).

Furthermore, residents having good mobility, with challenging behaviors, living in residential campuses, not having been diagnosed with epilepsy, and not having moved to the current residence from a family home predicted higher use of antipsychotics (Robertson et al., 2000). Similarly, having a diagnosis of autism and dementia or being older was also associated with the use of antipsychotics in individuals with ID (Sheehan et al., 2015). Nevertheless, challenging behaviors are commonly examined as a rationale for the higher use of antipsychotics. For instance, Holden and Gitlesen (2004a) indicate that nearly 50% of individuals with ID have been using psychotropic medications in the treatment of psychiatric disorder and/or challenging behaviors in the last 20 years. As such, although antipsychotic medications are often used to manage psychosis in the general population, it is evident that adults with ID are treated at a rate far exceeding that of recorded mental illness and that individuals with ID and challenging behaviors are significantly more likely to receive antipsychotics (Sheehan et al., 2015).

As such, there is concern that antipsychotics are overused and might often be prescribed for challenging behavior in itself rather than for diagnosed mental illness, despite inconsistent evidence of efficacy (Aman, Buitelaar, Smedt, Wapenaar, & Binder, 2005; Aman, De, Derivan, Lyons, & Findling, 2002; Duggan & Brylewski, 2004; Deb, Sohanpal, Soni, Lenotre, & Unwin, 2007; Duggan & Brylewski, 1999; La Malfa, Bertelli, & Conte, 2001; Scheifes, Stolker, Egberts, Nijman, & Heerdink, 2011). In fact, the most common reason for the prescription of antipsychotic medication to individuals with ID is the management of challenging behaviors (Aman, Sarphare, & Burrow, 1995; Matson & Mahan, 2010; Oliver-Africano, Murphy, & Tyrer, 2009). For example, clozapine has been shown to reduce aggression and self-injurious behavior independent of psychiatric diagnosis (Thalayasingam, Alexander, & Singh, 2004).

In a recent review of electronic case records, approximately 92% of a sample of 60 individuals with ID had been on antipsychotic medication. The vast majority of these individuals were prescribed antipsychotics to reduce challenging behaviors or psychotic symptoms; however, some were using them as a mood stabilizer or to reduce anxiety (Thalitaya, Reynolds, & Ismail, 2017). In a similar study exploring the association between prescription of psychotropic drugs, mental illness, and challenging behaviors in individuals with ID, 49% (of a sample of 33,016 adults with ID) had a record prescription of psychotropic drugs (Sheehan et al., 2015). Most antipsychotics were prescribed to individuals without a record of severe mental illness. In fact, challenging behavior was associated with the prescription of antipsychotics after adjusting for psychiatric diagnoses, suggesting that these drugs are being used to manage challenging behavior (Sheehan et al., 2015).

Commonly prescribed antipsychotics include risperidone, aripiprazole, olanzapine, chlorpromazine, clozapine, depixol, fluenthixol, zuclopenthixol, quetiapine, and benperidol (Thalitaya et al., 2017). Similar antipsychotics are reported in the literature as the most frequently reported psychotropic drugs include thioridazine, haloperidol, and chlorpromazine (Aman et al., 1995; Robertson et al., 2000) as well as risperidone, olanzapine, and quetiapine (Connor & Posever, 1998; Williams, Clarke, Bouras, Martin, & Holt, 2009). There are concerns due to whether the antipsychotics are actually effective in controlling challenging behaviors and there are a host of side effects associated with antipsychotics. Side effects of atypical antipsychotics are commonly identified as a concern with this population (Frighi et al., 2011; Mahan et al., 2010; Matson & Mahan, 2010). Thalitaya et al. (2017) reported common side effects of weight gain, sedation, hypersalivation, EPSE, hyperprolactinemia, hypertension, tremors, and hypercholesterolemia.

As such there are international guidelines for prescribing psychotropics for managing challenging behaviors (Deb et al., 2009) and for diagnosing mental health disorders in individuals with ID (Clarke & Gomez, 1999; Cooper, Melville, & Einfeld, 2003; Fletcher, Loschen, Stavrakaki, & First, 2007). For example, in 2006, the Royal College of Psychiatrists of the UK suggested that if there is no diagnosis of comorbid mental disorders and the challenging behaviors are a result of psychosocial factors, then use of antipsychotics should be used to alleviate short term risks to self and others, while other non-pharmacological programs are implemented to manage behavior. As such, researchers and clinicians assume that there will be a decline in use of antipsychotics (due to their questionable efficacy with challenging behaviors and reported side effects) and increased use of mood stabilizers, antidepressants, antianxiety medications given such international guidelines (Tsiouris et al., 2013). Recently it has been reported that current prescribing practice of antipsychotics in persons with ID in the UK is consistent with evidence-based practice and overall prescribing practice (Paton et al., 2011).

Antipsychotics as a Treatment of Psychosis in ID Population

The diagnosis of psychiatric disorders in individuals with ID is a complex process with difficulties increasing with lower IQ scores and poorer verbal communication (Thalayasingam et al., 2004). This would also mean that psychiatrists would have similar problems in establishing treatment refractoriness. Given the difficulty with diagnosis and treatment of psychosis in individuals with ID, it is not surprising that limited research exists. Many researchers outline the lack of studies on the efficacy of antipsychotics on schizophrenia or other psychotic disorder in individuals with ID (Duggan & Brylewski, 1999; Malfa, Lassi, Bertelli, & Castellani, 2006; Matson, Bielecki, Mayville, & Matson, 2003). There are also concerns regarding the greater risk of developing side effects and the impact of antipsychotics on learning and cognitive abilities of individuals with ID (Aman, 1993; Frighi et al., 2011; Mahan et al., 2010; Matson & Mahan, 2010).

Not only do individuals with ID receive psychotropic medications at high rates, but these drugs tend to be prescribed for many years (Yen, Lin, Loh, Shi, & Hsu, 2009), and with little or no current data collected to determine treatment efficacy (Matson & Neal, 2009). In a sample of 2069 adults with ID, almost half of the survey sample (45%) received antipsychotic medication, 39% of which was atypical antipsychotic, and the remaining 6% typical antipsychotic (Tsiouris et al., 2013). Tsiouris et al. (2013) noted the frequent use of antipsychotics for psychosis (70%) and bipolar disorder (67%).

Although typical antipsychotics such as haloperidol or chlorpromazine are most prescribed in ID, on a long-term basis they turn out to be less effective than new antipsychotics (Malfa et al., 2006). For instance, in the abovementioned sample of 2069 adults with ID, typical antipsychotics such as haloperidol, thioridazine, and chlorpromazine were infrequently prescribed, while atypical antipsychotics like risperidone, olanzapine, quetiapine, and aripiprazole were more commonly used (Tsiouris et al., 2013). This finding is consistent with other commonly used SGAs in the literature (Frighi et al., 2011; Leucht et al., 2009; Malfa et al., 2006). In a sample of 13 adults with mild ID and 8 with moderate ID and comorbid psychiatric disorders (with significant challenging behaviors in 10 cases), olanzapine and risperidone were well tolerated by individuals with ID and psychiatric disorders (with improvements in psychotic features), with only one case where olanzapine was discontinued due to side effects.

In a review of the literature, Connor and Posever (1998) concluded that atypical antipsychotics appeared better tolerated than typical antipsychotics, but there is a need for more research into their clinical efficacy for individuals with ID. Despite the efficacy for some antipsychotics, a Cochrane review did not find sufficiently detailed randomized controlled trials that examined antipsychotic medication versus placebo for individuals with both ID and psychosis, specifically schizophrenia (Duggan & Brylewski, 2004).

While there is limited evidence of antipsychotic medications, Clozapine has been shown to have a good response on treatment-resistant individuals with ID and DSM-IV diagnosis of schizophrenia, schizoaffective disorder, bipolar disorder, delusional disorder or other psychotic disorders (Ayub, Saeed, Munshi, & Naeem, 2015; Chakos, Lieberman, Hoffman, Bradford, & Sheitman, 2004; Farooq et al., 2011; Malfa et al., 2006; Thalayasingam et al., 2004; Singh, Singh, Kar, & Chan 2010). When at least two antipsychotics are not effective in treating the symptoms of psychosis, it is labelled as treatment-resistant psychosis. Individuals with ID who also suffer from psychosis are at a higher risk of developing treatment-resistant psychosis (Ayub et al., 2015). The extent of treatment resistance in individuals with ID is unclear (Ayub et al., 2015). Much of the evidence for treatment-resistance psychosis in ID is based on the fact that low IQ is a predictor of poor prognosis in psychosis and one manifestation of poor prognosis is treatment resistance (Thalayasingam et al., 2004).

A recent Cochrane review conducted by Ayub and colleagues (2015) revealed poorly designed randomized controlled trials (RCTs) that examined the efficacy and side effects of clozapine in individuals with ID and psychoses. Of 1224 titles and abstracts screens, 38 articles were initially included. However, these articles were subsequently excluded as they did not meet the inclusion criteria. Specifically, these studies were not RCTs. This makes sense as individuals with ID are a very heterogeneous population and psychiatric disorders are difficult to diagnose, which makes randomized controlled trials even more difficult to conduct. Nevertheless, small studies have shown that clozapine appears to efficacious and safe for individuals with ID (Antonacci & de Groot, 2000; Thalayasingam et al., 2004). Thalayasingam et al. (2004) found that clozapine may be used to effectively treat either refractory schizophrenia or bipolar affect disorder in individuals with ID. They highlighted that the dose of clozapine was comparable to those without ID and the effectiveness did not appear to diminish over time.

In line with the international guidelines, careful monitoring of side effects is recommended (Thalayasingam et al., 2004). The effect of psychotropic medication can be different in individuals with ID; that is, antipsychotics can have different (desired and undesired) side effects in different individuals. For instance, individuals with ID are reportedly more prone to side effects (Ayub et al., 2015). Common side effects of the use of clozapine include anemia and leukocytosis, drowsiness, hypersalivation, tremors, syncope, disturbed sleep or nightmares, restlessness, hypokinesia or akinesia, agitation, sedation, seizures, rigidity, akathisia, confusion, fatigue, insomnia, and hyperkinesia (Miller, 2000). Clozapine is also associated with weight gain (Leucht et al., 2009).

Psychological Treatment

Treatment models exist for individuals with comorbid major mental illness and ID. One of which is assertive community treatment (ACT), which was originally designed to support individuals with severe and enduring mental illness (Herman, 2014). The main goals of ACT include maximizing medication compliance, minimizing relapse, meeting basic social and occupational needs, enhancing quality of life, improving social and vocational functioning, promoting independent living skills, reducing carer burden, and facilitating community integration (Martin et al., 2005). Research has indicated that benefits are mixed (Balogh, Ouellette-Kuntz, Bourne, Lunsky, & Colantonio, 2008). Raftery et al. (2006) provided an intensive personalized support program for individuals with mild ID and comorbid psychosis over an 18-month period.

With regard to ecological treatments, these approaches are largely psychoeducational, tailored to particular presentations and levels of intensity. These approaches often target psychosis in a typically developing population, as opposed to in individuals with ID or developmental disabilities. There are no manualized approaches, but instead a variety of strategies are implemented together. Due to the idiopathic nature of psychosis, this approach is appropriate.

With regard to behavioral treatments, cognitive behavior therapy for psychosis (CBTp) has been implemented for typically developing individuals with psychosis. The cognitive behavior therapy model (combining cognitive exercises and behavioral strategies) has been extended for use with this population. In many ways, the standard CBT approach has been adjusted for use with psychotic thoughts (e.g., delusions, hallucinations). With regard to negative symptoms, a psychoeducational approach is taken, which focuses on behavioral support and traditional skills training. For positive symptoms, a more traditional CBT approach is taken, which focuses on identifying patterns and thoughts, and opting not to act upon them. The CBT package is not very different to traditional CBT, in that it focuses on both observable behavior and covert behavior. Despite many programs practicing this model, there is not a formalized treatment package available. Much of the research literature is conceptual and practice-based.

Morrison and Barratt (2010) conducted a Delphi study, which highlighted some of the key components of CBTp. It was found that there was not a clear consensus regarding the essential elements of treatment (Morrison & Barratt, 2010). No CBTp package has been found to consistently improve outcomes for individuals with ID and psychosis. Treatment packages that exist for comorbid ID/ASD are not well defined and are unclear in how they differ.

Support of CBTp for psychosis ranges from supportive to little or no support. Over 30 RCTs demonstrated positive outcomes for CBTp on positive and negative symptoms of schizophrenia (Wykes, Steel, Everitt, & Tarrier, 2008). In their review, Jauhar et al. (2014) found a small therapeutic effect for CBTp. In contrast, a meta-analysis of highly controlled CBTp found that CBTp does not reliably, if ever, lead to significant outcomes when compared to controls (Jones, Hacker, Cormac, Meaden, & Irving, 2012; Lynch, Laws, & McKenna, 2010; Newton-Howes & Wood, 2013). Various scales have been developed for use in CBTp, including the Cognitive Therapy for At Risk of Psychosis Adherence Scale (Bell et al., 2008), the Cognitive Therapy for Psychosis Adherence Scale (Rollinson et al., 2008), and the Cognitive Therapy Scale for Psychosis (Haddock et al., 2001). There is some evidence that CBTp can be effective in psychosis (with and without ID), though support is very limited. Future research is needed to determine the effectiveness of CBTp in individuals with ID and psychosis.

Support for acceptance and commitment therapy for psychosis (ACTp) ranges from having some benefit to minimal benefit in the general population. The approach found that there were some improvements displayed, though not much over psychosocial comparative treatment in typically developing individuals (Shawyer et al., 2017). Bach and Hayes (2002) found positive improvements compared to controls on self-report data in typically developing individuals. The ACTp approach is still in formative stages, though nothing to suggest as of yet that it provides anything beyond what CBTp offers. There are little standardized measures used in ACTp. ACTp is highly experimental at this point, without many well-controlled trials. Future research is needed on ACTp in the typically developing population, as well as expanding this research to include individuals with ID and psychosis.

In summary, ecological treatment models for psychosis (with or without developmental disabilities) do not lead to reliable differences in well-controlled trials. Some studies that indicate improvements with regard to self-reported data, though these are not highly controlled studies. Ultimately, these studies suggest these approaches lead to arguably small effects on real-world outcomes (e.g., hospital readmissions, symptom counts). No studies exist that directly compare CBTp to ACTp. As can be seen, the psychological treatment of psychosis in individuals with ID is an area where much more research is needed. Predominantly, treatment is pharmacological and therefore, research on psychological treatments are warranted.

Recommendations for Treatment

Recommendations for the treatment of psychosis in individuals include the following. First, the medication type that the individual is prescribed needs to be considered. There needs to be a focus on balancing the individual's needs with treatment and the side effect profile of the medication. Second, regular physical health checks, including cardiovascular and endocrine review should be conducted; for example, an ECG prior to starting antipsychotics and at regular intervals thereafter is useful to monitor any ECG abnormality (like QT prolongation). Third, medication needs to be monitored. This includes monitoring the benefit and response, as well as the side effects, and the possibility of reducing medication over time and as appropriate. Fourth, clinicians need to familiarize themselves with existing guidelines, such as the National Institute for Clinical Excellence (NICE) guidelines. Fifth, a full diagnostic evaluation with clear description of psychotic symptoms should be conducted. Sixth, there should be a clear statement outlined of the risks and use of antipsychotics, given the side effects. Seventh, informed consent to treatment needs to be given. Eighth, behavioral supports and therapy should be given, where appropriate.

Future Directions for Research

Research in this area is difficult as the literature is limited by inconsistent definitions, use of small or highly selected samples (Sheehan et al., 2015). Therefore, future researchers are encouraged to better define inclusion criteria (e.g., symptoms, adaptive functioning). Moreover, researchers should focus on RCTs (with double-blind vs. placebo or other medications) for clinical evidence to guide the medication for this population, especially given its use for those with challenging behaviors. Although pharmacotherapy is necessary to alleviate the symptoms of psychiatric disorders, it is only part of a coordinated comprehensive treatment plan.

There is limited research into cognitive impairment related to ID and psychotic disorders (McCarthy & Barbot, 2016), and as such further research is required to enable a better understanding of psychosis experienced by a person with ID. Further evidence is required to initiate effective, suitable treatment based on strong empirical evidence (McCarthy & Barbot, 2016).

Additional research is required to provide information about the extent to which existing support systems meet the needs of adults with ID with mental health disorders (Schűtzwohl et al., 2016). Research that contributes relevant information to the literature regarding mental health service provisions for adults with mild, moderate and profound ID is essential to guiding future policies. By creating a greater awareness of psychosis and the related genetic and environmental factors (Doherty & Owen, 2014; McCarthy &

Barbot, 2016) we may help to accumulate evidence that could support high quality clinical practice for those with ID and ASD (Allen, 2008; Canitano & Pallagrosi, 2017). Research evaluating a range of support systems would allow for comparison and consequently an improved understanding of the fundamental factors which contribute to an effective service provision for individuals with an ID or ASD who are diagnosed with psychotic mental health problem (Schűtzwohl et al., 2016). Further research into psychosis and ID could focus on support for health care practitioners, carers, and individuals with ID in reducing antipsychotic medication, if possible (McNamara et al., 2017; O'Dwyer et al., 2017).

Conclusion

This chapter provides an overview of psychosis in individuals with ID. Definitions of psychosis are discussed, as is the incidence and prevalence of psychosis in individuals with ID. The importance of studying psychosis is given, as well as the difficulties in diagnosing psychosis in individuals with ID. The risk factors of psychosis are given including baseline risks, early-life risks (in utero and during birth), childhood risks, and risks that develop later in life. Assessment of psychosis in ID is outlined, including recommendations for assessment. Following this, pharmacological and psychological treatment for psychosis in individuals with ID is discussed, including recommendations for treatment. Finally, future directions for research are suggested. In conclusion, psychosis in individuals with ID is an area where much more research is needed in order to better assess, understand, and treat psychosis.

References

Allen, D. (2008). The relationship between challenging behaviour and mental ill-health in people with intellectual disabilities: A review of current theories and evidence. *Journal of Intellectual Disabilities, 12*(4), 267–294.

Allington-Smith, P. (2006). Mental health of children with learning disabilities. *Advanced Psychiatric Treatment, 12*, 130–140.

Aman, M., Buitelaar, J., Smedt, G. D., Wapenaar, R., & Binder, C. (2005). Pharmacotherapy of disruptive behavior and item changes on a standardized rating scale: Pooled analysis of risperidone effects in children with subaverage IQ. *Journal of Child and Adolescent Psychopharmacology, 15*, 220–232.

Aman, M. G. (1993). Efficacy of psychotropic drugs for reducing self-injurious behavior in the developmental disabilities. *Annals of Clinical Psychiatry, 5*(3), 171–188.

Aman, M. G., De, S. G., Derivan, A., Lyons, B., & Findling, R. L. (2002). Double-blind, placebo-controlled study of risperidone for the treatment of disruptive behaviors in children with subaverage intelligence. *American Journal of Psychiatry, 159*, 1337–1346.

Aman, M. G., Sarphare, G., & Burrow, W. H. (1995). Psychotropic drugs in group homes: Prevalence & relation to demographic/psychiatric variables. *American Journal on Mental Retardation, 99*, 500–509.

American Psychiatric Association. (2013). Diagnostic and statistical manual of mental disorders (5th ed.) (DSM-5). American Psychiatric Association: Arlington, VA.

Antonacci, D. J., & de Groot, C. M. (2000). Clozapine treatment in a population of adults with mental retardation. *The Journal of Clinical Psychiatry, 61*(1), 22–25.

Axmon, A., Kristensson, J., Ahlström, G., & Midlöv, P. (2017). Use of antipsychotics, benzodiazepine derivatives, and dementia medication among older people with intellectual disability and/or autism spectrum disorder and dementia. *Research in Developmental Disabilities, 62*, 50–57.

Axmon, A., Sandberg, M., & Ahlström, G. (2017). Gender differences in psychiatric diagnoses in older people with intellectual disability: A register study. *BMC Psychiatry, 17*(1), 192.

Ayub, M., Saeed, K., Munshi, T. A., & Naeem, F. (2015). Clozapine for psychotic disorders in adults with intellectual disabilities. *The Cochrane Database of Systematic Reviews*, (9), CD010625-CD010625.

Bach, P., & Hayes, S. C. (2002). The use of acceptance and commitment therapy to prevent the rehospitalization of psychotic patients: A randomized controlled trial. *Journal of Consulting and Clinical Psychology, 70*(5), 1129–1139.

Bagalman, E., & Cornell, A. S. (2018). *Prevalence of mental illness in the United States: Data sources and estimates* (R43047). Congressional Research Service Report.

Bailey, N. M., & Andrews, T. M. (2003). Diagnostic criteria for psychiatric disorders for use with adults with learning disabilities/mental retardation (DC-LD) and the diagnosis of anxiety disorders: A review. *Journal of Intellectual Disability Research, 47*(s1), 50–61.

Bakken, T. L., Eilertsen, D. E., Smeby, N. A., & Martinsen, H. (2009). The validity of disorganized behavior as an indicator of psychosis in adults with autism and intellectual disability: A single case study. *Mental Health Aspects of Developmental Disabilities, 12*(1), 17–22.

Bakken, T. L., Friis, S., Løvoll, S., Smeby, N. A., & Martinsen, H. (2007). Behavioral disorganization as an indicator of psychosis in adults with intellectual disability and autism. *Mental Health Aspects of Developmental Disabilities, 10*(2), 37–46.

Bakken, T. L., & Høidal, S. H. (2014). Asperger syndrome or schizophrenia, or both? Case identification of 12 adults in a specialized psychiatric inpatient unit. *International Journal of Developmental Disabilities, 60*(4), 215–225.

Balogh, R., Ouellette-Kuntz, H., Bourne, L., Lunsky, Y., & Colantonio, A. (2008). Organising health care services for persons with an intellectual disability. *Cochrane Database of Systematic Reviews*, (4). Art. No.: CD007492.

Bell, K., Startup, M., French, P., Morrison, A. P., Bucci, S., & Fowler, D. (2008). Therapist treatment adherence in Cbt for people at ultra-high risk for psychosis: Development of a new rating scale. *Early Intervention in Psychiatry, 2*.

Bendall, S., Alvarez-Jimenez, M., Nelson, B., & McGorry, P. (2013). Childhood trauma and psychosis: New perspectives on aetiology and treatment. *Early Intervention in Psychiatry, 7*(1), 1–4.

Bourque, F., vander Ven, E., & Malla, A. (2011). A meta-analysis of the risk for psychotic disorders substantial variation in the treated incidence of psychotic disorders among first- and second-generation immigrants. *Psychological Medicine, 41*(5), 897–910.

Branford, D. (1994). A study of the prescribing for people with learning disabilities living in the community and in National Health Service care. *Journal of Intellectual Disability Research, 38*(6), 577–586.

Brylewski, J., & Duggan, L. (2001). Antipsychotic medication for challenging behaviour in people with learning disability. *The Cochrane Database of Systematic Reviews*, (3), CD000377.

Buckles, J., Luckasson, R., & Keefe, E. (2013). A systematic review of the prevalence of psychiatric disorders in adults with intellectual disability, 2003–2010. *Journal of Mental Health Research in Intellectual Disabilities, 6*(3), 181–207.

Canitano, R., & Pallagrosi, M. (2017). Autism spectrum disorders and schizophrenia spectrum disorders: Excitation/inhibition imbalance and developmental trajectories. *Frontiers in Psychiatry, 8*, 69.

Carthy, J., Hemmings, C., Kravariti, E., Dworzynski, K., Holt, G., Bouras, N., & Tsakanikos, E. (2010). Challenging behavior and co-morbid psychopathology in adults with intellectual disability and autism spectrum disorders. *Research in Developmental Disabilities, 31*(2), 362–366.

Chakos, M., Lieberman, J., Hoffman, E., Bradford, D., & Sheitman, B. (2004). Effectiveness of second-generation antipsychotics in patients with treatment-resistant schizophrenia: A review and meta-analysis of randomized trials. *Focus, 158*(1), 518–121.

Clarke, D. (2006). Psychiatric disorders and challenging behaviour. In A. Roy, M. Roy, & D. Clarke (Eds.), *The psychiatry of intellectual disability* (pp. 21–31). Oxford: Ratcliffe Publishing.

Clarke, D. J., & Gomez, G. A. (1999). Utility of modified DCR-10 criteria in the diagnosis of depression associated with intellectual disability. *Journal of Intellectual Disabilities Research, 43*(5), 413–420.

Cohen-Kettenis, P. T., & Pfäfflin, F. (2010). The DSM diagnostic criteria for gender identity disorder in adolescents and adults. *Archives of Sexual Behavior, 39*(2), 499–513.

Connor, D. F., & Posever, T. A. (1998). A brief review of atypical antipsychotics in individuals with developmental disability. *Mental Health Aspects of Developmental Disabilities, 1*, 93–101.

Conrad, A. M., Lewin, T. J., Sly, K. A., Schall, U., Halpin, S. A., Hunter, M., & Carr, V. J. (2017). Utility of risk-status for predicting psychosis and related outcomes: Evaluation of a 10-year cohort of presenters to a specialised early psychosis community mental health service. *Psychiatry Research, 247*, 336–344.

Cooper, S. A., & Bailey, N. M. (2001). Psychiatric disorders amongst adults with learning disabilities-prevalence and relationship to ability level. *Irish Journal of Psychological Medicine, 18*(2), 45–53.

Cooper, S. A., Melville, C. A., & Einfeld, S. L. (2003). Psychiatric diagnosis, intellectual disabilities and diagnostic criteria for psychiatric disorders for use with adults with learning disabilities/mental retardation (DC-LD). *Journal of Intellectual Disabilities Research, 47*(Suppl 1), 3–15.

Cooper, S. A., Smiley, E., Morrison, J., Allan, L., Williamson, A., Finlayson, J., … Mantry, D. (2007). Psychosis and adults with intellectual disabilities. *Social Psychiatry and Psychiatric Epidemiology, 42*(7), 530–536.

Cooper, S. A., Smiley, E., Morrison, J., Williamson, A., & Allan, L. (2007). Mental ill-health in adults with intellectual disabilities: Prevalence and associated factors. *The British Journal of Psychiatry, 190*(1), 27–35.

Crilly, J. (2007). The history of clozapine and its emergence in the US market: A review and analysis. *History of Psychiatry, 18*(1), 39–60.

Dean, K., & Murray, R. M. (2005). Environmental risk factors for psychosis. *Dialogues in Clinical Neuroscience, 7*(1), 69–80.

Deb, S., Kwok, H., Bertelli, M., Salvador-Carulla, L. U. I. S., Bradley, E., Torr, J., … Guideline Development Group of the WPA Section on Psychiatry of Intellectual Disability. (2009). International guide to prescribing psychotropic medication for the management of problem behaviours in adults with intellectual disabilities. *World Psychiatry, 8*(3), 181–186.

Deb, S., Matthews, T., Holt, G., & Bouras, N. (2001). *Practice guidelines for the assessment and diagnosis of mental health problems in adults who have intellectual disability*. London: European Association for Mental Health in Mental Retardation (EAMHMR).

Deb, S., Sohanpal, S. K., Soni, R., Lenotre, L., & Unwin, G. (2007). The effectiveness of antipsychotic medication in the management of behaviour problems in adults with intellectual disabilities. *Journal of Intellectual Disabilities Research, 51*, 766–777.

Deb, S., Thomas, M., & Bright, C. (2001). Mental disorder in adults with intellectual disability. 1: Prevalence of functional psychiatric illness among a community-based population aged between 16 and 64 years. *Journal of Intellectual Disability Research, 45*(6), 495–505.

Deb, S., Unwin, G., & Deb, T. (2014). Characteristics and the trajectory of psychotropic medication use in general and antipsychotics in particular among adults with an an intellectual disability who exhibit aggressive behaviour. *Journal of Intellectual Disability Research, 59*, 11–25.

Doherty, J. L., & Owen, M. J. (2014). Genomic insights into the overlap between psychiatric disorders: Implications for research and clinical practice. *Genome Medicine, 6*(4), 29.

Drake, R. E., Essock, S. M., Shaner, A., Carey, K. B., Minkoff, K., Kola, L., … Rickards, L. (2001). Implementing dual diagnosis services for clients with severe mental illness. *Psychiatric Services, 52*(4), 469–476.

Duggan, L., & Brylewski, J. (1999). Effectiveness of antipsychotic medication in people with intellectual disability and schizophrenia: A systematic review. *Journal of Intellectual Disability Research, 43*, 94–104.

Duggan, L., & Brylewski, J. (2004). Antipsychotic medication versus placebo for people with both schizophrenia and learning disability. *Cochrane Database of Systematic Reviews*, (4). (Art. No.: CD000030).

Emerson, E., Kiernan, C., Alborz, A., Reeves, D., Mason, H., Swarbrick, R., … Hatton, C. (2001). The prevalence of challenging behaviors: A total population study. *Research in Developmental Disabilities, 22*(1), 77–93.

Farooq, S., Nazar, Z., Irfan, M., Akhter, J., Gul, E., Irfan, U., & Naeem, F. (2011). Schizophrenia medication adherence in a resource-poor setting: Randomised controlled trial of supervised treatment in out-patients for schizophrenia (STOPS). *The British Journal of Psychiatry, 199*(6), 467–472.

Feinstein, C., Eliez, S., Blasey, C., & Reiss, A. L. (2002). Psychiatric disorders and behavioral problems in children with velocardiofacial syndrome: Usefulness as phenotypic indicators of schizophrenia risk. *Biological Psychiatry, 51*(4), 312–318.

Fletcher, R., Loschen, E., Stavrakaki, C., & First, M. (2007). *Diagnostic manual-intellectual disability (DM-ID): A textbook of diagnosis of mental disorders in persons with intellectual disability*. Kingston, NY: NADD Press.

Friedlander, R. I., & Donnelly, T. (2004). Early-onset psychosis in youth with intellectual disability. *Journal of Intellectual Disability Research, 48*(6), 540–547.

Frighi, V., Stephenson, M. T., Morovat, A., Jolley, I. E., Trivella, M., Dudley, C. A., … Barrow, B. (2011). Safety of antipsychotics in people with intellectual disability. *The British Journal of Psychiatry, 199*(4), 289–295.

Fusar-Poli, P., Borgwardt, S., Bechdolf, A., Addington, J., Riecher-Rössler, A., Schultze-Lutter, F., … Valmaggia, L. (2013). The psychosis high-risk state: A comprehensive state-of-the-art review. *JAMA Psychiatry, 70*(1), 107–120.

Gibson, L. E., Alloy, L. B., & Ellman, L. M. (2016). Trauma and the psychosis spectrum: A review of symptom specificity and explanatory mechanisms. *Clinical Psychology Review, 49*, 92–105.

Gründer, G., Hippius, H., & Carlsson, A. (2009). The "atypicality" of antipsychotics: A concept re-examined and re-defined. *Nature Reviews Drug Discovery, 8*(3), 197.

Haddock, G., Devane, S., Bradshaw, T., McGovern, J., Tarrier, N., Kinderman, P., … Harris, N. (2001). An investigation into the psychometric properties of the cognitive therapy scale for psychosis (CTS-Psy). *Behavioural and Cognitive Psychotherapy, 29*(2), 221–233.

Hassiotis, A., Barron, D. A., & Hall, I. (2009). *Intellectual disability psychiatry: A practical handbook* (1st ed.). Chichester, West Sussex: Wiley.

Heckers, S. (2009). Who is at risk for a psychotic disorder? *Schizophrenia Bulletin, 35*(5), 847–850.

Helverschou, S. B., Bakken, T. L., & Martinsen, H. (2009). The psychopathology in autism checklist (PAC): A pilot study. *Research in Autism Spectrum Disorders, 3*(1), 179–195.

Helverschou, S. B., Bakken, T. L., & Martinsen, H. (2011). Psychiatric disorders in people with autism spectrum disorders: Phenomenology and recognition. In J. L. Matson & P. Sturmey (Eds.), *International handbook of autism and pervasive developmental disorders* (pp. 53–74). New York, NY: Springer Science & Business Media.

Herman, D. B. (2014). Transitional support for adults with severe mental illness: Critical time intervention and its roots in assertive community treatment. *Research on Social Work Practice, 24*(5), 556–563.

Hippius, H. (1999). A historical perspective of clozapine. *The Journal of Clinical Psychiatry, 60*.(Suppl 12, 22–23.

Holden, B., & Gitlesen, J. P. (2004a). Psychotropic medication in adults with mental retardation: Prevalence, and prescription practices. *Research in Developmental Disabilities, 25*(6), 509–521.

Holden, B., & Gitlesen, J. P. (2004b). The association between severity of intellectual disability and psychiatric symptomatology. *Journal of Intellectual Disability Research, 48*(6), 556–562.

Hove, O., & Havik, O. E. (2010). Developmental level and other factors associated with symptoms of mental disorders and problem behaviour in adults with intellectual disabilities living in the community. *Social Psychiatry and Psychiatric Epidemiology, 45*(1), 105–113.

Jauhar, S., McKenna, P. J., Radua, J., Fung, E., Salvador, R., & Laws, K. R. (2014). Cognitive–behavioural therapy for the symptoms of schizophrenia:

Systematic review and meta-analysis with examination of potential bias. *British Journal of Psychiatry, 204*(01), 20–29.

Jones, C., Hacker, D., Cormac, I., Meaden, A., & Irving, C. B. (2012). Cognitive behavioural therapy versus other psychosocial treatments for Schizophrenia. *The Cochrane Database of Systematic Reviews, 4*, CD008712.

Jongsma, H. E., Gayer-Anderson, C., Lasalvia, A., Quattrone, D., Mulè, A., Szöke, A., … Berardi, D. (2018). Treated incidence of psychotic disorders in the multinational EU-GEI study. *JAMA Psychiatry, 75*(1), 36–46.

Kelly, M. (2013). *Understanding psychosis in individuals with intellectual disabilities*. Regional Support Services, Presentation September 7, 2013

Kessler, R. C., Chiu, W. T., Demler, O., & Walters, E. E. (2005). Prevalence, severity, and comorbidity of 12-month DSM-IV disorders in the National Comorbidity Survey Replication. *Archives of General Psychiatry, 62*(6), 617–627.

Khandaker, G. M., Dalman, C., Kappelmann, N., Stochl, J., Dal, H., Kosidou, K., … Karlsson, H. (2018). Association of Childhood Infection with IQ and adult nonaffective psychosis in Swedish men: A population-based longitudinal cohort and co-relative study. *JAMA Psychiatry, 75*(4), 356–362.

Kiernan, C., Reeves, D., & Alborz, A. (1995). The use of anti-psychotic drugs with adults with learning disabilities and challenging behaviour. *Journal of Intellectual Disability Research, 39*(4), 263–274.

Kildahl, A. N., Bakken, T. L., Holm, O. K., & Helverschou, S. B. (2017). Assessment of psychosis in ASD/ID: A case study. *Advances in Mental Health and Intellectual Disabilities, 11*(1), 17–23.

Kirkbride, J. B., Errazuriz, A., Croudace, T. J., Morgan, C., Jackson, D., Boydell, J., … Jones, P. B. (2012). Incidence of schizophrenia and other psychoses in England, 1950–2009: A systematic review and meta-analyses. *Plo S one, 7*(3), e31660.

Kirkbride, J. B., Hameed, Y., Ankireddypalli, G., Ioannidis, K., Crane, C. M., Nasir, M., … Spyridi, S. (2017). The epidemiology of first-episode psychosis in early intervention in psychosis services: Findings from the social epidemiology of psychoses in East Anglia [SEPEA] study. *American Journal of Psychiatry, 174*(2), 143–153.

Kozlowski, A. M., Matson, J. L., Sipes, M., Hattier, M. A., & Bamburg, J. W. (2011). The relationship between psychopathology symptom clusters and the presence of comorbid psychopathology in individuals with severe to profound intellectual disability. *Research in Developmental Disabilities, 32*(5), 1610–1614.

Krishnan, R. R., Kraus, M. S., & Keefe, R. S. (2011). Comprehensive model of how reality distortion and symptoms occur in schizophrenia: Could impairment in learning-dependent predictive perception account for the manifestations of schizophrenia? *Psychiatry and Clinical Neurosciences, 65*(4), 305–317.

La Malfa, G., Bertelli, M., & Conte, M. (2001). Fluvoxamine and aggression in mental retardation. *Psychiatric Services, 52*(8), 1105–1105.

Leucht, S., Kissling, W., & Davis, J. M. (2009). Second-generation antipsychotics for schizophrenia: Can we resolve the conflict? *Psychological Medicine, 39*(10), 1591–1602.

Lunsky, Y., Bradley, E., Durbin, J., & Koegl, C. (2008). A comparison of patients with intellectual disability receiving specialised and general services in Ontario's psychiatric hospitals. *Journal of Intellectual Disability Research, 52*(11), 1003–1012.

Lunsky, Y., Gracey, C., Bradley, E., Koegl, C., & Durbin, J. (2011). A comparison of outpatients with intellectual disability receiving specialised and general services in Ontario's psychiatric hospitals. *Journal of Intellectual Disability Research, 55*(2), 242–247.

Lynch, D., Laws, K. R., & McKenna, P. J. (2010). Cognitive behavioural therapy for major psychiatric disorder: Does it really work? A meta-analytical review of well-controlled trials. *Psychological Medicine, 40*(1), 9–24.

Madhavan, G. P. (2018). Psychotic symptoms in people with intellectual disability. *British Medical Journal*, 360.

Mahan, S., Holloway, J., Bamburg, J. W., Hess, J. A., Fodstad, J. C., & Matson, J. L. (2010). An examination of psychotropic medication side effects: Does taking a greater number of psychotropic medications from different classes affect presentation of side effects in adults with ID? *Research in Developmental Disabilities, 31*(6), 1561–1569.

Malfa, G. L., Lassi, S., Bertelli, M., & Castellani, A. (2006). Reviewing the use of antipsychotic drugs in people with intellectual disability. *Human Psychopharmacology: Clinical and Experimental, 21*(2), 73–89.

Martin, G., Costello, H., Leese, M., Slade, M., Bouras, N., Higgins, S., & Holt, G. (2005). An exploratory study of assertive community treatment for people with intellectual disability and psychiatric disorders: Conceptual, clinical, and service issues. *Journal of Intellectual Disability Research, 49*(7), 516–524.

Matson, J. L., & Neal, D. (2009). Psychotropic medication use for challenging behaviors in persons with intellectual disabilities: An overview. *Research in Developmental Disabilities, 30*(3), 572–586.

Mason, J., & Scior, K. (2004). 'Diagnostic overshadowing' amongst clinicians working with people with intellectual disabilities in the UK. *Journal of Applied Research in Intellectual Disabilities, 17*(2), 85–90.

Matson, J. L. (1995). *The diagnostic assessment for the severely handicapped revised (DASH-II)*. Baton Rouge, LA: Disability Consultants, LLC.

Matson, J. L. (1997). *The assessment for dual diagnosis (ADD)*. Baton Rouge, LA: Disability Consultants, LLC.

Matson, J. L., Bielecki, J., Mayville, S. B., & Matson, M. L. (2003). Psychopharmacology research for individuals with mental retardation: Methodological issues and suggestions. *Research in Developmental Disabilities, 24*(3), 149–157.

Matson, J. L., Kazdin, A. E., & Senatore, V. (1984). Psychometric properties of the psychopathology instrument for mentally retarded adults. *Applied Research in Mental Retardation, 5*, 81–89.

Matson, J. L., & Mahan, S. (2010). Antipsychotic drug side effects for persons with intellectual disability. *Research in Developmental Disabilities, 31*, 1570–1576.

Matson, J. L., Mayville, E. A., Lott, J. D., Bielecki, J., & Logan, R. (2003). A comparison of social and adaptive functioning in persons with psychosis, autism, and severe or profound mental retardation. *Journal of Developmental and Physical Disabilities, 15*(1), 57–65.

McCarthy, J. B., & Barbot, B. (2016). The need for research on intellectual disabilities and severe psychiatric disorders in children and adolescents. *Journal of Mental Disorders and Treatment, 2*(112), 2.

McGough, J. J., Smalley, S. L., McCracken, J. T., Yang, M., Del'Homme, M., Lynn, D. E., & Loo, S. (2005). Psychiatric comorbidity in adult attention deficit hyperactivity disorder: Findings from multiplex families. *American Journal of Psychiatry, 162*(9), 1621–1627.

McGrath, J., Sukanta, S., Chant, D., & Welham, J. (2008). Schizophrenia: A concise overview of incidence, prevalence, and mortality. *Epidemiologic Reviews, 30*(1), 67–76.

McNamara, R., Randell, E., Gillespie, D., Wood, F., Felce, D., Romeo, R., … Meek, A. (2017). A pilot randomised controlled trial of community-led ANtipsychotic drug REduction for adults with learning disabilities. *Health Technology Assessment, 21*(47), 1–92.

Messias, E., Chen, C.-Y., & Eaton, W. W. (2007). Epidemiology of schizophrenia: Review of findings and myths. *Psychiatric Clinics, 30*(3), 323–338.

Miller, D. D. (2000). Review and management of clozapine side effects. *The Journal of Clinical Psychiatry, 61*(Supple 8), 14–17.

Morrison, A. P., & Barratt, S. (2010). What are the components of CBT for psychosis? A Delphi study. *Schizophrenia Bulletin, 36*(1), 136–142.

Moss, S., & Hurley, A. D. (2014). Integrating assessment instruments within the diagnostic process. In *Handbook of psychopathology in intellectual disability* (pp. 43–61). New York, NY: Springer.

Moss, S., Prosser, H., Costello, H., Simpson, N., Patel, P., Rowe, S., … Hatton, C. (2002). Reliability and validity of the PAS-ADD checklist for detecting psychiatric disorders in adults with intellectual disability. *Journal of Intellectual Disability Research, 42*, 173–183.

Mungkhetklang, C., Crewther, S. G., Bavin, E. L., Goharpey, N., & Parsons, C. (2016). Comparison of measures of ability in adolescents with intellectual disability. *Frontiers in Psychology, 7*, 683.

Munir, K. M. (2016). The co-occurrence of mental disorders in children and adolescents with intellectual disability/intellectual developmental disorder. *Current Opinion in Psychiatry, 29*(2), 95.

Myrbakk, E., & von Tetzchner, S. (2008). Psychiatric disorders and behavior problems in people with intellectual disability. *Research in Developmental Disabilities, 29*(4), 316–332.

Newton-Howes, G., & Wood, R. (2013). Cognitive behavioural therapy and the psychopathology of schizophrenia: Systematic review and meta-analysis. *Psychology and Psychotherapy: Theory, Research and Practice, 86*(2), 127–138.

O'Dwyer, M., Peklar, J., Mulryan, N., McCallion, P., McCarron, M., & Henman, M. C. (2017). Prevalence, patterns and factors associated with psychotropic use in older adults with intellectual disabilities in Ireland. *Journal of Intellectual Disability Research, 61*(10), 969–983.

Oliver-Africano, P., Murphy, D., & Tyrer, P. (2009). Aggressive behaviour in adults with intellectual disability. *CNS Drugs, 23*(11), 903–913.

Patil, D., Keown, P., & Scott, J. (2013). A rising tide: The increasing age and psychiatric length of stay for individuals with mild intellectual disabilities. *Journal of Intellectual Disability Research, 57*(8), 783–790.

Paton, C., Flynn, A., Shingleton-Smith, A., McIntyre, S., Bhaumik, S., Rasmussen, J., … Barnes, T. (2011). Nature and quality of antipsychotic prescribing practice in UK psychiatry of intellectual disability services. *Journal of Intellectual Disability Research, 55*(7), 665–674.

Perälä, J., Suvisaari, J., Saarni, S. I., Kuoppasalmi, K., Isometsä, E., Pirkola, S., … Härkänen, T. (2007). Lifetime prevalence of psychotic and bipolar I disorders in a general population. *Archives of General Psychiatry, 64*(1), 19–28.

Perry, B. I., Cooray, S. E., Mendis, J., Purandar, K., Wijeratne, A., Manjubhashini, S., … Kwok, H. F. (2018). Problem behaviours and psychotropic medication use in intellectual disability: A multinational cross-sectional survey. *Journal of Intellectual Disability Research, 62*, 140–149.

Prosser, H., Moss, S., Costello, H., Simpson, N., Patel, P., & Rowe, S. (2002). Reliability and validity of the Mini PAS-ADD for assessing psychiatric disorders in adults with intellectual disability. *Journal of Intellectual Disability Research, 42*(4), 264–272.

Radua, J., Ramella-Cravaro, V., Ioannidis, J., Reichenberg, A., Phiphopthatsanee, N., Amir, T., … McGuire, P. (2018). What causes psychosis? An umbrella review of risk and protective factors. *World Psychiatry, 17*(1), 49–66.

Raftery, M., Burke, K., Murray, N., O'Duinn, O., Murray, I., & Hallahan, B. (2006). An intensive personalised support approach to treating individuals with psychosis and co-morbid mild intellectual disability. *Irish Journal of Psychological Medicine*, 1–11.

Reeve, S., Sheaves, B., & Freeman, D. (2015). The role of sleep dysfunction in the occurrence of delusions and hallucinations: A systematic review. *Clinical Psychology Review, 42*, 96–115.

Robertson, J., Emerson, E., Gregory, N., Hatton, C., Kessissoglou, S., & Hallam, A. (2000). Receipt of psychotropic medication by people with intellectual disability in residential settings. *Journal of Intellectual Disability Research, 44*, 666–676.

Rollinson, R., Smith, B., Steel, C., Jolley, S., Onwumere, J., Garety, P. A., … Fowler, D. (2008). Measuring adherence in CBT for psychosis: A psychometric analysis of an adherence scale. *Behavioural and Cognitive Psychotherapy, 36*(2), 163–178.

Sadock, B. J., & Sadock, V. A. (2009). *Concise textbook of child and adolescent psychiatry*. Philadelphia: Lippincott Williams & Wilkins.

Sass, L. A., & Parnas, J. (2003). Schizophrenia, consciousness, and the self. *Schizophrenia Bulletin, 29*(3), 427–444.

Scheifes, A., Egberts, T. C., Stolker, J. J., Nijman, H. L., & Heerdink, E. R. (2016). Structured medication review to improve pharmacotherapy in people with intellectual disability and behavioural problems. *Journal of Applied Research in Intellectual Disabilities, 29*, 346–355.

Scheifes, A., Stolker, J. J., Egberts, A. C., Nijman, H. L., & Heerdink, E. R. (2011). Representation of people with intellectual disabilities in randomised controlled trials on antipsychotic treatment for behavioural problems. *Journal of Intellectual Disabilities Research, 55*, 650–664.

Schützwohl, M., Koch, A., Koslowski, N., Puschner, B., Voß, E., Salize, H. J., … Vogel, A. (2016). Mental illness, problem behaviour, needs and service use in adults with intellectual disability. *Social Psychiatry and Psychiatric Epidemiology, 51*(5), 767–776.

Selten, J. P., Lundberg, M., Rai, D., & Magnusson, C. (2015). Risks for nonaffective psychotic disorder and bipolar disorder in young people with autism spectrum disorder: A population-based study. *JAMA Psychiatry, 72*(5), 483–489.

Singh, S. P., Singh, V., Kar, N., & Chan, K. (2010). Efficacy of antidepressants in treating the negative symptoms of chronic schizophrenia: meta-analysis. *The British Journal of Psychiatry, 197*(3), 174–179.

Shawyer, F., Farhall, J., Thomas, N., Hayes, S. C., Gallop, R., Copolov, D., & Castle, D. J. (2017). Acceptance and commitment therapy for psychosis: Randomised controlled trial. *The British Journal of Psychiatry, 210*(2), 140–148.

Sheehan, D. V., Lecrubier, Y., Sheehan, K. H., Amorim, P., Janavs, J., Weiller, E., … Dubar, G. (1998). The Mini-international neuropsychiatric interview (M.I.N.I.): The development and validation of a structured diagnostic psychiatric interview for DSM-IV and ICD-10. *Journal of Clinical Psychiatry, 59*(20), 22–33.

Sheehan, R., Hassiotis, A., Walters, K., Osborn, D., Strydom, A., & Horsfall, L. (2015). Mental illness, challenging behaviour, and psychotropic drug prescribing in people with intellectual disability: UK population based cohort study. *BMJ, 351*, 1–9.

Skokauskas, N., & Gallagher, L. (2010). Psychosis, affective disorders and anxiety in autistic Spectrum disorder: Prevalence and Nosological considerations. *Psychopathology, 43*(1), 8–16.

Smiley, E. (2005). Epidemiology of mental health problems in adults with learning disability: An update. *Advances in Psychiatric Treatment, 11*(3), 214–222.

Spauwen, J., Krabbendam, L., Lieb, R., Wittchen, H. U., & Van Os, J. (2006). Impact of psychological trauma on the development of psychotic symptoms: Relationship with psychosis proneness. *The British Journal of Psychiatry, 188*(6), 527–533.

Steel, Z., Marnane, C., Iranpour, C., Chey, T., Jackson, J. W., Patel, V., & Silove, D. (2014). The global prevalence of common mental disorders: A systematic review and meta-analysis 1980–2013. *International Journal of Epidemiology, 43*(2), 476–493.

Tandon, R., Gaebel, W., Barch, D. M., Bustillo, J., Gur, R. E., Heckers, S., … Van Os, J. (2013). Definition and description of schizophrenia in the DSM-5. *Schizophrenia Research, 150*(1), 3–10.

Tang, Y. L., Mao, P. X., Jiang, F., Chen, Q., Wang, C. Y., Cai, Z. J., & Mitchell, P. B. (2008). Clozapine in China. *Pharmacopsychiatry, 41*, 1–9.

Thalayasingam, S., Alexander, R. T., & Singh, I. (2004). The use of clozapine in adults with intellectual disability. *Journal of Intellectual Disability Research, 48*, 572–579.

Thalitaya, M. D., Reynolds, C., & Ismail, I. (2017). Study on the use of antipsychotic medication in an intellectual disability: Community caseload. *Psychitria Danubiana, 29*, 578–583.

Tone, E. B., Garn, C. L., & Pine, D. S. (2016). Anxiety regulation: A developmental psychopathology perspective. *Development and Psychopathology*, 1–34.

Tsakanikos, E., Bouras, N., Costello, H., & Holt, G. (2007). Multiple exposure to life events and clinical psychopathology in adults with intellectual disability. *Social Psychiatry and Psychiatric Epidemiology, 42*(1), 24–28.

Tsiouris, J. A., Kim, S. Y., Brown, W. T., Pettinger, J., & Cohen, I. L. (2013). Prevalence of psychotropic drug use in adults with intellectual disability: Positive and negative findings from a large scale study. *Journal of Autism and Developmental Disorders, 43*(3), 719–731.

Turner, T. H. (1989). Schizophrenia and mental handicap: An historical review, with implications for further research. *Psychological Medicine, 19*(2), 301–314.

Underwood, R., Kumari, V., & Peters, E. (2016). Appraisals of psychotic experiences: An experimental investigation of symptomatic, remitted and non-need-for-care individuals. *Psychological Medicine, 46*(6), 1249–1263.

United Nations General Assembly. (2006). *United Nations Convention on the Rights of Persons with Disabilities (UNCRPD)*. New York: UN General Assembly.

van Os, J., Rutten, B. P., & Poulton, R. (2008). Gene-environment interactions in schizophrenia: Review of epidemiological findings and future directions. *Schizophrenia Bulletin, 34*(6), 1066–1082.

van Os, J., & Tamminga, C. (2007). Deconstructing psychosis. *Schizophrenia Bulletin, 33*(4), 861–862.

Varese, F., Smeets, F., Drukker, M., Lieverse, R., Lataster, T., Viechtbauer, W., … Bentall, R. P. (2012). Childhood adversities increase the risk of psychosis: A meta-analysis of patient-control, prospective-and cross-sectional cohort studies. *Schizophrenia Bulletin, 38*(4), 661–671.

Wang, C., & Li, L. (2012). Proper use of clozapine: Experiences in China. *Shanghai Archives of Psychiatry, 24*(2), 108.

Watson, D. (2005). Rethinking the mood and anxiety disorders: A quantitative hierarchical model for DSM-V. *Journal of Abnormal Psychology, 114*(4), 522.

Welch, K. A., Lawrie, S. M., Muir, W., & Johnstone, E. C. (2011). Systematic review of the clinical presentation of schizophrenia in intellectual disability. *Journal of Psychopathology and Behavioral Assessment, 33*(2), 246–253.

Williams, H., Clarke, R., Bouras, N., Martin, J., & Holt, G. (2009). Use of atypical antipsychotics olanzapine and risperidone in adults with intellectual disability. *Journal of Intellectual Disability Research, 44*, 164–169.

Wing, L., Gould, J., & Gillberg, C. (2011). Autism spectrum disorders in the DSM-V: Better or worse than the DSM-IV? *Research in Developmental Disabilities, 32*(2), 768–773.

World Health Organization. (2004). *International statistical classification of diseases and related health problems* (Vol. 1). World Health Organization.

Wykes, T., Steel, C., Everitt, B., & Tarrier, N. (2008). Cognitive behavior therapy for schizophrenia: Effect sizes, clinical models, and methodological rigor. *Schizophrenia Bulletin, 34*(3), 523–537.

Yen, C. F., Lin, J. D., Loh, C. H., Shi, L., & Hsu, S. W. (2009). Determinants of prescription drug use by adolescents with intellectual disabilities in Taiwan. *Research in Developmental Disabilities, 30*(6), 1354–1366.

Yin, R. K. (2013). Validity and generalization in future case study evaluations. *Evaluation, 19*(3), 321–332.

Yung, A. R., Nelson, B., Thompson, A., & Wood, S. J. (2010). The psychosis threshold in ultra high risk (prodromal) research: Is it valid? *Schizophrenia Research, 120*(1), 1–6.

Yung, A. R., Phillips, L. J., Yuen, H. P., & McGorry, P. D. (2004). Risk factors for psychosis in an ultra high-risk group: Psychopathology and clinical features. *Schizophrenia Research, 67*(2), 131–142.

47

Evidence-Based Treatments for Comorbid Autism Spectrum Disorders

Karolina Štětinová, Michelle S. Lemay, Megan O. Bird, and Robert D. Rieske

Introduction

Autism spectrum disorder (ASD) is a neurodevelopmental disorder which presents with deficits in social communication and social interaction across settings. It includes restricted and repetitive patterns of behavior, interests, or activities. Symptoms typically become apparent in early childhood development and limit or impair everyday functioning (American Psychiatric Association, 2013). ASD first appeared in the third edition of the Diagnostic and Statistical Manual (DSM-III). Prior to the DSM-III diagnosis of ASD as a neurodevelopmental disorder, it was considered to be a dimensionally defined developmental disorder, both behaviorally and categorically. Understanding the neurological nature of the disorder has allowed for more accurate diagnosis and intervention (Mintz, 2016). Previously, the DSM included Asperger's disorder, which was added in 1994 to the DSM-IV, in addition to autistic disorder (Kite, Gullifer, & Tyson, 2012). However, with the update of the DSM-5, both Asperger's disorder and autistic disorder fall under the diagnosis of ASD, which allowed both previously separate diagnoses to fall into the same spectrum of symptoms, although the altered criteria now often excludes those with higher functioning forms of ASD (McPartland, Reichow, & Volkmar, 2012). Symptoms of ASD usually are first recognizable between 12 and 24 months of age. However, if developmental delays are severe, symptoms can be seen prior to 12 months; and if the symptoms are less severe, such as with high functioning autism, they may not be identified until after 24 months or until adulthood (American Psychiatric Association, 2013). Symptoms can include loss of social or language skills, delayed language development, odd play patterns, unusual communication, unusual sensory interests, and repetitive behaviors (Faras, Al Ateeqi, & Tidmarsh, 2010). The severity of symptoms is directly correlated to the individual's adaptive functioning.

Although scientists have still not been able to determine the exact cause of ASD, the prevalence of those diagnosed has increased significantly in recent years. It is reported that between 1970 and 2010, autism diagnoses have increased from 0.5 to 14.7 per 1000 children (Dave & Fernandez, 2014). There are multiple theories as to this increase, which include an increased awareness of the disorder, expansion of the diagnostic criteria, and an actual increase in ASD. Dave & Fernandez (2014), examined whether or not autism is increasing independently of other mental health disorders or if autism diagnoses are

---

K. Štětinová · M. S. Lemay · M. O. Bird
R. D. Rieske (✉)
Department of Psychology, Idaho State University, Pocatello, ID, USA
e-mail: riesrobe@isu.edu

J. L. Matson (ed.), *Handbook of Intellectual Disabilities*, Autism and Child Psychopathology Series, https://doi.org/10.1007/978-3-030-20843-1_47

displacing other disorders (i.e., diagnostic substitution) and found that there is, in fact, an increase in the true prevalence of autism. However, determining the true increase in prevalence in relation to the multitude of other factors involved in the diagnosis and treatment of ASD continues to be difficult.

There are three core areas of development that are affected by ASD: communication, social interaction, and behavior. In a 2010 article Faras, Ateeqi, and Tidmarsh explained that for preschool-age children with ASD, it is common to see communication impairment in the form of delayed or absent speech and a lack of nonverbal communication, such as little to no pointing and struggling to follow one's gaze or point. In school-age children, it is common to see persistent echolalia, or immediate repetition of words, muteness, and unusual/abnormal vocabulary for their age group. Adolescents are likely to struggle with communicating even if they have a wide vocabulary and understand grammar (Faras et al., 2010). According to Faras et al. (2010) they are also likely to talk *at* people instead of *with* people and provide excessive information about topics of their own interest. Their speech can be flat and repetitive and they may have a hard time understanding sarcasm or metaphors.

In regard to social impairment, Faras et al. (2010) stated that preschool-age children often lack response to others' facial expressions, imagination, initiation of activity, ability to share pleasure, and play with peers. School-age children often inappropriately attempt joint play and lack awareness of classroom rules and norms. Adolescents may have a hard time making friends and lack awareness of personal space or not tolerate others intruding on their own space.

The third area includes impairments in behavior, interests, and/or activities. These impairments for preschool-age children can include repetitive hand movements, inability to deal with change and repetitive play. Like preschool-age children, school-age children have a hard time coping with change. They may also struggle with imaginative play and creativity, as well as have an unusual response to sensory stimuli. Adolescents also have a hard time with imagination, unusual responses to sensory stimuli, and prefer a familiar routine. Adolescents with ASD also tend to have specific, narrow interests/hobbies (Faras et al., 2010). While all of these impairments are common, it is important to keep in mind that the effect ASD has on these core areas of development can be different for each person with ASD. Additionally, with proper interventions, the majority of these symptoms can be corrected or improved upon, especially when the intervention begins earlier in life.

Relationship Between ASD and ID

As previously stated, prevalence rates of ASD have steadily increased over the past decade, with 1 in 59 children being diagnosed in 2014 compared to 1 in 125 ten years earlier (Autism and Developmental Disabilities Monitoring Network, 2018). With these increased rates, the mental health and medical fields have been able to devote more research and time to the disorder and have formed a better understanding of what symptoms and comorbid disorders are seen. One of the disorders often seen with ASD is an intellectual disability (ID). In fact, based on a study from 2004, approximately 70% of individuals with ASD have a comorbid diagnosis of intellectual disability (La Malfa, Lassi, Bertelli, Salvini, & Placidi, 2004). Conversely, ASD is the second highest comorbid disorder with ID, with approximately 40% of those with a diagnosis of ID having a co-occurring diagnosis of ASD (Matson & Shoemaker, 2009).

Although this text describes in some detail the criteria and process related to a diagnosis of ID, it is important for the purposes of this chapter to explain in further detail the similarities and differences between ASD and ID and how the symptom presentation may be different depending upon severity of ASD symptoms or upon level of intellectual functioning. One of the most notable symptoms seen within both disorders is a language or communication deficit (Pinborough-Zimmerman et al., 2007). Although this does not mean that all those with either or both diagnoses will have a language/communication problem, people with either of these diagnoses have a strong tendency to fall under the umbrella of communication disorders. Another diagnostic criterium that is seen with both disorders is the developmental component. More specifically, both a diagnosis of ID and ASD require symptoms to be present in the early developmental period and are considered to be lifelong diagnoses (American Psychiatric Association, 2013).

As previously stated, the symptoms observed in an individual are strongly dependent upon the severity of ASD symptoms as well as the deficit in intellectual functioning. Additionally, having a comorbid diagnosis is also strongly dependent upon the severity of these symptoms. For example, the term “high-functioning autism” applies to individuals diagnosed with ASD without having a comorbid ID diagnosis. Conversely, individuals with “low functioning autism” have a comorbid diagnosis of ID. This is due to the fact that one’s cognitive ability directly affects their level of functioning in social situations and otherwise, which is tied to the diagnosis of autism. Due to the fact that we predominantly see mid-to-low functioning autism diagnosed with ID, we will be focusing predominantly on this population within this chapter. In terms of symptoms seen within a low functioning ASD-ID population, there is a trend towards increased rates of tantrums, repetitive, and avoidant behaviors compared to children with higher IQs (Tureck, Matson, Cervantes, & Konst, 2014). Similarly, research suggests that there are more individuals with impaired verbal and nonverbal communication with an ASD-ID population compared to an ASD only population (Deb & Prasad, 1994). A similar result is seen when comparing those with comorbid ASD-ID to those with a diagnosis of ID such that individuals with comorbid symptoms would respond less frequently to social presses than individuals with an ID diagnosis alone (Jackson et al., 2003). Lastly, we also see more severe deficits in adaptive behavior in those with a comorbid diagnosis of ASD-ID compared to either diagnosis alone (Bolte & Poustka, 2002; Fodstad & Matson, 2008; Matson, Mayville, Lott, Bielecki, & Logan, 2003).

The severity of symptoms within this population emphasizes the importance of therapeutic interventions to improve the quality of life and improved level of functioning for these individuals, but it also highlights the fact that typical interventions implemented for either of these diagnostic clusters will have to be modified in order to effectively treat someone with a comorbid ASD-ID diagnosis. There are several treatments that have been developed over the years to improve functioning and decrease negative symptoms (e.g., self-injurious behaviors, tantrums). Many of those treatments have been proven to be efficacious, although some non-efficacious treatments are still used today. The following sections will discuss various treatments including behavioral and cognitive-behavioral interventions, naturalistic intervention, early-intervention programs, social skills training, language training, as well as parent and peer training. These treatments have been shown to be effective in treating ASD populations and will further discuss how they may be modified for an ASD-ID population.

Behavioral and Cognitive-Behavioral Interventions

The largest group of evidence-based treatments suitable for individuals with ASD and ID diagnoses are behavioral and cognitive-behavioral interventions. This group includes but is not limited to the following interventions: antecedent-based intervention (ABI), discrete trial teaching (DTT), functional behavior assessment (FBA), functional communication training (FCT), and cognitive behavioral intervention (CBI).

Common ABA Components

Many of the above mentioned behavioral and cognitive-behavioral interventions are based on the traditional learning principles of applied behavior analysis (ABA), which is grounded in the ideology proposed by operant conditioning. The ABA approach was built on three theoretical frameworks stating that all human behaviors are learned, purposeful, and could be changed through modification of the client's environment (Hanley, 2012). The goal of the ABA approach is to promote learning and positively shape the client's behavior by reinforcing desirable behaviors and punishing the undesirable ones (Hanley, 2012). This goal can be achieved using a variety of behavioral components, which are essential for the ABA approach. Some of the commonly used ABA components include prompting, time delay, extinction, differential reinforcement, modeling, video modeling, visual support, scripting, and exercise.

Prompting and time delay. In the treatment of ASD and ID, many teachers, parents, and professionals will use prompts to help teach the child different things such as names of objects or people or appropriate item labels (e.g., "Show me the card with the bird"). Prompts have several types such as physical (e.g., pointing to a correct response), verbal (e.g., stating the correct response), modeling (e.g., showing the correct behavior), decreasing the number of possible choices, or hand-over-hand prompting (e.g., physically guiding the child to the correct response; Leaf, Leaf, Taubman, McEachin, & Delmolino, 2014). Time delay procedures are often implemented within a prompting framework by first having the discriminative stimulus and prompt presented simultaneously. After a number of trials with no time delay, the prompt is faded, with more time between the discrete stimulus and prompt, allowing for a greater opportunity for learning the discrete stimulus (Walker, 2008).

Extinction. By definition, extinction is a behavioral modification technique used to reduce the frequency of a specific behavior and is often used as a form of treatment for disorders in which a specific behavior is intended to be reduced such as in obsessive-compulsive disorders and for specific phobias (Wolff, Hupp, & Symons, 2013). Within an ASD/ID population, this treatment can help to reduce ritualistic behaviors or harmful behaviors by preventing the child from completing the behavior within the presence of the controlling stimulus such as by blocking a child from biting their arm any time they make an attempt. After a set of time in which the extinction is in place, the behavior will typically decrease.

Differential reinforcement. In order to treat problem behaviors, one method often implemented is differential reinforcement of an alternative behavior (DRA). DRA, often implemented with extinction includes attention given to an alternative behavior that the caregiver wants to see while ignoring the unwanted behavior, putting it on extinction (e.g., providing the child with positive attention and praise when appropriately requesting an item of food but ignoring the child when they are throwing a tantrum because they want the food; MacNaul & Neely, 2018). However, there are instances in which differential reinforcement cannot be implemented with extinction such as with aggression, although several studies continue to support the fact that DRA can be effective in treating unwanted problem behaviors in individuals with ASD (Athens & Vollmer, 2010; Ingvarsson, Kahng, & Hausman, 2008; Slocum & Vollmer, 2015). Additional methods of differential reinforcement include differential reinforcement of other behaviors (DRO) and incompatible behaviors (DRI) and implementation depends on the behavior being treated.

Modeling and video modeling. In vivo, or live, modeling has been implemented for decades in the establishment and modification of many different behaviors for children with ASD and ID alike. Children could learn appropriate behaviors by viewing another person, typically their caregiver, engaging in that same behavior and then they would practice and be reinforced for engaging in the modeled behavior. However, with new technology advancements, it is argued that video modeling is more effective in several ways including the ability to involve same-aged peers within the video modeling routine which may not be something possible within a traditional modeling routine (Wilson, 2013). Modeling cannot only improve behaviors such as toileting and similar self-care activities but can also help the child practice social-communication skills as well.

Visual support. Similar to video modeling, visual supports are tools created to help children with ASD. Visual supports include items with images or tangible objects that are intended to represent language (such as items or tasks) and are used as a method of communication. As an example, families can have a card with a picture of a cup on it representing a drink and when the child presents the card to a caregiver it indicates that they are thirsty. Another example of visual supports is the use of a visual schedule which includes pictures of tasks or activities that the child can see and manipulate to indicate the completion of a task. Visual schedules can address issues related to struggles with transitions between activities and help the child understand language tied to the completion of tasks. The use of visual supports in an ASD/ID population has been shown to reduce symptoms of cognitive and communication delay as well as social disabilities (Hayes et al., 2010).

Scripting. Scripting is often a component involved in social skills training in which the child is given a script or narrative that describes an activity and template for a conversation or may include only the initiation of a conversation. These scripts can be helpful to a child who struggles with social interactions but is also limiting because the child may not learn to generalize to different "scripts" and may become dependent upon the taught script (Bellini & Peters, 2008). However, script fading, in which the child practices fading the use of the script through sequential practices has been shown to be an effective method of helping the child generalize social interaction skills.

Exercise. The treatment of stereotypic behaviors such as hand-flapping or body rocking has successfully been done through the use of exercise. Although the underlying cause behind improvements in stereotypic behaviors with exercise are unclear, it is suggested that the sensation provided with exercises is similar to that of the behavior, or it could be due to implementing a competing behavior such as repeatedly tapping a ball instead of engaging in hand-flapping behaviors (Tse, Pang, & Lee, 2018).

The various approaches and components utilized are dependent upon the specific symptoms the child is presented with, and some have significantly more research than others, but all have been shown to provide some degree of improvement in ASD symptomology. The effectiveness of the traditional ABA approach was, for example, supported by McEachin, Smith, and Lovaas (1993) who found that ABA leads to improvements in cognitive functioning in children diagnosed with ASD. The following treatments utilize many of the components discussed, and although each varies in the implementation and approach, they are based on the basics of behaviorism.

Antecedent-Based Intervention (ABI)

An intervention closest to the traditional ABA approach is the antecedent-based intervention (ABI). The function of ABI is to modify the environment in order to enhance the client's efforts to engage in the desired behaviors. ABI operates on the principle of the three-term behavioral contingency, often called ABC, where A is the Antecedent, B is the Behavior, and C is the Consequence (Hart & Carr, 2016).

Antecedents are environmental stimuli which trigger a specific behavior of the client. Therefore, a behavior and the consequence linked to that behavior (reinforcement or punishment) follow up after an antecedent is presented in the environment. For this reason, the goal of ABI is to eliminate the presence of antecedents triggering an undesirable behavior through a variety of modifications done in the client's environment (Hart & Carr, 2016).

The first step of the modification process involves identification of the environmental stimuli which reinforce the undesirable behaviors. Subsequent removal of these stimuli ought to prevent the client from engagement in the undesirable behaviors. Advantageously, an environment free of antecedents triggering the undesirable behaviors prevents their interference with the learning process, facilitating the client's attention and motivation. Aside from modification of the client's environment, ABI utilizes components commonly found in the ABA approach including scripting, scheduling, and priming.

According to the report published by the National Professional Development Center on ASD (NPDC) which covers literature published between years 1990 and 2011, the effectiveness of ABI has been validated by 32 single-case studies (Wong et al., 2015). Empirical support regarding the effectiveness of ABI has been found for toddlers, preschool and school-aged children, and adolescents with ASD and ID diagnoses (Adcock & Cuvo, 2009; Butler & Luiselli, 2007; Conroy, Asmus, Sellers, & Ladwig, 2005; Dunlap & Plienis, 1991; Ladd, Luiselli, & Baker, 2009; Reinhartsen, Garfinkle, & Wolery, 2002; Schreibman, Whalen, & Stahmer, 2000; Sigafoos, Green, Payne, O'Reilly, & Lancioni, 2009).

ABI can also be effectively implemented in conjunction with other evidence-based treatments. For example, Stichter, Randolph, Kay, and Gage (2009) used ABI in combination with structural analysis (SA) to develop a treatment addressing difficulties in social and adaptive behaviors in three students diagnosed with autism. All three students were found to become more socially engaged with their classmates and teachers.

In summary, the goal of the antecedent-based intervention (ABI) is to modify the client's environment in order to minimize the occurrence of a specific event (antecedents) resulting in unwanted behavior. The ABI approach was found to have a great empirical base addressing various behavioral problems in children and adolescent clients diagnosed with ASD and ID.

Discrete Trial Training (DTT)

While ABI focuses on the antecedents preceding a specific behavioral response, the discrete trial training (DTT) method focuses more on the consequences of the behavior (i.e., reinforcement and punishment). The function of DTT is to provide a client with a gradual and achievable learning experience using scaffolding and repetition. DTT is typically implemented by a teacher or a behavioral specialist, who uses a scripted instructional process to promote client's learning.

When implementing DTT, the teacher should first identify the targeted complex behavior and break it down into small steps. The complex behavior is then taught step by step using discrete trials or teaching attempts (Smith, 2001). In DTT, it is very important to clearly define and script each step before any teaching trial begins. Scripting not only ensures consistency between the discrete trials but also allows for evaluation of the client's progress. As mentioned, consequences play an important part in the DTT intervention. Clients are reinforced for a successful competition of each step, but no reinforcement is provided for incorrect responses. Traditionally, discrete training trials have been divided into the five following steps (Smith, 2001):

1. Antecedent (i.e., teacher's actions).
2. Prompt.
3. Client's response.
4. Consequence for a correct and incorrect response.
5. Inter-trial interval (i.e., spacing between the trials).

According to the NPDC report, the effectiveness of DTT has been supported by 13 studies (for more detail, see Wong et al., 2015). Those studies included individuals diagnosed with ASD ages 3–11 years and were found to lead to improvements in social, academic, and language skills (Goldsmith, LeBlanc, & Sautter, 2007; Jahr, 2001; Jones, Feeley, & Takacs, 2007; Lang et al., 2011; McHugh, Bobarnac, & Reed, 2011; Summers et al., 2011; Wynn & Smith, 2003). The DTT teaching technique is a popular form of intervention due to its simple structure, consistency, and applicability to a variety of problematic behaviors.

Functional Behavior Assessment (FBA)

Functional behavioral assessment (FBA) and ABI share a similar task—determining the origins of a specific, often problematic, behavioral response. However, while ABI is a comprehensive intervention whose goal is to address the problematic behaviors, FBA is a standardized method used solely for the identification process. FBA, therefore, should not be perceived as a stand-alone treatment, but rather as a technique designed to assist in the development of individualized treatment plans. For optimal results, FBA should always be paired with an additional treatment addressing the problematic behaviors.

According to Walker, Lyon, Loman, and Sennott (2018) functions of behavior could be grouped into the following categories:

1. Escape—avoiding undesirable activity.
2. Attention.
3. Reinforcement—obtaining access to a reinforcer.
4. Sensory stimulation.

To ensure convergence of evidence during the identification process, FBA is often implemented by a functional assessment team composed of clinicians, speech therapists, teachers, school administrators, and the caregivers (Fettig, 2013). The FBA process follows six standardized steps starting with a description of the problematic behavior. Once the problematic behavior is clearly defined, the team collects the baseline data and formulates a hypothesis regarding the antecedents and reinforcers which maintain the problematic behavior. The hypothesis is then tested and data about the client's behavior are collected. If the FBA hypothesis is accurate, the team develops an intervention and continues monitoring its effectiveness (Fettig, 2013).

FBA was found to be effective when developing interventions for academic, adaptive, and communication skills, as well as when determining origins of aggression and self-injurious behaviors (Fettig, 2013; Minshawi, Hurwitz, Morriss, & Mcdougle, 2015). Even though FBA is a behavioral strategy rather than a complex intervention, its effectiveness has been widely documented, meeting the evidence-based practice criteria (Wong et al., 2015).

Functional Communication Training (FCT)

Many individuals diagnosed with ASD and ID face daily challenges when communicating their needs and thoughts (Heath, Ganz, Parker, Burke, & Ninci, 2015). functional communication training (FCT) was developed by Carr and Durand (1985) as an intervention designed to address these exact problems. The goal of FCT is to determine the origins of the communication difficulties and provide the client with appropriate substitute skills. For example, if a child client shows physical aggression towards a younger sibling every time the sibling starts playing with the client's trains, the goal of FCT would be to identify the antecedents leading to the physical aggression and teaching the client appropriate substitute behaviors (verbally or nonverbally asking for the trains or seeking out assistance from the caregiver).

Similar to FBA, FCT is typically implemented by a team composed of mental health professionals, speech therapists, teachers, and the caregivers. FCT also uses FBA methods when identifying the functions of problematic interpersonal behaviors. Once the function of the problematic behavior is determined, a functionally equivalent substitute (or replacement) behavior has to be selected by the team (Walker et al., 2018). The substitute behavior is then taught using differential reinforcement, prompting, and shaping.

Several literature reviews have been conducted in order to evaluate effectiveness of FCT for a variety of populations including individuals diagnosed with ASD and ID (Heath et al., 2015; Kurtz, Boelter, Jarmolowicz, Chin, & Hagopian, 2011; Tiger, Hanley, & Bruzek, 2008; Walker et al., 2018). The NPDC report by Wong et al. (2015) identified studies which included participants diagnosed with ASD between ages 3 and 18 years. Overall, FCT interventions have been found to be very effective when addressing maladaptive interpersonal communication skills in individuals with ASD and ID diagnoses.

Cognitive Behavioral Intervention (CBI)

A combination of cognitive and behavioral approaches resulted in the development of a popular intervention for addressing comorbid anxiety for individuals diagnosed with ASD. Cognitive behavioral intervention (CBI) is grounded in the beliefs that some level of cognition underlies all human behaviors and that human behaviors are purposeful. Modification of the cognition might, therefore, result in observable behavioral changes.

In practice, CBI relies on teaching strategies which assist the client with understanding their own thoughts and emotions. For example, when a client experiences physical symptom of anxiety, CBI is designed to help the client to examine its source. Comorbidity of anxiety disorders in individuals with ASD and ID is very common; according to Steensel, Bögels, and Perrin (2011), about 40% of children diagnosed with ASD have at least one diagnosis of anxiety disorder. Aside from anxiety, CBI was found to effectively address communication, social, and adaptive difficulties (Wong et al., 2015). In fact, the NPDC report identified one single-case design study (Singh et al., 2011) and three group design studies (Drahota, Wood, Sze, & Van Dyke, 2011; Sofronoff, Attwood, & Hinton, 2005; Sofronoff, Attwood, Hinton, & Levin, 2007) whose results suggested that CBI is an effective intervention for individuals between 6 and 18 years of age.

CBI is often implemented by a trained professional who assumes the role of a teacher helping the client to acquire insight and develop strategies addressing maladaptive thoughts and behaviors (Brock, 2013). CBI could be divided into two types of components, cognitive and behavioral. The cognitive components include strategies promoting the client's engagement in problem-solving and self-regulation. Implementation of the cognitive components could be summarized in the following steps (Brock, 2013):

1. Assisting the client to recognize and define the problem.
2. Modeling steps of problem-solving to the client.
3. Generating alternative strategies and substitute behaviors.
4. Evaluating possible consequences of the alternative strategies.
5. Selecting the most suitable strategy.
6. Evaluating the effectiveness of the selected strategy.

The behavioral components include reinforcement and contingency. Implementation of behavioral components could be summarized in following steps (Brock, 2013):

1. Scripting an intervention outline.
2. Identification of reinforcers.
3. Writing up a behavioral contract including a description of the desirable behaviors.
4. Explaining the contract to the client.
5. Rewarding the client for appropriate behavior.
6. Monitoring the client's outcomes.

Implementation of CBI should involve an interactive relationship between the teacher and the client, and the client should assume an active role in the learning process. CBI can also be implemented in conjunction with other interventions (e.g., social narratives) (Brock, 2013). In the implementation of CBI, it is important to take into account the verbal and cognitive abilities of the individual being treated and making adaptations when needed. In many cases of comorbid ASD and ID, CBI would not be the treatment of choice due to the barriers in obtaining insight into their cognitions and behaviors; however, those with mild (and even moderate) intellectual disabilities may benefit from CBI depending on verbal abilities and successful adaptations.

Naturalistic Interventions

Another group of evidence-based treatments commonly used in a population with overlapping ASD and ID diagnoses are naturalistic interventions. Those interventions are based on traditional ABA practices, utilizing behavioral treatment components such as reinforcement and modeling. However, compared to the traditional ABA approaches, naturalistic interventions are more interactive and developmentally oriented (Schreibman et al., 2015) and focus on learning that occurs in the child's natural environment.

Naturalistic interventions build on the ideas of famous developmental psychologists such as Lev Vygotsky and Jean Piaget, who proposed that children are active agents in their environment and that learning is most effectively facilitated through instructional scaffolding. They aim to enhance the naturally occurring learning process through gradual exposure to new skill sets as explained by Vygotsky's Zone of Proximal Development theory (Schreibman et al., 2015). The relationship between the trainer and the client is interactive and fluid, depending on the client's interests, motivation, and achievements. Because of this interactive structure, naturalistic interventions are mainly designed to address difficulties with social behaviors and communication, which are commonly found in individuals with comorbid ASD and ID diagnoses. Clients are also being reinforced for initiative, attention, and progress which further facilitates learning.

Advantageously, naturalistic interventions can be implemented at a very early age regardless of the client's level of language proficiency (Schreibman et al., 2015). In fact, clients receiving naturalistic interventions in early childhood were found to achieve significant improvement in specific learning as well as enhanced ability to generalize the learned knowledge in different situations (Carr & Kologinsky, 1983; McGee, Krantz, Mason, & McClannahan, 1983; McGee, Krantz, & McClannahan, 1985). Children diagnosed with ASD were also found to learn better when associations between the targeted behaviors and reinforces were natural and interrelated (e.g., saying "car" and receiving a car to play with versus saying "car" and receiving a piece of candy for correct labeling of the car; Schreibman et al., 2015; McGee et al., 1985).

Another advantage of naturalistic interventions is that they can be implemented by a broad team including not only the clinicians but also the caregivers, siblings, peers, and teachers. The results of a systematic review done by Trembath, Madler, and Hurdy (2016) support the idea that, for children with several types of disabilities, parents can effectively implement many different naturalistic interventions, leading to improvements in receptive and expressive language skills.

The umbrella of naturalistic interventions includes natural language teaching paradigm, incidental teaching, and pivotal response training (PRT). Natural language teaching paradigm traditionally relies on activities and play during which the trainer models usage of proper language to the client. While incidental learning typically focuses on the promotion of specific communication intent such as making a request for food, Pivotal Response Training takes interest in broader, "pivotal" areas of human development such as skills related to motivation and self-management. Researchers have found that children who underwent Pivotal Response Training exhibited improvements in self-management, engagement in peer interaction and symbolic play, creativity, and joint-attention behaviors (Pierce & Schreibman, 1995; Stahmer, 1995). In one clinical trial, Pivotal Response Training has also been found to be more effective with children diagnosed with ASD than the traditional, structured ABA approach (Mohammadzaheri, Koegel, Rezaee, & Rafiee, 2014).

The effectiveness of naturalistic interventions has been widely researched, meeting the criteria for evidence-based treatment for ID and ASD diagnoses (Wong et al., 2015). Naturalistic interventions were found to be effective in various age groups including preschool-aged children (Ingersoll, Lewis, & Kroman, 2007; Koegel, Camarata, Koegel, Ben-Tall, & Smith, 1998; McGee, Almeida, Sulzer-Azaroff, & Feldman, 1992; Yoder & Stone, 2006), school-aged children (Charlop, Schreibman, & Thibodeau, 1985; Charlop & Trasowech, 1991; Charlop & Walsh, 1986; Koegel et al., 1998; Laski, Charlop, & Schreibman, 1988; McGee et al., 1985), and adolescents (Hamilton & Snell, 1993).

There are four strategies that are key to the teaching style used by naturalistic interventions, which are not commonly present in traditional didactic teaching (McGee et al., 1983). Those strategies include waiting for the child's initiative (e.g., request for a toy), teaching in context of activities enjoyable to the child such as play, usage of stimuli and reinforces selected and preferred by the child, and promotion of language by variation of prompting strategies dependent on the child's initiating behavior (McGee et al., 1983). In addition to these four strategies, steps for implementation include: identifying the target behavior and the intervention setting, collecting baseline data, training the team members, eliciting the desired behavior through environmental rearrangement, and data collection (Franzone, 2009).

Early Interventions

The Early Start Denver Model (ESDM)

The early start Denver model (ESDM) was originally developed by Sally Rogers, Ph.D., and Geraldine Dawson, Ph.D., as an intervention suitable for clients with ASD diagnosis between ages 12 and 36 months (Rogers & Dawson, 2010). The ESDM mainly builds on four intervention approaches designed for ASD clients including the Denver Model, Dawson's social motivation hypothesis of autism, pivotal response training (PRT), and the Rogers and Pennington's model of interpersonal development in autism (Rogers & Dawson, 2010).

Similar to naturalistic interventions, the ESDM is developmentally oriented, based on traditional ABA techniques, and utilizes settings and behaviors natural for the client. However, the focus of the ESDM is more complex, addressing a variety of developmental milestones including imitation, theory of mind, play, verbal and nonverbal language, and other socially relevant abilities (Baril & Humphreys, 2017). The ESDM promotes the acquisition of these abilities through structured and intensive education, which can be modified for the needs and developmental level of the client (Rogers & Dawson, 2010). The individualized ESDM curriculum is then taught through play and activities enjoyable for the client. Components utilized by the ESDM may include prompting, fading, and chaining behaviors.

While naturalistic interventions may be primarily implemented by the caregivers and the teachers, the ESDM has to be implemented by a professional with ESDM training and certification. Implementation of the ESDM may be guided by a multidisciplinary team including psychologists, language pathologists, and physicians. The process of implementation begins with a collection of the baseline data, which provides the starting point for the individualized ESDM intervention. The ESDM Curriculum Checklist is used for the assessment of baseline abilities of the client (Rogers & Dawson, 2010). The client's abilities are typically reassessed in 12-week intervals in order to record and evaluate the client's progress and the effectiveness of the intervention (Rogers & Dawson, 2010). The caregivers may be provided with a specialized Training Manual explaining the ESDM intervention strategies.

The ESDM has a strong empirical basis and has been recommended by the National Research Council (2001) as an effective intervention for toddlers diagnosed with ASD. According to Waddington, van der Meer, and Sigafoos (2016) who created a systematic review of 15 studies with a total of 209 participants, the ESDM has been linked to positive client outcomes for language, cognitive and social skills including initiation, imitation, and attentiveness (Dawson et al., 2010, 2012; Eapen, Črnčec, & Walter, 2013; Estes et al., 2015; Fulton, Eapen, Crnčec, Walter, & Rogers, 2014; Vismara, Colombi, & Rogers, 2009; Vismara, McCormick, Young, Nadhan, & Monlux, 2013; Vismara & Rogers, 2008; Vivanti et al., 2014; Vivanti, Dissanayake, Zierhut, Rogers,, & Team, 2013). Nonetheless, the systematic review reported mixed findings regarding the effectiveness of the ESDM in relation to other adaptive behaviors as well as the severity of ASD symptoms (Waddington et al., 2016).

State Early Intervention Programs

One of the most common resources utilized and available to infants and toddlers with developmental disabilities is the individual states early intervention program. Currently, every state (and eligible territory) in the United States participates in the federal grant Early Intervention Program for Infants and Toddlers with Disabilities (0–3 years of age) which is a component (Part C) of the Individuals with Disabilities Education Act

(IDEA, 2004). Every state is responsible for developing their early intervention program following guidelines regarding structure, evaluation, assessment, and identification; however, in regard to treatment approaches, states differ quite dramatically. Although the goals of these state programs follow those established by Part C of IDEA (enhance development, reduce educational costs, minimize the need for special education and institutionalization, maximize independent living, etc.) the method by which each state works towards these goals can be quite different. Because of these differences, it is not currently possible to describe state early intervention programs as a whole, but rather to discuss individual treatment modalities that various states may incorporate into their state-run early intervention program. However, there are components and concepts that are relatively consistent across all state early intervention programs including eligibility determination, the Individualized Family Service Plan (IFSP), and transition services from early intervention (IDEA-Part C) to special education (IDEA-Part B).

Identification and eligibility are one of the goals at the forefront of state early intervention programs. IDEA requires that each state participates in a type of "Child Find" program that helps to identify and evaluate all children with disabilities between 0 and 21 years of age. While many children with ASD may not be identified in time to receive services in state early intervention programs, those with comorbid ID and ASD are more likely to be identified within this age range and qualify for services. Eligibility for services does vary from state to state and is based on the state's definition of developmental delay. Additionally, some states extend services to those found to be "at risk" for disabilities while others may not.

Once a child is found to be eligible for services, according to state guidelines, all families will receive an Individualized Family Service Plan or IFSP. The IFSP is meant to guide the early intervention process and determine which available services will be most beneficial for the child and their family. For children with ASD/ID these services may vary widely from state to state and the availability of ASD specific programming may be even further limited in many states. One of the major commonalities amongst states provided by Part C of IDEA is the push to provide services in naturalistic environments. This most often takes place in the home but could also take place in other community settings that include children without disabilities. Because of this, states that are providing ASD specific treatments as part of their early intervention programming have leaned away from less naturalistic treatments such as Discrete Trial Training and more towards naturalistic programs such as the Early Start Denver Model or similar programs as well as those that promote parent involvement and parent-child interaction.

Due to the time that it often takes to identify children with disabilities, access to services through the state early intervention program is often short-lived. A final component of these programs is to ensure an effective transition from early intervention programs into special education programs. This period of time is an area in which families often fall into gaps or lapses in service provision either due to differing eligibility criteria from early intervention to special education or due to ineffective transition planning and lack of available resources. An effective transition generally starts 4–6 months prior to the child's third birthday and includes a meeting between the early intervention program, the preschool special education program, and the family and after eligibility is determined an Individuated Education Plan (IEP) is developed to help guide the educational needs of the child into preschool.

Social Skills Training

Severe social impairments are a common concern in the ASD population. Therefore, social skill interventions are essential. Social skills interventions are treatments that focus on social behaviors, such as eye contact, appropriate speech content, and intonation, appropriate motor movements, appropriate facial affect, etc. Without effective intervention, social impairments are likely to become more severe with age (Matson,

Matson, & Rivet, 2007). A study performed by Taheri, Perry, and Minnes (2016) examined the social interaction between developing children and adolescents, those with ID, and those with ASD-ID. The study found that those with ASD-ID participated in fewer social activities than their typically developing peers and peers with ID. Typically developing participants had significantly more friends than participants with ID and ASD-ID. Finally, participants with ID were more likely to have friends or even a best friend compared to the participants with ASD-ID (Taheri et al., 2016). Minimal social interaction is common in individuals with ASD-ID; and, in order to obtain a range of basic social skills, they require intensive instruction (Plavnick, Kaid, & MacFarland, 2015).

Various interventions have been used in the treatment of social deficits in this population. Evidenced-based practices for social skills treatments include self-management, social narratives, and socials skills training. Self-management consists of instructions that focus on teaching learners with ASD to discriminate between appropriate and inappropriate behaviors. The learners are also taught to monitor and record their own behaviors accurately. When the learners behave appropriately they then reward themselves. Self-management has been found effective for learners with ASD ranging from age 3 to 22 (Wong et al., 2015 with specific studies examining its effectiveness).

A study conducted by Newman and Eyck (2005) attempted to teach three participants with ASD self-management skills. The goals were to help them learn to make initiations with others and require less interventionist supervision, and in the study, participants were able to learn to make social initiations to an adult (Newman & Eyck, 2005). Another study found self-management procedures to be effective in reducing inappropriate vocalizations in it participants (Kern, Marder, Boyajian, Elliot, & McElhattan, 1997). Koegel, Koegel, Hurley, and Frea (1992) found that lack of social responsivity can be successfully treated with self-management procedures. This is beneficial in schools, homes, and in the community because it permits less direct contact, is teachable in a short time, and provides relatively quick results (Koegel et al., 1992).

Social narratives were first introduced in 1993 by a former teacher named Carol Gray (Kokina & Kern, 2010) and typically give short descriptions of social situations. Criteria for social narratives must include several types of sentences, such as descriptive, perspective, directive, cooperative, affirmative, and control. The control sentences are written by the child in order to allow for an understanding of the child's own strategies to recall and use information. It is recommended that terms such as "usually," "sometimes," and "probably" be used (Kokina & Kern, 2010). The descriptions may include pictures and other visual aids and the learners are provided with relevant cues and examples of appropriate responding. Learners are also provided with explanations of the thoughts and feelings of others (Sam & Team, 2015). Social narratives have shown to be effective for those with ASD ranging from ages 3 to 18 years (Wong et al., 2015) with studies performed to examine the extent of their effectiveness.

Social narratives, also known as social stories, are one of the most common evidence-based procedures used in treatments for individuals diagnosed with ASD. Teachers who have used this in their classrooms have perceived social narratives to be effective in changing behavior (Leaf et al., 2016). However, meta-analyses have found limitations with research regarding social narratives (Kokina & Kern, 2010; Leaf et al., 2016). Often the studies included other procedures as well, making it difficult to distinguish the effectiveness of social narratives. Leaf et al. (2016) decided to compare one behavior intervention called the "cool vs. not cool" procedure (CNC) and social narratives with an individual diagnosed with ASD and the results indicated that CNC was more effective than social narratives. The data indicates that for the social narratives there were some improvements from the baseline level to the intervention sessions, though the participant never showed more than 80% of the steps correctly (Leaf et al., 2016). Kokina and Kern (2010) conducted a meta-analysis also examining the effectiveness of social narratives and found that social narrative interventions were more effective for targeting behavior reduction compared to teaching social skills.

Social skills training teaches people with ASD how to appropriately interact with others around them. Instructions of basic concepts, such as role-playing and feedback, are often used to help the learners obtain and practice communication and social skills. It is used to promote positive interactions with the learners' peers and has been effective for learners with ASD from age 3 to age 22 (Wong et al., 2015). A combined theory-of-mind and social skills training program was found to greatly improve positive social interactions of the participants across multiple settings (Hua, Ya-yu, Shuling, & Cartledge, 2008). Another study found that young children with ASD showed increased frequency and duration of interactions with typically developing peers after a social skills training. The participants were taught social skills that would be beneficial in sundry situations. Free play was implemented after the training to see if the training procedures were effective (Gonzalez-Lopez & Kamps, 1997). Social skill interventions are important in helping those with ASD-ID function in their lives. Self-management, social narratives, and socials skills training are just a few of the evidenced-based practices that are used to help individuals with ASD-ID. Overall, social skill interventions have been found effective in helping those with ASD-ID improve their social skills.

Language Training

Computer-Aided Instruction (CAI)

Computer-Aided Instruction (CAI), falling under a broader category of evidence-based treatments called Technology-Aided Instruction and Intervention (TAII), has been developed as an instructional method promoting the development and facilitation of interpersonal communication in individuals diagnosed with ASD and ID. CAI has been primarily designed to assist school-aged children, addressing school-readiness, academic skills, and communication. Traditionally, the implementation of CAI is led by a team including the caregivers, clinicians, and the school staff. Through the usage of a computer, CAI aims to provide the learner with systematic instructions, making the learning process easier and more achievable (Collet-Klingenberg, 2009). Various CAI programs are available on the market, depending on the learner's age and targeted skill. The following steps of CAI implementation were discussed by Collet-Klingenberg (2008):

> *Identification of the targeted skill to be learned (e.g. increased vocabulary), Collection of the baseline data, Identification, installation, and familiarization with an appropriate software, Competition of a task analysis of steps for using the selected software, Introducing the learner to software, Providing the learner with multiple opportunities to use the computer and ongoing support, and Collecting data on the acquisition of the target skill.*

The effectiveness of CAI itself has been supported by six studies, which included participants between preschool and middle-school ages with a variety of communication problems and language delays (Bosseler & Massaro, 2003; Hetzroni & Shalem, 2005; Hetzroni & Tannous, 2004; Massaro & Bosseler, 2006; Moore & Calvert, 2000; Silver & Oakes, 2001).

Nevertheless, since the number of treatments utilizing modern technologies expanded in recent years, CAI has been included into a broader Technology-Aided Instruction and Intervention (TAII) evidence-based category (Wong et al., 2015). TAII relies on the use of an electronic device, including computers, tablets, smartphones, and other technology, which can assist individuals diagnosed with ASD. The idea that an ASD population benefits from using modern technologies has been supported by 20 group-design and case studies (Beaumont & Sofronoff, 2008; Choi, O'Reilly, Sigafoos, & Lancioni, 2010; Cihak, Wright, & Ayres, 2010; Faja, Aylward, Bernier, & Dawson, 2007; Stromer, Mackay, Howell, McVay, & Flusser, 1996; Whalen et al., 2010).

Speech Generated and Communication Devices

Another sub-category of TAII evidence-based treatments suitable for individuals with comorbid ASD and ID diagnoses is Speech Generated and Communication Devices (SGCD). SGCD include a variety of electronic, portable devices whose aim is to assist the individual with daily communication. This is done through synthetic speech produced by the device across different settings. Franzone and Collet-Klingenberg (2008) discussed the following steps for implementation of SGCD:

> *Identifying and setting up the device, Identification of settings where the device will be initially used, Identification of the appropriate vocabulary, Introduction of the device to the learner, Providing as few prompts as the learner requires, Honoring the communication, Reducing prompts as soon as possible, Increasing the number of settings where the SGD is used, and Increasing vocabulary.*

Skills that were found to be effectively addressed by SGCD are the initiation of conversation, expressive language, nonverbal language, reading, and mathematics (Franzone & Collet-Klingenberg, 2008). In fact, the utility of SGCD for school-aged children has been widely supported (Olive et al., 2007; Olive, Lang, & Davis, 2008; Parsons & La Sorte, 1993; Van Acker & Grant, 1995).

Picture Exchange Communication System (PECS)

The Picture Exchange Communication System (PECS) is an augmentative communication system that uses pictures/icons (Ganz, Davis, Lund, Goodwyn, & Simpson, 2012). It is used to help initiate communicative exchanges and interaction for those with limited functional communication skills within a social context (Sam & AFIRM Team, 2016). PECS is designed to help learners with autism and other developmental disorders. It has been predominately used with children, specifically preschool-aged children (Ganz et al., 2012). There are six phases of training and they get progressively more complex. Learners with severe ASD are often only taught the first three phases (Carre, LeGrice, Blampied, & Walker, 2009; Ganz et al., 2012). While PECS is primarily picture-based, it does use sentence strips combined with pictures in more advanced phases. This is to increase complexity and potential utility of communication. It is suggested that PECS is particularly well-suited for those with ASD because of its primarily visual nature. PECS attempts to reduce the uncertainty in communication for those with ASD by using symbols that have a one-to-one ratio correspondence with objects, people, and concepts (Ganz et al., 2012). Ganz et al. (2012) also states that along with the reduction of ambiguity in communication, there is also an opportunity for recognition of meaning instead of requiring recall.

Many single case research designs have found PECS is indeed effective in improving communication skills in individuals with ASD. Not only has it been found to be effective, but for some learners PECS has also been found to be *more* effective than other forms of alternative and augmentative communication (AAC). PECS has also been found to be more effective than other forms of picture-based AAC and just as effective as speech generating devices (SGDs) (Ganz et al., 2012). PECS is a beneficial treatment in helping learners with ASD and other developmental disabilities improve their communication skills.

Parent and Peer Interventions

In addition to the many interventions used for children with ASD-ID diagnoses, two effective treatments to implement in the home or at school are Parent-Implemented Intervention (PII) and Peer-Mediated Instruction and Intervention (PMII). The emphasis on these two types of interventions is based on research indicating four characteristics of efficacious interventions for the treatment of infants and younger children with a developmental disability such as autism or intellectual/developmental delay. The characteristics described by Wallace and Rogers (2010) include:

1. Parent involvement in interaction such as continuous coaching and sensitivity to cues provided by the child.
2. Individualized intervention planning tailored to the child's specific needs.
3. Attending to a broad range of targeted stimuli.
4. Providing treatment as early in the lifespan as possible and providing treatment at least once per week for an extended duration.

Based on these qualities of an effective intervention, PII meets all of these criteria in addition to added benefits such as decreased family stress (Koegel & Schreibman, 1996). Parent-implemented intervention is a form of behaviorally based intervention in which the clinician helps to teach the parent(s) ways in which they can help their child improve specific behaviors outside of the office. With parents being the identified "agents of change," it allows the child to generalize their gains across multiple settings, the parent is able to implement the intervention(s) throughout the day, it provides the parent with a greater sense of success, it is more cost-effective, and it has even been shown to have improved gains (Ingersoll & Gergans, 2007).

The majority of the current literature on PII has focused predominantly on socially based interventions such as social communication and imitation. For example, when looking at outcomes of parents that have been coached and received reciprocal imitation training (RIT), children's correct use of imitation and descriptive language increased significantly (Zaghlawan & Ostrosky, 2016). Similarly, children whose parents received RIT and were implementing PII generalized their object imitation skills to the homes and parents reported that their child's social-communication skill had improved (Ingersoll & Gergans, 2007). Children also will experience greater gains in verbal skills, communication and daily living skills, and social communication skills when provided with individual parent-provided early social interaction in which parents are trained on ways to improve and increase the interactions with their nonverbal children with ASD (Wetherby et al., 2014). In addition to these treatments being effective for improving symptoms of ASD, PII has also been shown to be effective in treating children with developmental delays or intellectual disabilities to help improve language (Akamoglu & Meadan, 2018).

Not only have treatments been developed to be implemented by parents; peer interventions have also been shown to be effective in treating social symptoms of children with diagnoses of ASD and ID. Peer-mediated intervention (PMI) is described as a treatment that teaches typically developing peers ways to interact and increase social skills in children with a primary diagnosis of ASD (Płatos & Wojaczek, 2018). This type of treatment is beneficial in the development of social skills because most children with a diagnosis of ASD typically spend most of their time engaged in solitary play and struggle to form peer relationships. By implementing PMI, peers are taught either by trained teachers or research assistants how to use strategies such as inviting the child to play, offering assistance, offering to share, and demonstrating warmth to the child with the primary ASD diagnosis (Katz & Girolametto, 2013). This form of treatment, which has been shown to be effective across childhood, from preschool to middle school, is beneficial because it allows the child to interact with his or her same-aged peers in their natural setting (Collet-Klingenberg, Neitzel, & Laberge, 2012; Katz & Girolametto, 2013). In addition to having frequent peer interactions which provide the child with a sense of social support, research has also suggested that increased regular social interaction leads to better verbal skills as well as nonverbal skills (Allen & Marshall, 2011; Laski et al., 1988). This form of treatment can be modified for ASD/ID children by training typically developing peers on how to interact with simpler verbalizations and allow greater response time for the child. By making these modifications, an ASD/ID child can see improvements in nonverbal and verbal skills and will likely have a greater self-concept because they are increasing their social group (Agaliotis & Kalyva, 2008).

Implications and Future Directions

The treatment of ASD in the context of ID has been the focus of many child and developmental disability researchers for several decades. Many of the interventions discussed throughout this chapter were developed for infants, toddlers, and school-aged children with comorbid ASD and ID and the science supporting these treatments show promising results. The current status of the field, however, requires greater focus on the aging population of individuals with ASD and ID. Very few interventions have been developed targeting adults or even adolescents and the transitions between various phases of life are an area that interventionists and support programs have failed to successfully address. While many have attempted to adapt programs to fit the needs of adolescents and adults (and some rather successfully) further research and resources should be targeting the appropriate care of this aging population.

Though the ASD and ID treatment and support community has come a long way from the days of mass institutionalization, integration into our communities continues to be a challenge. While acceptance by employers and community members has continued to grow, appropriate support programs and legislation continue to be grossly deficient. Further program development should continue to focus on the development of work training, supported independent living, and community integration programs that are individualized to the needs and abilities of the individual. Although many such programs exist, very few have the ability to be tailored to the individual and can place unrealistic expectations and goals on the client (e.g., teaching a nonverbal adult how to manage personal finances who has not mastered counting).

Finally, treatment services for those with ASD and ID have traditionally centered around major medical universities and hospitals in urban areas; however, a large portion of this population (and possibly even the majority) live in rural area. Access to appropriate services continues to be lacking including not only treatment and support services but even diagnostic services. Further research and resources should be focused on the delivery of evidence-based interventions and services to these rural populations and may include existing technologies such as telehealth, monitoring and support systems, and cell phone apps.

Since appropriate and timely intervention in the early years of the individual's life may contribute to a reduction in severity of ASD and ID symptoms, the need for effective, evidence-based treatments suitable for this population is paramount. Even though a majority of the evidence-based treatments discussed in this chapter have been designed for and studied on preschool and school-aged children, adolescent and adult clients would likely benefit from those programs as well. More research is, however, needed to validate the effectiveness of the discussed treatments for a variety of age groups and adaptations will likely be necessary dependent on the individual client.

References

Adcock, J., & Cuvo, A. J. (2009). Enhancing learning for children with autism spectrum disorders in regular education by instructional modifications. *Research in Autism Spectrum Disorders, 3*(2), 319–328. https://doi.org/10.1016/j.rasd.2008.07.004

Agaliotis, I., & Kalyva, E. (2008). Nonverbal social interaction skills of children with learning disabilities. *Research in Developmental Disabilities, 29*(1), 1–10. https://doi.org/10.1016/j.ridd.2006.09.002

Akamoglu, Y., & Meadan, H. (2018). Parent-implemented language and communication interventions for children with developmental delays and disabilities: A scoping review. *Review Journal of Autism and Developmental Disorders.*, 1–16. https://doi.org/10.1007/s40489-018-0140-x

Allen, J., & Marshall, C. R. (2011). Parent-child interaction therapy (PCIT) in school-aged children with specific language impairment. *International Journal of Language & Communication Disorders, 46*(4), 397. https://doi.org/10.3109/13682822.2010.517600

American Psychiatric Association. (2013). *Diagnostic and statistical manual of mental disorders* (5th ed.). Arlington, VA: American Psychiatric Publishing.

Athens, E. S., & Vollmer, T. R. (2010). An investigation of differential reinforcement of alternative behavior without extinction. *Journal of Applied Behavior Analysis, 43*, 569–589. https://doi.org/10.1901/jaba.2010.43-569

Autism and Developmental Disabilities Monitoring Network Surveillance Year 2014 Principal

Investigators. (2018). Prevalence of autism spectrum disorder among children aged 8 years—Autism and developmental disabilities monitoring network, 11 Sites, United States, 2014. *Morbidity and Mortality Weekly Report: Surveillance Summaries, 67*(6), 1–23. https://doi.org/10.15585/mmwr.ss6706a1

Baril, E. M., & Humphreys, B. P. (2017). An evaluation of the research evidence on the Early Start Denver Model. *Journal of Early Intervention, 39*(4), 321–338. https://doi.org/10.1177/1053815117722618

Beaumont, R., & Sofronoff, K. (2008). A multi-component social skills intervention for children with Asperger syndrome: The Junior Detective Training Program. *Journal of Child Psychology and Psychiatry, 49*(7), 743–753. https://doi.org/10.1111/j.1469-7610.2008.01920.x

Bellini, S., & Peters, J. K. (2008). Social skills training for youth with autism spectrum disorders. *Child and Adolescent Psychiatric Clinics of North America, 17*(4), 857–873. https://doi.org/10.1016/j.chc.2008.06.008

Bolte, S., & Poustka, F. (2002). The relation between general cognitive level and adaptive behavior domains in individuals with autism with and without co-morbid mental retardation. *Child Psychiatry and Human Development, 33*(2), 165–172. https://doi.org/10.1023/A:1020734325815

Bosseler, A., & Massaro, D. W. (2003). Development and evaluation of a computer-animated tutor for vocabulary and language learning in children with autism. *Journal of Autism and Developmental Disorders, 33*(6), 653–672.

Brock, M. E. (2013). *Cognitive behavioral intervention (CBI) fact sheet*. Chapel Hill: The University of North Carolina, Frank Porter Graham Child Development Institute, The National Professional Development Center on Autism Spectrum Disorders.

Butler, L. R., & Luiselli, J. K. (2007). Escape-maintained problem behavior in a child with autism antecedent functional analysis and intervention evaluation of noncontingent escape and instructional fading. *Journal of Positive Behavior Interventions, 9*(4), 195–202. https://doi.org/10.1177/10983007070090040201

Carr, E. G., & Durand, V. M. (1985). Reducing behavior problems through functional communication training. *Journal of Applied Behavior Analysis, 18*(2), 111–126. https://doi.org/10.1901/jaba.1985.18-111

Carr, E. G., & Kologinsky, E. (1983). Acquisition of sign language by autistic children. II: Spontaneity and generalization effects. *Journal of Applied Behavior Analysis, 16*, 297–314.

Carre, A. J. M., LeGrice, B., Blampied, N. M., & Walker, D. (2009). Picture Exchange Communication (PECS) Training for young children: Does training transfer at school and to home? *Behavior Change, 26*, 54–66.

Charlop, M. H., Schreibman, L., & Thibodeau, M. G. (1985). Increasing spontaneous verbal responding in autistic children using a time delay procedure. *Journal of Applied Behavior Analysis, 18*(2), 155–166.

Charlop, M. H., & Trasowech, J. E. (1991). Increasing autistic children's daily spontaneous speech. *Journal of Applied Behavior Analysis, 24*(4), 747–761.

Charlop, M. H., & Walsh, M. E. (1986). Increasing autistic children's spontaneous verbalizations of affection: An assessment of time delay and peer modeling procedures. *Journal of Applied Behavior Analysis, 19*(3), 307–314.

Choi, H., O'Reilly, M., Sigafoos, J., & Lancioni, G. (2010). Teaching requesting and rejecting sequences to four children with developmental disabilities using augmentative and alternative communication. *Research in Developmental Disabilities: A Multidisciplinary Journal, 31*(2), 560–567. https://doi.org/10.1016/j.rasd.2010.08.005

Cihak, D. F., Wright, R., & Ayres, K. M. (2010). Use of self-modeling static-picture prompts via a handheld computer to facilitate self-monitoring in the general education classroom. *Education and Training in Developmental Disabilities, 45*(1), 136–149.

Collet-Klingenberg, L. (2008). *Computer-aided instruction: Steps for implementation*. Madison, WI: The National Professional Development Center on ASD, Waisman Center, University of Wisconsin.

Collet-Klingenberg, L. (2009). *Overview of computer-aided instruction*. Madison, WI: The National Professional Development Center on Autism Spectrum Disorders, Waisman Center, The University of Wisconsin.

Collet-Klingenberg, L., Neitzel, J., & Laberge, J. (2012). Power-PALS (peers assisting, leading, supporting): Implementing a peer-mediated intervention in a rural middle school program. *Rural Special Education Quarterly, 31*(2), 3–11. https://doi.org/10.1177/875687051203100202

Conroy, M. A., Asmus, J. M., Sellers, J. A., & Ladwig, C. N. (2005). The use of an antecedent-based intervention to decrease stereotypic behavior in a general education classroom: A case study. *Focus on Autism and Other Developmental Disabilities, 20*(4), 223–230. https://doi.org/10.1177/10883576050200040401

Dave, D. M., & Fernandez, J. M. (2014). Rising autism prevalence: Real or displacing other mental disorders? Evidence from demand for auxiliary healthcare workers in California. *Economic Inquiry, 53*(1), 448–468. https://doi.org/10.1111/ecin.12137

Dawson, G., Jones, E. J. H., Merkle, K., Venema, K., Lowy, R., Faja, S., & Webb, S. J. (2012). Early behavioral intervention is associated with normalized brain activity in young children with autism. *Journal of the American Academy of Child and Adolescent Psychiatry, 51*(11), 1150–1159. https://doi.org/10.1016/j.jaac.2012.08.018

Dawson, G., Rogers, S., Munson, J., Smith, M., Winter, J., Greenson, J., & Varley, J. (2010). Randomized, controlled trial of an intervention for toddlers with autism: The Early Start Denver Model. *Pediatrics, 125*(1), 17–23. https://doi.org/10.1542/peds.2009-0958

Deb, S., & Prasad, K. (1994). The prevalence of autistic disorder among children with a learning disability. *The British Journal of Psychiatry, 165*(3), 395–399. https://doi.org/10.1192/bjp.165.3.395

Drahota, A., Wood, J. J., Sze, K. M., & Van Dyke, M. (2011). Effects of cognitive behavioral therapy on daily living skills in children with high-functioning autism and concurrent anxiety disorders. *Journal of Autism and Developmental Disorders, 41*(3), 257–265. https://doi.org/10.1007/s10803-010-1037-4

Dunlap, G., & Plienis, A. J. (1991). The influence of task size on the unsupervised task performance of students with developmental disabilities. *Education and Treatment of Children, 14*(2), 85–95.

Eapen, V., Črnčec, R., & Walter, A. (2013). Clinical outcomes of an early intervention program for preschool children with autism spectrum disorder in a community group setting. *BMC Pediatrics, 13*(1), 1–9. https://doi.org/10.1186/1471-2431-13-3

Estes, A., Munson, J., Rogers, S. J., Greenson, J., Winter, J., & Dawson, G. (2015). Long-term outcomes of early intervention in 6-year-old children with autism spectrum disorder. *Journal of the American Academy of Child & Adolescent Psychiatry*. https://doi.org/10.1016/j.jaac.2015.04.005. Advance online publication.

Faja, S., Aylward, E., Bernier, R., & Dawson, G. (2007). Becoming a face expert: A computerized face-training program for high-functioning individuals with autism spectrum disorders. *Developmental Neuropsychology, 33*(1), 1–24. https://doi.org/10.1080/87565640701729573

Faras, H., Al Ateeqi, N., & Tidmarsh, L. (2010). Autism spectrum disorders. *Annals of Saudi Medicine, 30*(4), 295–300. https://doi.org/10.4103/0256-4947.65261

Fettig, A. (2013). *Functional behavior assessment (FBA) fact sheet*. Chapel Hill: The University of North Carolina, Frank Porter Graham Child Development Institute, The National Professional Development Center on Autism Spectrum Disorders.

Fodstad, J. C., & Matson, J. L. (2008). A comparison of feeding and mealtime problems in adults with intellectual disabilities with and without autism. *Journal of Developmental and Physical Disabilities, 20*(6), 541–550. https://doi.org/10.1007/s10882-008-9116-6

Franzone, E. (2009). *Overview of naturalistic intervention*. Madison, WI: National Professional Development Center on Autism Spectrum Disorders, Waisman Center, University of Wisconsin.

Franzone, E., & Collet-Klingenberg, L. (2008). *Overview of speech generating devices for children and youth with autism spectrum disorders*. Madison, WI: The National Professional Development Center on Autism Spectrum Disorders, Waisman Center, University of Wisconsin.

Fulton, E., Eapen, V., Crnčec, R., Walter, A., & Rogers, S. (2014). Reducing maladaptive behaviors in preschool-aged children with autism spectrum disorder using the early start Denver model. *Frontiers in Pediatrics, 2*(40), 1–9. https://doi.org/10.3389/fped.2014.00040

Ganz, J. B., Davis, J. L., Lund, E. M., Goodwyn, F. D., & Simpson, R. L. (2012). Meta-analysis of PECS with individuals with ASD: Investigation of targeted versus non-targeted outcomes, participant characteristics, and implementation phase. *Research in Developmental Disabilities, 33*(2), 406–418. https://doi.org/10.1016/j.ridd.2011.09.023

Goldsmith, T. R., LeBlanc, L. A., & Sautter, R. A. (2007). Teaching intraverbal behavior to children with autism. *Research in Autism Spectrum Disorders, 1*(1), 1–13. https://doi.org/10.1016/j.rasd.2006.07.001

Gonzalez-Lopez, A., & Kamps, D. M. (1997). Social skills training to increase social interactions between children with autism and their typical peers. *Focus on Autism & Other Developmental Disabilities, 12*(1), 2.

Hamilton, B. L., & Snell, M. E. (1993). Using the milieu approach to increase spontaneous communication book use across environments by an adolescent with autism. *Augmentative and Alternative Communication, 9*, 259–272.

Hanley, G. P. (2012). Functional assessment of problem behavior: Dispelling myths, overcoming implementation obstacles, and developing new lore. *Behavior Analysis in Practice, 5*(1), 54–72. https://doi.org/10.1007/bf03391818

Hart, A., & Carr, S. (2016). *Antecedent based intervention [pamphlet]*. Richmond, VA: Autism Center for Excellence.

Hayes, G. R., Hirano, S., Marcu, G., Monibi, M., Nguyen, D. H., & Yeganyan, M. (2010). Interactive visual supports for children with autism. *Personal and Ubiquitous Computing, 14*(7), 663–680. https://doi.org/10.1007/s00779-010-0294-8

Heath, A. K., Ganz, J., Parker, R., Burke, M., & Ninci, J. (2015). A meta-analytic review of functional communication training across mode of communication, age, and disability. *Review Journal of Autism and Developmental Disorders, 2*, 155–166. https://doi.org/10.1007/s40489-014-0044-3

Hetzroni, O. E., & Shalem, U. (2005). From logos to orthographic symbols: A multilevel fading computer program for teaching nonverbal children with autism. *Focus on Autism and Other Developmental Disabilities, 20*(4), 201–212.

Hetzroni, O. E., & Tannous, J. (2004). Effects of a computer-based intervention program on the communicative functions of children with autism. *Journal of Autism and Developmental Disorders, 34*(2), 95–113.

Hua, F., Ya-yu, L., Shuling, T., & Cartledge, G. (2008). The effects of theory-of-mind and social skill training on the social competence of a sixth-grade student with autism. *Journal of Positive Behavior Interventions, 10*(4), 228.

IDEA. (2004) Individuals with Disabilities Education Act, 20 U.S.C. § 1400.

Ingersoll, B., & Gergans, S. (2007). The effect of a parent-implemented imitation intervention on spontaneous imitation skills in young children with autism. *Research in Developmental Disabilities, 28*(2), 163–175. https://doi.org/10.1016/j.ridd.2006.02.004

Ingersoll, B., Lewis, E., & Kroman, E. (2007). Teaching the imitation and spontaneous use of descriptive gestures in young children with autism using a naturalistic behavioral intervention. *Journal of Autism and Developmental Disorders, 37*, 1446–1456.

Ingvarsson, E. T., Kahng, S., & Hausman, N. L. (2008). Some effects of noncontingent positive reinforcement on multiply controlled problem behavior and compliance in a demand context. *Journal of Applied Behavior Analysis, 41*, 435–440. https://doi.org/10.1901/jaba.2008.41-435

Jackson, C. T., Fein, D., Wolf, J., Jones, G., Hauck, M., Waterhouse, L., & Feinstein, C. (2003). Responses and sustained interactions in children with mental retardation and autism. *Journal of Autism and Developmental Disorders, 33*(2), 115–121. https://doi.org/10.1023/A:1022927124025

Jahr, E. (2001). Teaching children with autism to answer novel wh-questions by utilizing a multiple exemplar strategy. *Research in Developmental Disabilities, 22*(5), 407–423. https://doi.org/10.1016/S0891-4222(01)00081-6

Jones, E. A., Feeley, K. M., & Takacs, J. (2007). Teaching spontaneous responses to young children with autism. *Journal of Applied Behavior Analysis, 40*(3), 565–570. https://doi.org/10.1901/jaba.2007.40-565

Katz, E., & Girolametto, L. (2013). Peer-mediated intervention for preschoolers with ASD implemented in early childhood education settings. *Topics in Early Childhood Special Education, 33*(3), 133–143. https://doi.org/10.1177/0271121413484972

Kern, L., Marder, T. J., Boyajian, A. E., Elliot, C. M., & McElhattan, D. (1997). Augmenting the independence of self-management procedures by teaching self-initiation across settings and activities. *School Psychology Quarterly, 12*(1), 23–32. https://doi.org/10.1037/h0088944

Kite, D. M., Gullifer, J., & Tyson, G. A. (2012). Views on the diagnostic labels of autism and Asperger's disorder and the proposed changes in the DSM. *Journal of Autism and Developmental Disorders, 43*(7), 1692–1700. https://doi.org/10.1007/s10803-012-1718-2

Koegel, L. K., Koegel, R. L., Hurley, C., & Frea, W. D. (1992). Improving social skills and disruptive behavior in children with autism through self-management. *Journal of Applied Behavior Analysis, 25*(2), 341–353. https://doi.org/10.1901/jaba.1992.25-341

Koegel, R. L., Camarata, S., Koegel, L. K., Ben-Tall, A., & Smith, A. E. (1998). Increasing speech intelligibility in children with autism. *Journal of Autism and Developmental Disorders, 28*(3), 241–251.

Koegel, R. L., & Schreibman, L. (1996). Fostering self-management: Parent-directed pivotal response training for children with autistic disorder. In E. D. Hibbs & P. S. Jenson (Eds.), *Psychosocial treatments for child and adolescent disorders* (pp. 525–552). Washington, DC: American Psychological Association.

Kokina, A., & Kern, L. (2010). Social Story™ interventions for students with autism spectrum disorders: A meta-analysis. *Journal of Autism and Developmental Disorders, 40*(7), 812–826. https://doi.org/10.1007/s10803-009-0931-0

Kurtz, P. F., Boelter, E. W., Jarmolowicz, D. P., Chin, M. D., & Hagopian, L. P. (2011). An analysis of functional communication training as an empirically supported treatment for problem behavior displayed by individuals with intellectual disabilities. *Research in Developmental Disabilities, 32*, 2935–2942. https://doi.org/10.1016/j.ridd.2011.05.009

La Malfa, G., Lassi, S., Bertelli, M., Salvini, R., & Placidi, G. F. (2004). Autism and intellectual disability: A study of prevalence on a sample of the Italian population. *Journal of Intellectual Disability Research, 48*(3), 262–267. https://doi.org/10.1111/j.1365-2788.2003.00567.x

Ladd, M. V., Luiselli, J. K., & Baker, L. (2009). Continuous access to competing stimulation as intervention for self-injurious skin picking in a child with autism. *Child & Family Behavior Therapy, 31*(1), 54–60. https://doi.org/10.1080/07317100802701400

Lang, R., Rispoli, M., Sigafoos, J., Lancioni, G., Andrews, A., & Ortega, L. (2011). Effects of language of instruction on response accuracy and challenging behavior in a child with autism. *Journal of Behavioral Education, 20*(4), 252–259. https://doi.org/10.1007/s10864-011-9130-0

Laski, K. E., Charlop, M. H., & Schreibman, L. (1988). Training parents to use the natural language paradigm to increase their autistic children's speech. *Journal of Applied Behavior Analysis, 21*(4), 391–400. https://doi.org/10.1901/jaba.1988.21-391

Leaf, J. A., Leaf, J. B., Milne, C., Townley-Cochran, D., Oppenheim-Leaf, M. L., Cihon, J. H., … Leaf, R. (2016). The effects of the cool versus not cool procedure to teach social game play to individuals diagnosed with autism spectrum disorder. *Behavior Analysis in Practice, 9*(1), 34–49. https://doi.org/10.1007/s40617-016-0112-5

Leaf, J. B., Leaf, R., Taubman, M., McEachin, J., & Delmolino, L. (2014). Comparison of flexible prompt fading to error correction for children with autism spectrum disorder. *Journal of Developmental and Physical Disabilities, 26*(2), 203–224. https://doi.org/10.1007/s10882-013-9354-0

MacNaul, H. L., & Neely, L. C. (2018). Systematic review of differential reinforcement of alternative behavior without extinction for individuals with autism. *Behavior Modification, 42*(3), 398–421. https://doi.org/10.1177/0145445517740321

Massaro, D. W., & Bosseler, A. (2006). Read my lips: The importance of the face in a computer-animated tutor for vocabulary learning by children with autism. *Autism, 10*(5), 495–510.

Matson, J. L., Matson, M. L., & Rivet, T. T. (2007). Social-skills treatments for children with autism spectrum disorders: An overview. *Behavior Modification, 31*(5), 682–707. https://doi.org/10.1177/0145445507301650

Matson, J. L., Mayville, E. A., Lott, J. D., Bielecki, J., & Logan, R. (2003). A comparison of social and adaptive functioning in persons with psychosis, autism, and severe or profound mental retardation. *Journal of Developmental and Physical Disabilities, 15*(1), 57–65. https://doi.org/10.1023/A:1021404304361

Matson, J. L., & Shoemaker, M. (2009). Intellectual disability and its relationship to autism spectrum disorders. *Research in Developmental Disabilities, 30*(6), 1107–1114. https://doi.org/10.1016/j.ridd.2009.06.003

McEachin, J. J., Smith, T., & Lovaas, I. O. (1993). Long-term outcome for children with autism who received early intensive behavioral treatment. *American Association on Mental Retardation, 97*(4), 359–372.

McGee, G. G., Almeida, M. C., Sulzer-Azaroff, B., & Feldman, R. S. (1992). Promoting reciprocal teaching via peer incidental teaching. *Journal of Applied Behavior Analysis, 25*, 117–126.

McGee, G. G., Krantz, P. J., Mason, D., & McClannahan, L. E. (1983). A modified incidental teaching procedure for autistic youth. *Journal of Applied Behavior Analysis, 16*, 329–338.

McGee, G. G., Krantz, P. J., & McClannahan, L. E. (1985). The facilitative effects of incidental teaching on preposition use by autistic children. *Journal of Applied Behavior Analysis, 18*, 17–31.

McHugh, L., Bobarnac, A., & Reed, P. (2011). Brief report: Teaching situation-based emotions to children with autistic spectrum disorder. *Journal of Autism and Developmental Disorders, 41*(10), 1423–1428. https://doi.org/10.1007/s10803-010-1152-2

McPartland, J. C., Reichow, B., & Volkmar, F. R. (2012). Sensitivity and specificity of proposed *DSM-5* diagnostic criteria for Autism Spectrum Disorder. *Journal of the American Academy of Child & Adolescent Psychiatry, 51*(4), 368–383. https://doi.org/10.1016/j.jaac.2012.01.007

Minshawi, N. F., Hurwitz, S., Morriss, D., & Mcdougle, C. J. (2015). Multidisciplinary assessment and treatment of self-injurious behavior in autism spectrum disorder and intellectual disability: Integration of psychological and biological theory and approach. *Journal of Autism and Developmental Disorders, 45*(6), 1541–1568. https://doi.org/10.1007/s10803-014-2307-3

Mintz, M. (2016). Evolution in the understanding of autism spectrum disorder: Historical perspective. *The Indian Journal of Pediatrics, 84*(1), 44–52. https://doi.org/10.1007/s12098-016-2080-8

Mohammadzaheri, F., Koegel, L. K., Rezaee, M., & Rafiee, S. M. (2014). A randomized clinical trial comparison between pivotal response treatment (PRT) and structured applied behavior analysis (ABA) intervention for children with autism. *Journal of Autism and Developmental Disorders, 44*(11), 2769–2777. https://doi.org/10.1007/s10803-014-2137-3

Moore, M., & Calvert, S. (2000). Brief report: Vocabulary acquisition for children with autism: Teacher or computer instruction. *Journal of Autism and Developmental Disorders, 30*(4), 359–362.

National Research Council. (2001). *Educating children with autism*. Committee on Educational Interventions for Children with Autism. In C. Lord & J. P. McGee (Eds.), *Division of behavioral and social sciences and education*. Washington, DC: National Academy Press. https://doi.org/10.17226/10017

Newman, B., & Eyck, P. T. (2005). Self-management of initiations by students diagnosed with autism. *Analysis of Verbal Behavior, 21*, 117–122.

Olive, M. L., de la Cruz, B., Davis, T. N., Chan, J. M., Lang, R. B., O'Reilly, M. F., & Dickson, S. M. (2007). The effects of enhanced milieu teaching and a voice output communication aid on the requesting of three children with autism. *Journal of Autism and Developmental Disorders, 37*, 1505–1513.

Olive, M. L., Lang, R. B., & Davis, T. N. (2008). An analysis of the effects of functional communication and a voice output communication aid for a child with autism spectrum disorder. *Research in Autism Spectrum Disorders, 2*(2), 223–236. https://doi.org/10.1016/j.rasd.2007.06.002

Parsons, C., & La Sorte, D. (1993). The effects of computers with synthesized speech and no speech on the spontaneous communication of children with autism. *Australian Journal of Human Communication Disorders, 21*, 12–31.

Pierce, K., & Schreibman, L. (1995). Increasing complex social behaviors in children with autism: Effects of peer-implemented pivotal response training. *Journal of Applied Behavior Analysis, 283*, 285–295.

Pinborough-Zimmerman, J., Satterfield, R., Miller, J., Bilder, D., Hossain, S., & McMahon, W. (2007). Communication disorders: Prevalence and comorbid intellectual disability, autism, and Emotional/Behavioral disorders. *American Journal of Speech-Language Pathology, 16*(4), 359–367. https://doi.org/10.1044/1058-0360(2007/039)

Płatos, M., & Wojaczek, K. (2018). Broadening the scope of peer-mediated intervention for individuals with autism spectrum disorders. *Journal of Autism and Developmental Disorders, 48*(3), 747–750. https://doi.org/10.1007/s10803-017-3429-1

Plavnick, J., Kaid, T., & MacFarland, M. (2015). Effects of a school-based social skills training program for adolescents with autism spectrum disorder and intellectual disability. *Journal of Autism & Developmental Disorders, 45*(9), 2674–2690. https://doi.org/10.1007/s10803-015-2434

Reinhartsen, D. B., Garfinkle, A. N., & Wolery, M. (2002). Engagement with toys in two-year-old children with autism: Teacher selection versus child choice. *Research and Practice for Persons with Severe Disabilities, 27*(3), 175–187. https://doi.org/10.2511/rpsd.27.3.175

Rogers, S. J., & Dawson, G. (2010). *Early Start Denver Model for young children with autism: Promoting language, learning, and engagement*. New York: Guilford Press.

Sam, A., & Team, A. F. I. R. M. (2015). *Social narratives*. Chapel Hill, NC: National Professional Development Center on Autism Spectrum Disorder, FPG Child Development Center, University of North Carolina. Retrieved from http://afirm.fpg.unc.edu/social-narratives

Sam, A., & AFIRM Team. (2016). *Picture Exchange Communication System*. Chapel Hill, NC: National Professional Development Center on Autism Spectrum Disorder, FPG Child Development Center, University of North Carolina. Retrieved from http://afirm.fpg.unc.edu/picture-exchange-communication-system

Schreibman, L., Dawson, G., Stahmer, A. C., Landa, R., Rogers, S. J., Mcgee, G. G., … Halladay, A. (2015). Naturalistic developmental behavioral interventions: Empirically validated treatments for autism spectrum disorder. *Journal of Autism and Developmental Disorders, 45*(8), 2411–2428. https://doi.org/10.1007/s10803-015-2407-8

Schreibman, L., Whalen, C., & Stahmer, A. C. (2000). The use of video priming to reduce disruptive transition behavior in children with autism. *Journal of Positive Behavior Interventions, 2*(1), 3–11. https://doi.org/10.1177/109830070000200102

Sigafoos, J., Green, V. A., Payne, D., O'Reilly, M. F., & Lancioni, G. E. (2009). A classroom-based antecedent intervention reduces obsessive-repetitive behavior in an adolescent with autism. *Clinical Case Studies, 8*(1), 3–13. https://doi.org/10.1177/1534650108327475

Silver, M., & Oakes, P. (2001). Evaluation of a new computer intervention to teach people with autism or Asperger syndrome to recognize and predict emotions in others. *Autism, 5*(3), 299–316.

Singh, N. N., Lancioni, G. E., Manikam, R., Winton, A. S., Singh, A. N., Singh, J., & Singh, A. D. (2011). A mindfulness-based strategy for self-management of aggressive behavior in adolescents with autism. *Research in Autism Spectrum Disorders, 5*(3), 1153–1158. https://doi.org/10.1016/j.rasd.2010.12.012

Slocum, S. K., & Vollmer, T. R. (2015). A comparison of positive and negative reinforcement for compliance to treat problem behavior maintained by escape. *Journal of Applied Behavior Analysis, 48*, 563–574. https://doi.org/10.1002/jaba.216

Smith, T. (2001). Discrete trial training in the treatment of autism. *Focus on Autism and Other Developmental Disabilities, 16*(2), 86–92. https://doi.org/10.1177/108835760101600204

Sofronoff, K., Attwood, T., & Hinton, S. (2005). A randomised controlled trial of a CBT intervention for anxiety in children with Asperger syndrome. *Journal of Child Psychology and Psychiatry, 46*(11), 1152–1160. https://doi.org/10.1111/j.1469-7610.2005.00411.x

Sofronoff, K., Attwood, T., Hinton, S., & Levin, I. (2007). A randomized controlled trial of a cognitive behavioural intervention for anger management in children diagnosed with Asperger syndrome. *Journal of Autism and Developmental Disorders, 37*(7), 1203–1214. https://doi.org/10.1007/s10803-006-0262-3

Stahmer, A. C. (1995). Teaching symbolic play skills to children with autism using Pivotal Response Training. *Journal of Autism and Developmental Disorders, 25*, 123–141.

Steensel, F. J., Bögels, S. M., & Perrin, S. (2011). Anxiety disorders in children and adolescents with autistic spectrum disorders: A meta-analysis. *Clinical Child and Family Psychology Review, 14*(3), 302–317. https://doi.org/10.1007/s10567-011-0097-0

Stichter, J. P., Randolph, J. K., Kay, D., & Gage, N. (2009). The use of structural analysis to develop antecedent-based interventions for students with autism. *Journal of Autism and Developmental Disorders, 39*(6), 883–896. https://doi.org/10.1007/s10803-009-0693-8

Stromer, R., Mackay, H. A., Howell, S. R., McVay, A. A., & Flusser, D. (1996). Teaching computer-based spelling to individuals with developmental and hearing disabilities: Transfer of stimulum control to writing tasks. *Journal of Applied Behavior Analysis, 29*(1), 25–42. https://doi.org/10.1901/jaba.1996.29-25

Summers, J., Tarbox, J., Findel-Pyles, R. S., Wilke, A. E., Bergstrom, R., & Williams, W. L. (2011). Teaching two household safety skills to children with autism. *Research in Autism Spectrum Disorders, 5*(1), 629–632. https://doi.org/10.1016/j.rasd.2010.07.008

Taheri, A., Perry, A., & Minnes, P. (2016). Examining the social participation of children and adolescents with intellectual disabilities and autism spectrum disorder in relation to peers. *Journal of Intellectual Disability Research, 60*(5), 435–443.

Tiger, J. H., Hanley, G. P., & Bruzek, J. (2008). Functional communication training: A review and practical guide. *Behavior Analysis in Practice, 1*, 16–23. https://doi.org/10.1007/BF03391716

Trembath, D., Mahler, N., & Hudry, K. (2016). Evidence from systematic review indicates that parents can learn to implement naturalistic interventions leading to improved language skills in their children with disabilities. *Evidence-Based Communication Assessment and Intervention, 10*(2), 101–107. https://doi.org/10.1080/17489539.2016.1231387

Tse, C. Y. A., Pang, C. L., & Lee, P. H. (2018). Choosing an appropriate physical exercise to reduce stereotypic behavior in children with autism spectrum disorders: A non-randomized crossover study. *Journal of Autism and Developmental Disorders, 48*(5), 1666–1672. https://doi.org/10.1007/s10803-017-3419-3

Tureck, K., Matson, J. L., Cervantes, P., & Konst, M. J. (2014). An examination of the relationship between autism spectrum disorder, intellectual functioning, and comorbid symptoms in children. *Research in Developmental Disabilities, 35*(7), 1766–1772. https://doi.org/10.1016/j.ridd.2014.02.013

Van Acker, R., & Grant, S. (1995). An effective computer-based requesting system for persons with Rett syndrome. *Journal of Childhood Communication Disorders, 16*, 31–38.

Vismara, L., McCormick, C., Young, G., Nadhan, A., & Monlux, K. (2013). Preliminary findings of a telehealth approach to parent training in autism. *Journal of Autism and Developmental Disorders, 43*(12), 2953–2969. https://doi.org/10.1007/s10803-013-1841-8

Vismara, L. A., Colombi, C., & Rogers, S. J. (2009). Can one hour per week of therapy lead to lasting changes in young children with autism? *Autism, 13*(1), 93–115. https://doi.org/10.1177/1362361307098516

Vismara, L. A., & Rogers, S. J. (2008). The early start Denver model: A case study of an innovative practice.

Journal of Early Intervention, 31(1), 91–108. https://doi.org/10.1177/1053815108325578

Vivanti, G., Dissanayake, C., Zierhut, C., Rogers, S. J., & Victorian ASELCC Team. (2013). Brief report: Predictors of outcomes in the early start Denver model delivered in a group setting. *Journal of Autism and Developmental Disorders, 43*(7), 1717–1724. https://doi.org/10.1007/s10803-012-1705-7

Vivanti, G., Paynter, J., Duncan, E., Fothergill, H., Dissanayake, C., Rogers, S. J., & the Victorian ASELCC Team. (2014). Effectiveness and feasibility of the early start Denver model implemented in a group-based community childcare setting. *Journal of Autism and Developmental Disorders, 44*(12), 3140–3153. https://doi.org/10.1007/s10803-014-2168-9

Waddington, H., Van der Meer, L., & Sigafoos, J. (2016). Effectiveness of the Early Start Denver model: A systematic review. *Review Journal of Autism and Developmental Disorders, 3*, 93–106. https://doi.org/10.1007/s40489-015-0068-3

Walker, G. (2008). Constant and progressive time delay procedures for teaching children with autism: A literature review. *Journal of Autism and Developmental Disorders, 38*(2), 261–275. https://doi.org/10.1007/s10803-007-0390-4

Walker, V. L., Lyon, K. J., Loman, S. L., & Sennott, S. (2018). A systematic review of Functional Communication Training (FCT) interventions involving augmentative and alternative communication in school settings. *Augmentative and Alternative Communication, 34*(2), 118–129. https://doi.org/10.1080/07434618.2018.1461240

Wallace, K. S., & Rogers, S. J. (2010). Intervening in infancy: Implications for autism spectrum disorders Katherine Wallace and Sally Rogers Intervening in infancy. *Journal of Child Psychology & Psychiatry, 51*(12), 1300–1320. https://doi.org/10.1111/j.1469-7610.2010.02308.x

Wetherby, A. M., Guthrie, W., Woods, J., Schatschneider, C., Holland, R. D., Morgan, L., & Lord, C. (2014). Parent-implemented social intervention for toddlers with autism: An RCT. *Pediatrics, 134*(6), 1084–1093. https://doi.org/10.1542/peds.2014-0757

Whalen, C., Moss, D., Ilan, A. B., Vaupel, M., Fielding, P., Macdonald, K., … Symon, J. (2010). Efficacy of TeachTown: Basics computer-assisted intervention for the intensive comprehensive autism program in Los Angeles unified school district. *Autism, 14*(3), 179–197. https://doi.org/10.1177/1362361310363282

Wilson, K. P. (2013). Teaching social-communication skills to preschoolers with autism: Efficacy of video versus in vivo modeling in the classroom. *Journal of Autism and Developmental Disorders, 43*(8), 1819–1831. https://doi.org/10.1007/s10803-012-1731-5

Wolff, J. J., Hupp, S. C., & Symons, F. J. (2013). Brief report: Avoidance extinction as treatment for compulsive and ritual behavior in autism. *Journal of Autism and Developmental Disorders, 43*(7), 1741–1746. https://doi.org/10.1007/s10803-012-1721-7

Wong, C., Odom, S. L., Hume, K. A., Cox, A. W., Fettig, A., Kucharczyk, S., … Schultz, T. R. (2015). Evidence-based practices for children, youth, and young adults with autism spectrum disorder: A comprehensive review. *Journal of Autism and Developmental Disorders, 45*(7), 1951–1966. https://doi.org/10.1007/s10803-014-2351-z

Wynn, J. W., & Smith, T. (2003). Generalization between receptive and expressive language in young children with autism. *Behavioral Interventions, 18*(4), 245–266. https://doi.org/10.1002/bin.142

Yoder, P., & Stone, W. L. (2006). A randomized comparison of the effect of two prelinguistic communication interventions on the acquisition of spoken communication in preschoolers with ASD. *Journal of Speech, Language, and Hearing Research, 49*, 698–711.

Zaghlawan, H. Y., & Ostrosky, M. M. (2016). A parent-implemented intervention to improve imitation skills by children with autism: A pilot study. *Early Childhood Education Journal, 44*(6), 671–680. https://doi.org/10.1007/s10643-015-0753-y

48

ADHD in Individuals with Intellectual Disability

Laura Flanigan, Emma A. Climie, Christina Gray, and Cristina Fernández Conde

Individuals with intellectual disabilities (ID) may be impacted by any number of other comorbid conditions. Indeed, those with ID may be at greater risk for co-occurring conditions such as autism spectrum disorder, learning disorders, mental health concerns, and/or other physical impairments. More recently, researchers and practitioners have become aware that individuals with ID can also be affected by attentional disorders, such as Attention-Deficit/Hyperactivity Disorder (ADHD).

This chapter will provide an overview of ADHD and ID, with a specific emphasis on both the overlap and the distinctions between the two disorders. We will begin with brief descriptions of ADHD and ID diagnoses and continue with a summary of current literature surrounding the strengths and challenges of those with a combined ID/ADHD diagnosis. The majority of the chapter will focus on reviewing and describing evidence-based recommendations and suggestions for treatment and support. Specifically, we will highlight supports in a number of domains, including social interaction, behavioral interventions, academic recommendations, adaptive challenges, and in other more general supports for adults. We will conclude by highlighting some areas of continual need for those with ID/ADHD as they transition into adulthood.

Attention-Deficit/Hyperactivity Disorder

ADHD is a neurodevelopmental disorder that is characterized by persistent symptoms of hyperactivity, impulsivity, and/or inattention that affect an individual's ability to adapt to his or her environment (American Psychiatric Association, 2013). Attention encompasses a series of mental operations that allow individuals to focus, encode, disengage, and shift their focus from diverse stimuli in their environment (Seidman, 2006). Inattention, or the lack of attention, can be observed behaviorally by disengagement with presented tasks, issues with persistence, and challenges organizing or understanding information (APA, 2013). Inattentive children may be prone to careless mistakes, losing their belongings, and missing relevant information when they are involved in play and schoolwork (Friedberg & McClure, 2015). Those who present with the hyperactive component of the disorder display inappropriate motor activity, which can present as fidgeting, garrulousness, and continual movement (APA, 2013). Impulsivity becomes evident in actions undertaken without consideration of

L. Flanigan · E. A. Climie (✉) · C. Gray · C. F. Conde
School & Applied Child Psychology, Werklund
School of Education, University of Calgary,
Calgary, AB, Canada
e-mail: eaclimie@ucalgary.ca

J. L. Matson (ed.), *Handbook of Intellectual Disabilities*, Autism and Child Psychopathology
Series, https://doi.org/10.1007/978-3-030-20843-1_48

the environment and consequences that could result in harm (APA, 2013).

According to the *Diagnostic and Statistical Manual of Mental Disorders, Fifth Edition* (DSM-5), three main subtypes of ADHD are identified: predominantly inattentive presentation, predominantly hyperactive/impulsive presentation, and combined presentation (APA, 2013). Those with the predominantly inattentive presentation must demonstrate six or more inattentive behaviors (five or more if the individual is over the age of 17 years or an adult), such as lack of attention to detail, lack of sustained attention, failure to complete tasks or follow through on instructions, loses things (e.g., keys, books, pencils), or is frequently forgetful. Those with the predominantly hyperactive/impulsive presentation must demonstrate six or more hyperactive/impulsive symptoms (five symptoms for adolescents/adults), such as fidgeting, tapping, playing loudly, difficulty waiting, talking excessively, interrupting others, or blurting out answers. If an individual meets the diagnostic criteria for both inattentive and hyperactive/impulsive presentations independently, then a combined presentation diagnosis is warranted.

Identification of an ADHD subtype requires thorough observation or report of the core symptoms of inattention and hyperactivity/impulsivity to verify they are negatively impacting the individuals functioning. According to the DSM-5, symptoms must be present before age 12 years and can persist throughout the life span (APA, 2013). As well, it is noted that the challenges derived from the observed symptoms must interfere with the person's functioning in at least two settings (e.g., home and work or home and school) to ensure that there is continuity in symptom presentations and that the behaviors are not simply environmentally controlled (APA, 2013). Finally, the severity of symptomatology (e.g., mild, moderate, or severe) can also be specified (APA, 2013).

Historical Background

Symptoms of what is now referred to as ADHD were first described in 1775 in a chapter by physician Melchior Adam Weikard (Barkley & Peters, 2012). Decades later, Alexander Crichton described a child who could not control himself (Taylor, 2011). Although single cases were described in the literature, it was not until the beginning of the nineteenth century that Benjamin Rush focused on the description of impulsive behavior and lack of control in children (Nigg & Barkley, 2014; Taylor, 2011). Years later, Frederick Still provided significant description of behavior that can be linked to ADHD by describing children with difficulties in inhibitory behavioral control and attention (Taylor, 2011). He was also one of the first to identify higher prevalence in males than females (Barkley, 2014). Tredgold provided further insight on this area by highlighting that through medication and environmental changes, behavioral improvements could be observed (Barkley, 2014).

Many children that survived the encephalitis epidemic of 1917 presented with disruptive behaviors and symptoms similar to those described for ADHD. Thus, doctors started describing these children as having "postencephalitic behavior disorder" (Barkley, 2014). At this time, heightened interest existed in understanding the link between disruptive behaviors and brain dysfunction. By focusing on brain damage in the early twentieth century, ADHD became known as Minimal Brain Dysfunction (MBD) as part of a conceptualization that considered the possibility of the existence of a unitary syndrome of brain damage in young populations (Barkley, 2014; Nigg & Barkley, 2014).

In 1968, the *Diagnostic and Statistical Manual of Mental Disorders, Second Edition* (DSM-II) included a disorder called hyperkinetic reaction of childhood. In the late 1960s, questions began around understanding hyperactivity through brain damage, ultimately resulting in a shift towards considering behavioral symptoms to describe hyperactivity (Barkley, 2014). Furthermore, evidence was provided to suggest that there was not always neurological evidence to suggest the diagnosis of hyperkinesis (Barkley, 2014). It was postulated that hyperactivity could be linked to intellectual disabilities, brain damage, and psychotic disorders and other serious mental illnesses. At this point, it was believed that hyperactivity was outgrown by adolescence (Barkley, 2014).

Wender, at the beginning of the 1970s, brought to attention that individuals with hyperactivity also presented with a short attention span, impulsivity, difficulties in concentration, and challenges with learning and impulse control. However, seminal work by Virginia Douglas suggested that difficulties with attention and impulse control represented considerable challenge for hyperactive children above and beyond hyperactivity itself (Barkley, 2014). Through follow up studies, extreme variability in performance in this population was demonstrated and it was found that challenges with sustained attention and impulse control persisted into adulthood (Barkley, 2014). With the changes in research trends, Hyperkinetic Reaction of Childhood was renamed Attention Deficit Disorder (ADD) in the *Diagnostic and Statistical Manual of Mental Disorders, Third Edition* (DSM-III; Nigg & Barkley, 2014). The change in terminology showed a shift in the conceptualization of the disorder by focusing on its psychological components rather than unknown neurological causes (Taylor, 2011). Further research on ADD started showing the impact that culture, environment, and a multidisciplinary approach to treatment (e.g., use of behavioral management interventions, medication, and parent training to treat ADD) had on the affected population (Barkley, 2014).

In the revised version of the DSM-III (DSM-III-R), factor analysis indicated the presence of three symptom groups: inattention, hyperactivity, and impulsivity (Barkley, 2014; Nigg & Barkley, 2014). Thus, the hyperactive impulsive components of the disorder were highlighted by renaming the condition attention-deficit/hyperactivity disorder (Nigg & Barkley, 2014). The diagnosis further specified whether hyperactivity was present or not (e.g., ADHD with or without hyperactivity; Barkley, 2014).

Significant progress in neuroimaging and genetic research led to the fourth edition of the DSM (DSM-IV; APA, 2013) being the strongest empirical set of criteria of ADHD (Barkley, 2014). In the most current DSM edition (DSM-5; Nigg & Barkley, 2014), changes were made to include environmental considerations and symptom counts that accounted for the challenges associated with a late adolescence or adult diagnosis (Barkley, 2014). In the twenty-first century, the study of ADHD focuses not only on attention and hyperactivity-impulsivity but also in understanding comorbidity (Barkley, 2014) and executive functions, which are relevant cognitive abilities thought to underlie ADHD (Nigg & Barkley, 2014).

Etiology and Epidemiology

Etiology. There are several etiological explanations for ADHD. The disorder is highly heritable (APA, 2013), suggesting that those with relatives with ADHD are more likely to develop the disorder. Neurologically, the frontostriatal model of ADHD has focused on understanding ADHD as a disorder that affects the prefrontal cortex, a part of the brain responsible for executive control and behavioral regulation (Seidman, 2006). This theory gathered substantial support when some studies reported a smaller prefrontal cortex in children with ADHD, as well as a slight asymmetry between the left and right prefrontal cortex (Krain & Castellanos, 2006). Research has also focused on structures that have been linked to attention (cerebellum), motor control (putamen), coordination of executive functioning (basal ganglia), and white and gray matter development (Krain & Castellanos, 2006).

Currently, although there is no core consensus on the definition of executive functioning (EF), it is generally understood as an overarching control mechanism of high-order activities that encompass several subprocesses such as working memory, inhibitory control, selective attention, planning, and problem-solving (Castellanos & Tannock, 2002; Lambek et al., 2011; Miyake et al., 2000). For individuals to regulate their behaviors and organize EF, inhibitory response is especially relevant (Castellanos & Tannock, 2002). Based on this idea, Barkley hypothesized executive dysfunction as being the primary deficit in ADHD (Castellanos & Tannock, 2002). In this theory, Barkley theorized that self-regulation is dependent on the inhibition of behavioral responses through four EF components: verbal and nonverbal working memory, self-regulation of affect, motivation and arousal, and effective analysis and synthesis of behavior (Nigg & Barkley, 2014). Disinhibition and impairment in the above-mentioned EF areas affect motor responses and adaptive functioning negatively by impairing self-regulation (Nigg & Barkley, 2014).

Prevalence. Due to its neurodevelopmental nature, ADHD affects individuals over the course of their life (Abecassis, Isquith, & Roth, 2017). Estimates suggest that ADHD affects approximately 5% of school-aged children (APA, 2013; Steinberg & Drabick, 2015), making it one of the most common mental disorders in youth (Takeda, Ambrosini, deBerardinis, & Elia, 2012). The DSM-5 estimates a prevalence of 2.5% in adulthood (APA, 2013). However, prevalence rates vary in different regions of the world depending on age, with rates varying from 4 to 21% in young adults (Abecassis et al., 2017). With regards to gender, prevalence in childhood is higher in males than females (approximately 3:1 ratio), but this proportion evens out during adulthood (Nigg & Barkley, 2014).

Diagnostic methods. The primary form of diagnosis of this disorder relies on clinical judgements that are based on observations, behavioral rating scales completed by the individual and those around him/her (e.g., parents or teachers), as well as standardized measures (Climie, Mah, & Chase, 2017; Mueller, Hong, Shepard, & Moore, 2017). Based on the findings, a clinician or pediatrician may specify a predominantly inattentive, hyperactive/impulsive, or combined presentation of the disorder. To help increase diagnostic accuracy regarding ADHD, current research has focused on increasing understanding of anatomical (Krain & Castellanos, 2006) and neural circuits that could support a more specific classification of the disorder (Mueller et al., 2017) in addition to furthering the understanding of the behavioral connections to ADHD.

Developmental Presentation

It is important to study the presentation of ADHD over the life span to fully understand its characteristics and impact in different life stages, including early childhood, adolescence, and adulthood. Although the DSM-5 does not prohibit the diagnosis of ADHD prior to age 4 years, the validity of a diagnosis at such an early age is contentious (Nigg & Barkley, 2014). Most individuals with ADHD are identified at eight to 12 years of age (Nigg & Barkley, 2014). Children diagnosed with ADHD have difficulties with sustained attention (Mueller et al., 2017), working memory, and processing speed (Koziol & Budding, 2012). In a school setting, numerous studies show that students with ADHD tend to be disruptive, have difficulty staying on task, move around constantly, interrupt their teacher and peers, and are prone to leaving their seat without permission (Gaastra, Groen, Tucha, & Tucha, 2016). Furthermore, childhood ADHD can result in familial stress, and parents of children with ADHD are prone to feel less competent in their roles (Barkley et al., 2000). Thus, they are often faced with social, cognitive, familial, and occupational difficulties throughout their lives (Harvey, Breaux, & Lugo-Candelas, 2016; Nigg & Barkley, 2014).

Elevated levels of inattention and/or hyperactivity and impulsivity frequently persist into adolescence (Sibley, Kuriyan, Evans, Waxmonsky, & Smith, 2014). Challenges in social and family interactions are still present in this stage (Sibley et al., 2014). Adolescents with ADHD are at greater risk of substance abuse (Charach, Yeung, Climans, & Lillie, 2011) and risky driving practices (Thompson, Molina, Pelham, & Gnagy, 2007). For some adolescents, treatment adherence to medication is challenging and behavioral training and therapy may not be as effective as in childhood, due to adolescent or parental disengagement (Sibley et al., 2014).

Young adults with ADHD often experience numerous challenges as a result of reduced supervision and a cognitively demanding environment. Executive functioning challenges affect their academics and they may engage in risky sexual behavior (Abecassis et al., 2017). Research has shown that adults with this diagnosis tend to have job-related difficulties and family and peer conflict are still present (Magnin & Maurs, 2017). It is important to highlight that symptomatology can improve through the implementation of an appropriate treatment plan.

Intellectual Disability

The term *intellectual disability (ID)* refers to a neurodevelopmental disorder characterized by deficits in general mental abilities that result in impairments in adaptive functioning (APA, 2013). For example, an individual with intellectual disability may present with deficits in mental reasoning, problem-solving, abstract thinking, planning, and learning from academics and/or experience (APA, 2013). These general mental deficits impact daily adaptive functioning to the extent that the individual is unable to meet standards of independence and responsibility, including in the areas of communication, academic or occupational functioning, and independent living at home or in their community. Onset of ID must occur during the developmental period in an individual's life. Previously, ID has been referred to using other terms, such as *mental retardation* and *intellectual developmental disorder* (APA, 2013). At present, the term ID is most commonly used by medical professionals and across public professions (APA, 2013; World Health Organization, 2009).

Historical Background

There have been numerous changes in the conceptualization and terminology relating to ID. Historically, the term *mental retardation* (MR) was used to refer to a cluster of disorders characterized by low intelligence and associated limitations in adaptive behavior function (Carulla et al., 2011). Initially, the World Health Organization (WHO) revised the *International Classification of Diseases and Related Health Problems, Eleventh Revision* (ICD-11) to reflect updated clinical research, frameworks, and further advance a unified global classification system relating to the previously termed MR syndromes (World Health Organization, 2009). The ICD-10 conceptualization of MR was replaced with the concept of *intellectual developmental disorders* (IDD) in ICD-11 (Carulla et al., 2011). This shift away from MR towards ID has occurred not just with the ICD-11 but also in policy and legislation in many developed countries (Carulla et al., 2011).

The DSM-5 classifies ID under the neurodevelopmental disorders family (APA, 2013). Previous editions of the DSM included MR, which was later changed to align with ICD and other organizations shifting to the label ID (Mahour & Panday, 2015). The current DSM-5 criteria for ID now encompasses a focus on impact on the individual's cognitive, social, and practical adaptive functioning as well as a decreased reliance on low intelligence quotient (IQ) scores (i.e., standard score ≤ 70 on a standardized intelligence test) to rule-in a diagnosis (Mahour & Panday, 2015). These changes in the diagnostic criteria of ID in the DSM place greater emphasis on adaptive reasoning in the individual's academic, social, and practical settings rather than focusing primarily on the individual's IQ score (Mahour & Panday, 2015). The historical views of ID have progressed to allow for a less stigmatizing terminology and view towards the disorder. These changes in conceptualization of ID have been adopted across dominant tools used to guide clinicians with diagnostic processes as well as shift ID associated administrative and legislative policies.

Etiology and Epidemiology

Etiology. The causes of ID are broad and variable; however, experts generally agree that there are six categories that cover the majority of ID cases: genetics, pre-, peri-, and post-natal difficulties, illness, and environmental factors

(Mouzakitis, 2012). Approximately 5% of ID cases are caused by genetic abnormalities. Other causes include pre-, peri-, and post-natal difficulties, including improper development or injury in utero, lack of oxygen during birth, and childhood diseases such as chicken pox or measles (Mouzakitis, 2012). General illness or health concerns, such as meningitis or malnutrition, can cause IDs. Lastly, IDs can be caused by environmental factors, such as lack of stimulation caused by neglect (Mouzakitis, 2012). It is important to determine the cause of the ID, as the root cause may result in additional difficulties that also need treatment (Mouzakitis, 2012).

Prevalence. ID has a general population prevalence of 1% in high-income countries, with prevalence rates varying by age (APA, 2013; Maulik, Mascarenhas, Mathers, Dua, & Saxena, 2011). In low income countries, the prevalence is around 2% (Durkin, 2002). The prevalence for severe ID is approximately 6 per 1000 individuals (APA, 2013). The highest rates of ID have been found in lower to middle-income countries, which presents challenges in terms of resource limitations (Maulik et al., 2011). Among adults with ID, there is a widely varying prevalence of comorbid psychiatric illness that affects between 10% and 39% of individuals, depending on the diagnostic criteria used (Deb, Thomas, & Bright, 2001).

Diagnostic methods. ID is typically identified through the use of standardized intelligence measures, such as the Wechsler Adult Intelligence Scale (WAIS; Corporation, 2008) or the Stanford-Binet (Roid, 2003), as well as an assessment of adaptive skills, such as the Vineland Adaptive Behavior Scale (Sparrow, Cicchetti, & Balla, 2005). Previously, under the guidelines of the DSM-IV, ID was identified solely through IQ determined by a cognitive assessment (APA, 2013). An individual could be identified as intellectually disabled if they achieved an IQ two standard deviations below the population mean (APA, 2013). For example, if the WAIS has a population mean of 100 and a standard deviation of 15, an individual would be considered intellectually disabled if they achieved an IQ of 70 or below on this scale (APA, 2013). It is now recognized, however, that adaptive skills are just as, if not more, important than IQ in identifying IDs. The DSM-5 (APA, 2013) assesses the severity of an ID (mild, moderate, severe, profound) based on an individual's ability to complete age-appropriate skills in the conceptual (e.g., literacy), social (e.g., developing appropriate relationships), and practical (basic hygiene) domains.

Developmental Presentation

ID is a disorder that continues throughout the life span of the affected person (APA, 2013). Individuals with ID follow similar sequences of cognitive development as typically developing individuals with respect to their progression of cognitive problem-solving, reasoning, and understanding abilities. However, the progression of these stages tends to stop earlier than those without ID, prior to completion of progression in conceptual development (Harris, 2006). An individual who is appropriately diagnosed with ID will continue to be intellectually disabled throughout her or his life span, experiencing a failure of typical cognitive progression (Harris, 2006). Early cognitive development tends to be more universal and neurobiologically based, while later, more complex cognitive development may vary with social and cultural experiences for the individual with ID (Harris, 2006). Understanding the contributory conditions (e.g., genetic/familial causes, brain damage) that result in ID may be useful with determining appropriate personal supports and focused education to support the individual's needs through adolescence, adulthood, and elderly years.

Contrary to misconceptions surrounding ID, many individuals with ID live to old age, continue to work, and in many instances can live independently and make contributions in their communities (Harris, 2006). Some individuals with ID may experience premature aging, shortened life expectancy, and poor health, particularly those with severe ID and multiple disabilities (Harris, 2006). Individuals with ID tend to have a lower capacity for self-determination, independent functioning, and limitations with work and social environments, adaptations allow them to meet their potential as they age (Harris, 2006).

Co-occurring ID and ADHD

Individuals with ID are at significant risk of developing comorbid psychiatric conditions. The most common comorbid mental disorder experienced by those with ID is ADHD (Baker, Neece, Fenning, Crnic, & Blancher, 2010). However, it was only recently that it became acceptable to diagnose these two disorders simultaneously. Historically, it was believed that an individual with ID could not also have ADHD, as any inattentive, hyperactive, and/or impulsive symptoms were largely the result of the more global mental impairment associated with ID (Ahuja, Martin, Langley, & Thapar, 2013). Additionally, and for similar reasons, the majority of research on ADHD has excluded participants with lower cognitive functioning (Lindsey, 2002). This phenomenon, known as "diagnostic overshadowing," where symptoms of one disorder are explained away by the presence of another (Fuller & Sabatino, 1998), has led not only to considerable under-diagnosis of ID with comorbid ADHD, but also a dearth of research examining this population (Ahuja et al., 2013). The following section will outline what is known about the etiology/epidemiology, presentation, and primary concerns among individuals with co-occurring ID and ADHD.

Etiology and Epidemiology

Etiology. Individually, ADHD and ID both have strong genetic components (APA, 2013). No studies have examined the shared genetic variance among those with ADHD and ID; however, Aureli et al. (2010) found that specific polymorphisms of the brain-derived neurotrophic factor (BDNF, polymorphisms Val66Met and 270 C/T) are significantly associated with both ADHD and ID, and are overrepresented in these populations compared to controls. This finding suggests that these BDNF polymorphisms could play a role in the comorbidity between ADHD and ID. Kuntsi et al. (2004) found that 100% of the variance between ADHD diagnosis and IQ was the result of shared genetics.

Epidemiology. As mentioned, the most common psychiatric condition associated with ID is ADHD (Baker et al., 2010). Depending on methodological variables, ADHD is associated with ID in 8–39% of cases (Baker et al., 2010; Dekker & Koot, 2003), notably higher than that expected in individuals without ID (approximately 5–7%; APA, 2013). Voigt, Barbaresi, Colligan, Weaver, and Katusic (2006) found an odds ratio of co-occurring ADHD with versus without ID to be 6.3, suggesting that those with ID are at a higher risk of also having ADHD. Both ADHD and ID, individually, occur much more frequently in males than females (APA, 2013); however, it is not known how this compares to populations with co-occurring ADHD and ID, although it is likely that ID/ADHD is more prevalent in males.

Presentation

To be diagnosed with ADHD in addition to ID, the individual must display symptoms of inattention, hyperactivity, and/or impulsivity over and above what would be expected from someone with ID alone (APA, 2013). Additionally, as per the diagnostic criteria for ADHD, the individual must display these symptoms in multiple settings, have displayed these symptoms since before age 12, and experience significant impairment in multiple domains of functioning as a result of the symptoms of ADHD (APA, 2013). Individuals with ID who meet criteria for ADHD may represent a distinct group in terms of their presentation and associated comorbid conditions.

Symptomology. Ahuja et al. (2013) found that children with both ADHD and ID had significantly lower IQs than those with ID-only. Interestingly, research suggests that there are no differences in the total number of ADHD symptoms displayed by those with ADHD and ID

(Ahuja et al., 2013; Johnson, Lubetsky, & Sacco, 1995), or in the presentations of ADHD (e.g., predominantly inattentive or hyperactive-impulsive; Baker et al., 2010) compared to ADHD-only. However, Neece, Baker, Blacher, and Crnic (2011) found that ADHD diagnoses appeared earlier and remained more stable among children with ID than without ID. Interestingly, individuals with ADHD and ID may be less responsive to medication and experience more side effects than those with only ADHD (Hässler & Thome, 2012).

Additional comorbid conditions. Ahuja et al. (2013) found that youth with both ADHD and ID, compared to youth with only ADHD and only ID, had higher symptoms and diagnoses of Oppositional Defiant Disorder (ODD) and Conduct Disorder (CD), potentially due to the increased level of impulsivity and lower social skills among those with ADHD/ID. Children with ADHD and ID were similar in their symptoms and diagnoses of depression and anxiety, compared to those with ADHD-only and ID-only. However, Aman, Armstrong, Buican, and Sillick (2002) found that adolescents with ADHD and ID had increased rates of separation anxiety disorder than those with ADHD only.

Primary Concerns

It is recognized that individuals with both ID and ADHD experience a "double deficit," whereby they experience significantly more impairment in functioning than either ADHD or ID alone (Carmeli, Klein, & Sohn, 2007). Specifically, Pearson et al. (2000) found that adolescents with ID and ADHD experienced significantly more depression, family conflict, defiance, anxiety, social skill deficits, and academic difficulties than adolescents with ID without ADHD. Carmeli et al. (2007) compared males with ID, ADHD, and co-occuring ID and ADHD and found that those with co-occurring ID and ADHD had significantly higher impairment in social skills than those with ID or ADHD only. Youth with ID and ADHD tend to have greater rates of behavioral problems than those with ADHD or ID alone (Aman et al., 2002; Carmeli et al., 2007). Additionally, hyperactivity among adolescents with ID is associated with bullying and violent behavior (Reiter, Bryen, & Shachar, 2007). Interestingly, Di Nuovo and Buono (2007) described that the co-occurrence of ID with ADHD has less of an impact on motor skills, verbal communication, and visual spatial skills than it does on areas that require judgment, reasoning, and memory. They did not find any differences in adaptive functioning between those with both ID and ADHD and ID only.

Treatment of Individuals with ID/ ADHD

This section outlines a number of areas in which those with ID/ADHD may benefit from treatment or support. Although the following intervention suggestions are not exhaustive, they may provide practitioners and families with some guidance as to how to support those with co-occurring ID/ADHD. This section specifically outlines evidence-based interventions related to four key domains: social, behavioral, academic, and adaptive support. Additionally, we have included a final section that notes a number of other interventions that may be beneficial specifically for adults with ID/ADHD.

Social Interventions

The development of appropriate peer interactions is one area in which those with ID or ADHD may have difficulties. For example, those with ID often struggle to engage conversationally with their peers, as their ability to keep up with or discuss age-appropriate topics may be limited by their cognitive skills. For those with ADHD, the inattentive and impulsive symptoms associated with the disorder may frequently impact a child's ability to follow a conversation or provide meaningful input at appropriate times (i.e., without blurting out or interrupting others). Together, when individuals are impacted by the challenges associated with both ID and ADHD, the social difficulties may multiply. Consequently, these individuals may not only struggle to find areas of common ground with peers, but may also have difficulties with the social norms related to conversational turn taking.

Broadly speaking, having close relationships with others and meaningful social interaction is linked to emotional well-being (Emerson & McVilly, 2004). Furthermore, within the ID population specifically, peer friendships have been found to be related to a number of additional beneficial outcomes, including increased adaptive skills and lower levels of severe behaviors (Kozma, Mansell, & Beadle-Brown, 2009). Those with ADHD are also known to have social challenges, with making and maintaining appropriate peers relationships among the primary concerns noted by parents and teachers (e.g., Hoza, 2007). Given the necessary link between social skills and peer relationships, those with ID/ADHD may benefit from additional social supports that may allow them to build skills and, more importantly, apply these skills consistently, effectively, and appropriately.

Building appropriate peer relations. One area in which children with ID/ADHD may benefit from additional supports is in the direct instruction of social skills, often achieved through social skill training program. Social skills training programs are often the primary manner in which individuals are taught skills that they may otherwise be lacking. However, as Gresham, Sugai, and Horner (2001) note, it is important to understand that those with greater levels of disability also require more frequent and intense instruction. A one hour per week social skills training group is unlikely to provide any significant change and may not show any sustained long-term benefit. Instead, more intensive or targeting intervention may produce more effective results, allowing those with disabilities the opportunity to build their skills more readily.

As well, those with ID/ADHD may benefit from additional support while building their social skills, rather than simply relying on group training. For example, including parents in the social skills training process may allow parents to understand the skills their children are being taught and help them work on those skills outside of the training sessions. As well, the inclusion of parent–child homework (e.g., social tasks that the parent helps the child complete, such as inviting another child to a play date) may help to generalize some of these skills to new or novel situations. This parent-inclusion model has been shown to be beneficial for those individuals on the autism spectrum, such as through the UCLA PEERS program (Laugeson, Frankel, Gantman, Dillon, & Mogil, 2012; Laugeson, Frankel, Mogil, & Dillon, 2009) and for those with ADHD (Frankel, Myatt, Cantwell, & Feinberg, 1997) individually. Tenets of these programs may also be particularly applicable to those with ID/ADHD, as the increased support provided by these models may create greater opportunities for success.

Understanding social conventions. Social conventions or social cooperation skills incorporate the ability to understand and abide by social rules, adhere to social structure, and attune to social expectations (Merrell & Wolfe, 1998). These skills may include things like sharing, turn taking, addressing adults, making polite requests (e.g., the use of “please” and “thank you”), appropriate greetings (e.g., with a peer or adult), or joining an existing conversation. While many typically developing children pick up these skills through experience and exposure, the challenges associated with ID and/or ADHD may make this experiential learning more difficult for those with the disorder.

These social conventions are often particularly difficult for those with ADHD, as their impulsivity often impedes their ability to communicate appropriately. Additionally, children with ID/ADHD may require direct instruction in the area of understanding social conventions so as to build appropriate skills as well as to generalize these skills to new or novel situations. It is important to remember that these individuals may require significant practice and repetition and that their ability to extrapolate from one situation to another may be limited.

A number of techniques may be used to help teach students social convention skills. For example, at the most basic level, the use of modeling can be beneficial. If a teacher or student is able to model appropriate interaction skills, it provides a strong visual example for students who are weaker in this area. However, a more effective technique is to allow students the opportunity to practice the skills themselves through role-playing scenarios (Spence, 2003). In these situations, students are able to mimic situations in which they may find themselves and provide in-the-moment responses to the scenarios. For example, a student may practice beginning a conversation appropriately, allowing for conversational turn taking. As well, more advanced topics may be incorporated, such as responding appropriately or confidently to bully or peer pressure situations.

Together, the building of social skills may allow those with ID/ADHD a greater opportunity to engage with their peers at home, at school, and in their communities. Given the positive long-term outcomes related to friendships, ensuring that these children develop the necessarily skills for appropriate peer interaction should be an important consideration in support planning.

Behavioral Interventions

Behavioral interventions are also central to the treatment and skill building of individuals with ID and ADHD; they allow for an improvement in daily living skills and reduction of maladaptive behaviors (Benson & Havercamp, 2007). While there is limited research examining behavioral interventions among individuals with comorbid ID and ADHD, behavioral interventions commonly used to support individuals with ID can be easily adapted to accommodate for ADHD-related behaviors. The following section will discuss some effective behavioral interventions for individuals with ID or ADHD, including reinforcement, token economy, and relaxation training, and how they can be adapted to accommodate co-occurring ID/ADHD.

Reinforcement. The concept of reinforcement forms the basis for most behavioral interventions. A reinforcer is a stimulus that increases some form of behavior or outcome (Benson & Havercamp, 2007). Positive reinforcement, then, is rewarding positive or desired behavior with a motivating stimulus or outcome (Miltenberger, 2001). For example, a child may be more inclined to clean his or her room with the knowledge that he or she may be rewarded upon completion of the task (e.g., they may get a new toy or have an ice cream treat). Individuals with ID and ADHD (individually) both react well to positive reinforcement (Benson & Havercamp, 2007; Pfiffner & Haack, 2014). However, for positive reinforcement to work effectively, the outcome must be particularly rewarding for the individual. For some children, ice cream may be a strong motivator, but for others, it may have little to no motivating power. Consequently, it may be beneficial to conduct a preference assessment with an individual prior to using a positive reinforcement paradigm. Sturmey, Lee, Reyer, and Robek (2003) created the Choice Assessment Scale, which may be used to identify potential reinforcers for individuals with ID. The reinforcer should also be received or administered in close succession to the desired behavior, as individuals with ID and ADHD struggle to understand and appreciate delayed gratification (Pfiffner & Haack, 2014).

Negative reinforcement, or the encouraging a behavior by stopping or removing a stimulus, may also be used. For example, allowing a child to come out of a time out after they apologize may help to increase apologizing behavior. However, although negative reinforcement has some effect, it is often less motivating for individuals with ID and ADHD (Pfiffner & Haack, 2014); therefore, it may be more beneficial to use positive reinforcement more frequently.

Token economy. Token economy is a popular form of reinforcement used to support individuals with ID and ADHD. The reinforcer, or "token," acts in a similar manner to traditional money: it can be exchanged for a desired stimulus or outcome in the future (Benson & Havercamp, 2007). For example, if a child receives a marble for demonstrating good behavior, he or she can "save" the marbles until a marble jar is full. After that time, the full marble jar can be exchanged for a reward of his or her choosing. There are numerous benefits to using a token system: they are tangible, received immediately, and can be collected to earn a greater future reward (Benson & Havercamp, 2007).

To create an effective token economy, choose a negative behavior to be changed and a positive behavior to replace the negative behavior (e.g., stealing food from the cupboard should be replaced with asking for a snack). Tokens should be given based on predetermined intervals or outcomes. For example, a token could be given each time a positive replacement behavior is observed (i.e., the child asks for a snack). Once enough tokens have been earned (based on a predetermined threshold), they can then be exchanged for some greater reward (Pfiffner & Haack, 2014), such as a trip to the ice cream store. Token economy is an effective strategy for children, adolescents, and adults with ADHD and ID (Benson & Havercamp, 2007; Pfiffner & Haack, 2014). Matson (1982), for example, provided tokens to adult men with mild ID for each minute out of 30 that a target behavior (repetitive clothes checking) did not occur. That token could be later exchanged for snacks. Over time, the occurrences of the undesirable behavior reduced.

Relaxation training. Relaxation training, meant to reduce symptoms of anxiety, is an effective behavioral strategy for both children and adults (Benson & Havercamp, 2007). Individuals with both ADHD and ID experience tension and anxiety due to difficulty meeting the demands of their environment (Lim, 2009; Pfiffner & Haack, 2014). With appropriate accommodations, relaxation training can reduce aggressive and disruptive behaviors, hyperactivity, and anxiety in individuals with ID (Lim, 2009). It can also help to improve concentration and attention to tasks, an important component for those with ADHD (Lim, 2009). Lim (2009) completed a relaxation training program with teenagers with mild ID, some of whom also had ADHD. The relaxation exercises include deep breathing, listening to calming music, progressive muscle relaxation, and picturing a special place. Lim (2009) reported reductions in anger, and greater feelings of calmness, relaxation, and concentration.

One of the most common forms of relaxation training is that of progressive muscle relaxation, where the individual tenses, holds, and relaxes muscle groups (Sturmey, Lindsay, Vause, & Neil, 2014). Progressive muscle relaxation has been adapted to accommodate the needs of individuals with ID, and is referred to as abbreviated progressive muscle relaxation (APMR). Here, the instructor models and guides the individual through the movements, providing manual instructions and prompts as necessary. Progressive muscle relaxation can be further accommodated for the needs of those with ADHD by conducting the exercise in very short bursts, by allowing for “inattentive relaxation,” or by allowing movement during the activity (Pfiffner & Haack, 2014).

Academic Interventions

Given that success on academic tasks is strongly predicted by attentional abilities (e.g., Kirk, Gray, Ellis, Taffe, & Cornish, 2016), researchers have acknowledged the importance of studying evidence-based interventions that specifically support the success of those students who struggle academically and/or with attention (Roording-Ragetlie, Klip, Buitelaar, & Slaats-Willemse, 2017). Furthermore, changes in legislation that have made inclusion a priority in education have increased the need to create academic interventions that allow for greater access to education for diverse learners (Snodgrass, Israel, & Reese, 2016). Although many academic supports can be applicable for the ID/ADHD population, this section will highlight two of the most relevant and evidence-based techniques: Computer-Based Interventions, also known as Computer-Assisted Instruction, and Peer Tutoring.

Computer Assisted Instruction (CAI). CAI shows promise as an academic intervention to target attention and working memory (Weng, Maeda, & Bouck, 2014). Also known as Computer Based Learning or Computer Based Teaching, CAI is defined as educational technology in which instruction can be delivered through computer programs or other type of computer-based technology (Weng et al., 2014). Four primary types of CAI exist: Visual, Auditory, Mobile, and Cognitive skills-based (Weng et al., 2014). Visual and auditory computer assisted instruction can provide support for students that need computer devices to complete academic tasks. Examples of these computer-based interventions include speech-to-text software (Van Laere & van Braak, 2017). Mobile CAI includes the use of tablets, which have generated interest in educational settings because they can be easily transported and their use is simple and interactive (Weng et al., 2014). Mobile technologies have been used with students with autism spectrum disorder as well as moderate and severe intellectual disabilities to teach life skills such as cooking, time and behavioral management (Ayres, Mechling, & Sansosti, 2013). Cognitive skills-based CAI has shown to be the most effective for children with attentional concerns. Specifically, studies have shown that children with ADHD who use CAI tend to spend less time in off-task behaviors and appear to be more actively engaged compared to other methods (Murray & Rabiner, 2014), such as handwritten note-taking or just looking at information explained by drawing or writing on a whiteboard.

Furthermore, computer-based technologies also seemed to be preferred by individuals with intellectual disabilities, as the immediate and direct feedback provided by computer games and programs helps to maintain attention and engagement in the program (Kirk et al., 2016). Through CAI, the intensity or length of intervention can be reduced and, because software access is becoming more accessible, the duration of interventions can be extended of longer periods of time (Roording-Ragetlie et al., 2017). This increasing duration is important because prolonging the use of an intervention to reduce cognitive load may be beneficial for children with mild IDs and neuropsychiatric disorders (Roording-Ragetlie et al., 2017). Although CAI has also been used to develop academic skills in students with ADHD, including reading and mathematics (DuPaul et al., 2006; Mautone, DuPaul, & Jitendra, 2005), few studies have examined the use of CAI solely in the ID population. A recent study in Brazil targeted reading and writing skills with parents as monitors of the CAI intervention (Benitez & Domeniconi, 2016). The reading and writing program was available to students from a home computer and a high-school graduated family member was trained to provide monitoring during sessions (20 to 30 minutes, three times a week, before or after school). All students improved the target reading and writing skills after the intervention (Benitez & Domeniconi, 2016). These results suggest that academic interventions targeting reading and writing could also be effective in this population when they receive support from their caregivers or support workers.

Literature reviews aiming to identify the effectiveness of CAI programs in students with ADHD or other disorders have shown small effect sizes (Ramdoss et al., 2012; Weng et al., 2014). Some of the issues with the implementation of Cognitive training in research have been inconsistencies in training regimes (e.g., lack of clarity in the frequency of implementation; Kirk, Gray, Riby, & Cornish, 2015) as well as significant heterogeneity in the targeted skills: some studies have targeted cognitive skills like attention and memory, while others have focused on teaching basic living skills, mathematical, reading, or writing skills (Weng et al., 2014). With the broad range of skills that are being targeted, it is difficult to identify the academic areas cognitive training has a positive impact on. Research is still needed to establish CAI as an effective and evidence-based intervention for children and youth with comorbid ADHD/ID.

Peer tutoring. This instructional strategy involves two students collaborating in an academic activity by teaching, assisting, and providing feedback on an academic task (DuPaul, Ervin, Hook, & McGoey, 1998). Peer-assisted learning focuses on enhancing learning and motivation by recognizing that peers are able to instruct each other or act as positive academic models for academic tasks (Rohrbeck, Ginsburg-Block, Fantuzzo, & Miller, 2003). Some evidence of effectiveness of the use of peer tutoring with children with ADHD to learn academic material has been found (Raggi & Chronis, 2006; Richardson et al., 2015). However, few studies have researched its effectiveness for students with ID specifically and no literature has explored this technique for the ID/ADHD population. However, class-wide peer tutoring to improve spelling has been used in regular education classrooms by pairing typically developing children with students with disabilities (Mortweet et al., 1999). All of the children, including those with a either mild intellectual disabilities or ADHD, improved their spelling scores by the end of the intervention (Mortweet et al., 1999), suggesting that there may be benefits for using this type of intervention support to build academic skills in the ID/ADHD population.

Adaptive Supports

Individuals with ID show similar sequences of development to typically developing children; consequently, an understanding of universal sequences in development is essential for intervention with the ID/ADHD population (Harris, 2006). Adopting a developmental approach to ID will allow for emphasizing where the individual with ID/ADHD is at developmentally and then build upon existing skills across multiple domains. Given that individuals with ID/ADHD face a number of challenges relating to adaptive functioning across conceptual, practical, and academic domains, successful treatment requires adaptive supports to address challenges across these domains. When adaptive behaviors are assessed, the individual's behavior is compared to expectations based on culture and age, generally to national norm groups by age (Fiorello & Jenkins, 2018). Thus, adaptive supports may be most beneficially applied when consideration is given to the age and culturally based expectations that are relevant for the individual with ID/ADHD. These expectations for adaptive functioning will vary for children, adolescents, and adults with ID/ADHD.

Children. Children with ID/ADHD must demonstrate mastery of skills for there to be growth and development across adaptive functioning domains (Harris, 2006). For example, a child with ID/ADHD who has difficulty with their verbal comprehension and language skills has gaps in these foundational skill areas that may impact their adaptive functioning relating to academics, making it difficult for them to understand what they read and understand reading-based material in and outside of school. Demonstration of prerequisite skills is necessary for further development in other domains and allow for setting goals for next steps in treatment plans.

Across conceptual, practical, and social domains, the level of adaptive supports provided to target challenges across these areas will depend on the severity of intellectual disability that the child has as well as the type and severity of comorbid ADHD. In terms of the conceptual adaptive domain, school-age children with ID/ADHD will tend to have slower progress in reading, writing, and mathematics and understanding is markedly limited compared to peers (APA, 2013). Adaptive supports that address these challenges in the conceptual domain require educational supports with simplifying academic material, provisions of extra time to complete academic work, as well as use of a scribe, computer, calculator and other technological supports to accompany teaching strategies. With the social domain, the spoken language of a child with ID/ADHD may be less complex that that of peers, there may be challenges with perceiving or interpreting social cues accurately, and social judgment and decision-making is limited (APA, 2013). Adaptive supports to address challenges in the social domain will require that the child be explicitly taught about social concepts such as trust, how to be a good friend, and simple approaches for how to navigate social conflict, such as a friend hurting one's feelings. The practical domain includes challenges relating to one's capacity to meet his or her own personal needs involving dressing, eating, elimination, and hygiene (APA, 2013). Depending on the severity of adaptive functioning deficits experienced by the child with ID/ADHD, he or she may require daily support from a caregiver or aide with meeting their personal needs. Some children with milder ID/ADHD will require adaptive support in the form of modeling and one-on-one teaching with visual reminders for how to carry out personal care steps, such as how to get dressed and how to brush teeth.

Adolescents and adults. A child with ID/ADHD who enters adolescence may present as developmentally younger in comparison to their peers across conceptual, social, and practical domains of adaptive functioning. Unfortunately, the demands of social relationships, academics, and expectations for autonomy and independence increase from childhood into adolescence, which are difficult for a youth with ID/ADHD to meet. Adaptive supports in the form of an assistive aid worker within the home/school or living in a residential treatment program from childhood may have to continue to be in place for the adolescent with progression towards teaching specific skills for adulthood, such as employment and money-managing skills. For older adolescents, the transitional periods from school completion into adult life is difficult if the youth is in a position where they are leaving a monitored, individualized program with same-age peers to adult programming that encompasses a range of ages and diagnoses (Harris, 2006). Consideration of provincial funding eligibility options and program placement may be informed through formal assessment of adaptive functioning through social work and psychological services.

Other Adult Supports

As individuals with ID/ADHD age out of the youth system, their needs do not dissipate. Indeed, they may actually require more intensive support, as they are often not able to manage the typical expectations of being an adult, such as independent living, maintaining employment, and self-advocacy. This section will outline some relevant supports that may be beneficial when transition planning or when supporting adults with ID/ADHD in their homes, communities, and places of employment.

Occupational supports. Those with ID/ADHD often find satisfaction when they have some kind of meaningful employment. These individuals note that they enjoy the opportunity to connect with others, enjoy the benefits of pay, develop feelings of pride and satisfaction, and have the opportunity to develop new learning experiences (Lysaght, Ouellette-Kuntz, & Lin, 2012). Additionally, in some workplaces, these individuals may be valued for their contributions to the workplace, although there are often challenges to the integration of those with disabilities into the workforce (Lysaght et al., 2012).

For individuals with ID/ADHD, they may be supported through a number of means. For example, the use of agency partnerships may help to bridge the challenges that those with ID/ADHD may experience (Lysaght et al., 2012). These agencies may be able to provide an organization with training and guidance for their staff regarding the supports that individuals with ID/ADHD may need to be successful in the workplace (e.g., social supports, physical supports; Lysaght & Larmour-Trode, 2008) and help to manage any problems that arise. Having this outside support may be invaluable for ensuring that the individual with ID/ADHD is not only accepted into the workplace but can be a productive and contributing member of the work team.

As well, having a greater connection between K-12 schooling and occupational opportunities may be particularly beneficial for those with ID/ADHD. For example, allowing and encouraging those with ID/ADHD to become involved with vocational or work training programs while still in school may allow them to build occupational skills as well as develop connections with local organizations (Beyer & Kaehne, 2008). If schools are able to build collaborative connections with community agencies and partners, then vocational preparation may begin at an earlier age. As well, the range of opportunities available to these individuals may be expanded (Lysaght et al., 2012). For example, job shadowing or workplace visitations/observations may encourage these students to further explore educational opportunities beyond high school completion. As well, those with ID/ADHD may gain a greater awareness of employment opportunities available to them, which may increase their motivation to complete their high school coursework.

Independent living supports. As to be expected, those with ID/ADHD vary in their ability to live independently; most of these individuals have some desire to live as independently as possible, with some autonomy in their decision making (Kozma et al., 2009; Shaw, Cartwright, & Craig, 2011). Some individuals may be able to be more independent than others, depending on their level of ID. For example, some may be successful with minimal supports, such as with a supportive roommate or in a supported living facility, while others may require more intensive support, such as in a group or residential home with higher levels of supervision and guidance. In a review of placement settings, Kozma et al. (2009) report that a majority of studies indicated that deinstitutionalized placements, such as those offered through community care arrangements, had greater benefits than group living arrangements, although the needs of the individual, particularly around safety, must be considered. Either way, these individuals will require personalized planning to ensure that they are able to live as independently as possible, taking into consideration their strengths, limitations, needs, and desires.

As previously noted, building social skills in this population may have protective benefits. The development of social skills can also have an impact on an individual's quality of life, particularly in relation to independent living skills (Wehmeyer & Garner, 2003). As well, connectedness with others has been frequently linked to better quality of life, with those who live in supported living arrangements having a greater number of friends and visitors and are more likely to be known by those who live around them than those who live alone (Kozma et al., 2009). Ensuring that those in the ID/ADHD community have the opportunity to connect with others with ID, as well as have positive interactions in the community may help to foster their skills in these areas.

Finally, it should also be noted that working with individuals with ID/ADHD from a person-centered approach is also recommended. This approach acknowledges the preferences and desires of those with a disability and their caregivers in making placement decisions. As well, this approach encourages those with ID/ADHD to become active and involved in their own decision making and planning when choosing housing and support services. Individuals are more likely to enjoy their living arrangements when they feel as though they have had some input into the process.

Self-advocacy. Building on the idea of the person-centered approach, an additional area of persistent need is to ensure that those with ID/ADHD continue to build skills in the area of self-advocacy (Shogren & Plotner, 2012). The ability to advocate for oneself is a critical skill to ensure that those with ID/ADHD have a say in decisions that have direct impact on them. There have been a number of suggestions as to how to build leadership and self-advocacy skills in those with ID, although many of these recommendations would also apply to those with ID/ADHD. One manner in which to build self-advocacy skills is to encourage those with ID/ADHD to participate in volunteering or other community service opportunities (Caldwell, 2010). For example, involvement in one-off special community events (e.g., fun runs, celebration events) or more long-term volunteering opportunities (e.g., weekly shifts at the local humane society), based on the individual's interests or desires, may help to increase feelings of competence and independence. Enhancing leadership opportunities for this population may allow them to develop stronger communication, social, and life skills, all of which may lead to enhancement of their self-advocacy skills.

Additionally, those with ID/ADHD should be encouraged to participate on relevant boards or committees, particularly those who may benefit from the presence of someone who lives with a disability (Caldwell, 2010). For example, involvement on organizations associated with supporting those with disabilities (e.g., learning disability associations, independent living support groups) may provide valuable experience for both the organization and the individual. Encouraging those with ID/ADHD to provide direct input to decision making bodies allows them to develop a stronger sense of self-worth and importance and provide an opportunity for them to give honest and practical feedback that may have important and policy-related outcomes.

Finally, encouraging those with ID/ADHD to pursue some form of post-secondary education may also contribute to their ability to self-advocate (Caldwell, 2010). There is currently a greater push for accessibility supports for those with exceptional learning needs of all varieties within post-secondary environments. Consequently, encouraging the pursuit of higher education in areas of interest and ability may significantly benefit the lives of those with ID/ADHD. These educational skills may also have secondary benefits, such as an increase in the employment prospects of these individuals or additional opportunities to build social skills.

Conclusions and Future Directions

Together, this chapter highlights the unique needs of individuals with comorbid ID and ADHD. Due to issues such as diagnostic overshadowing, research examining the co-occurrence of ID and ADHD is extremely limited, and it is only with the recent iteration of the DSM that the possibility of both an ID and an ADHD diagnosis is available. However, it is clear that individuals with both disorders are at greater risk of numerous negative outcomes (e.g., depression, anxiety, aggression; Carmeli et al., 2007) than those with ID or ADHD alone, and thus the importance of specific, targeted, and individualized interventions should not be overlooked.

Although those with ID/ADHD may benefit from support in childhood and adolescence, as we highlighted above, the needs of these individuals as they transition from the youth system to the adult system remain high. Currently the ability to provide this wrap-around care and support as individuals make this transition is limited, sometimes resulting in individuals falling through the proverbial "crack" and ending up on a negative path. Continuing to advocate for supports and resources not only for adults but also to support and guide the transition to adulthood may be extremely beneficial. More specifically, building capacity to allow those with ID/ADHD to advocate for themselves would provide long term benefit for those with the disability as well as their families (Carter, Brock, & Trainor, 2014; Shogren & Plotner, 2012).

In summary, it is necessary to understand the unique and individual needs of those with a combined ID/ADHD diagnosis, as they may benefit from some interventions and recommendation relevant to the ID population as well as those more pertinent to individuals with ADHD. As with all intervention planning, providing individualized support and guidance is key to ensuring that each person can achieve to their potential.

References

Abecassis, M., Isquith, P. K., & Roth, R. M. (2017). Characteristics of ADHD in the emerging adult: An overview. *Psychological Injury and Law, 10*(3), 197–208.

Ahuja, A., Martin, J., Langley, K., & Thapar, A. (2013). Intellectual disability in children with attention deficit hyperactivity disorder. *The Journal of Pediatrics, 163*(3), 890–895.

Aman, M. G., Armstrong, S., Buican, B., & Sillick, T. (2002). Four-year follow-up of children with low intelligence and ADHD: A replication. *Research in Developmental Disabilities, 23*(2), 119–134.

American Psychiatric Association. (2013). *Diagnostic and statistical manual of mental disorders: DSM-5* (5th ed.). Washington, DC: American Psychiatric Association.

Aureli, A., Del Beato, T., Sebastiani, P., Marimpietri, A., Melillo, C. V., Sechi, E., & Di Loreto, S. (2010). Attention-deficit hyperactivity disorder and intellectual disability: A study of association with brain-derived neurotrophic factor gene polymorphisms. *International Journal of Immunopathology and Pharmacology, 23*(3), 873–880.

Ayres, K. M., Mechling, L., & Sansosti, F. J. (2013). The use of mobile technologies to assist with life skills/independence of students with moderate/severe intellectual disability and/or autism spectrum disorders: Considerations for the future of school psychology. *Psychology in the Schools, 50*(3), 259–271.

Baker, B. L., Neece, C. L., Fenning, R. M., Crnic, K. A., & Blancher, J. (2010). Mental disorders in five-year-old children with or without developmental delay: Focus on ADHD. *Journal of Clinical Child and Adolescent Psychology, 39*, 492–505.

Barkley, R. A. (2014). *Attention-deficit hyperactivity disorder: A handbook for diagnosis and treatment* (4th ed.). New York: Guilford Press.

Barkley, R. A., & Peters, H. (2012). The earliest reference to ADHD in the medical literature? Melchior Adam Weikard's description in 1775 of "attention deficit". *Journal of Attention Disorders, 16*(8), 623–630.

Barkley, R. A., Shelton, T. L., Crosswait, C., Moorehouse, M., Fletcher, K., Barrett, S., … Metevia, L. (2000). Multi-method psycho-educational intervention for preschool children with disruptive behavior: Preliminary results at post-treatment. *Journal of Child Psychology and Psychiatry, and Allied Disciplines, 41*(3), 319–332.

Benitez, P., & Domeniconi, C. (2016). Use of a computerized reading and writing teaching program for families of students with intellectual disabilities. *Psychological Record, 66*(1), 127–138.

Benson, B., & Havercamp, S. (2007). Behavioural approaches to treatment: Principles and practices. In N. Bouras & G. Holt (Eds.), *Behavioural disorders in intellectual and developmental disabilities* (2nd ed., pp. 283–309). Cambridge: Cambridge University Press.

Beyer, S., & Kaehne, A. (2008). The transition of young people with learning disabilities to employment: What works? *Journal on Developmental Disabilities, 14*(1), 81–90.

Caldwell, J. (2010). Leadership development of individuals with developmental disabilities in the self-advocacy movement. *Journal of Intellectual Disability Research, 54*(11), 1004–1014.

Carmeli, E., Klein, N., & Sohn, M. (2007). The implications of having attention-deficit/hyperactivity disorder in male adolescents with intellectual disability. *International Journal of Adolescent Medicine and Health, 19*(2), 209–214.

Carter, E. W., Brock, M. E., & Trainor, A. A. (2014). Transition assessment and planning for youth with severe intellectual and developmental disabilities. *The Journal of Special Education, 47*(4), 245–255.

Carulla, L. S., Reed, G. M., Vaez-Aziz, L. M., Cooper, S. A., Leal, R., Bertelli, M., … Girimaji, S. C. (2011). Intellectual developmental disorders: Towards a new name, definition and framework for "mental retardation/intellectual disability" in ICD-11. *World Psychiatry, 10*(3), 175–180.

Castellanos, F. X., & Tannock, R. (2002). Neuroscience of attention-deficit/hyperactivity disorder: The search for endophenotypes. *Nature Reviews Neuroscience, 3*(8), 617–628.

Charach, A., Yeung, E., Climans, T., & Lillie, E. (2011). Childhood attention-deficit/hyperactivity disorder and future substance use disorders: Comparative meta-analyses. *Journal of the American Academy of Child & Adolescent Psychiatry, 50*(1), 9–21.

Climie, E. A., Mah, J. W., & Chase, C. Y. (2017). Clinical reasoning in the assessment and intervention planning for attention-deficit/hyperactivity disorder. *Canadian Journal of School Psychology, 32*(1), 31–45.

Deb, S., Thomas, M., & Bright, C. (2001). Mental disorder in adults with intellectual disability. 1: Prevalence of functional psychiatric illness among a community-based population aged between 16 and 64 years. *Journal of Intellectual Disability Research, 45*(6), 495–505.

Dekker, M. C., & Koot, H. M. (2003). DSM-IV disorders in children with borderline to moderate intellectual disability. I: Prevalence and impact. *Journal of the American Academy of Child & Adolescent Psychiatry, 42*(8), 915–922.

Di Nuovo, S. F., & Buono, S. (2007). Psychiatric syndromes comorbid with mental retardation: Differences in cognitive and adaptive skills. *Journal of Psychiatric Research, 41*(9), 795–800.

DuPaul, G. J., Ervin, R. A., Hook, C. L., & McGoey, K. E. (1998). Peer tutoring for children with attention deficit hyperactivity disorder: Effects on classroom behavior and academic performance. *Journal of Applied Behavior Analysis, 31*(4), 579–592.

DuPaul, G. J., Jitendra, A. K., Volpe, R. J., Tresco, K. E., Lutz, J. G., Junod, R. E. V., … Mannella, M. C. (2006). Consultation-based academic interventions for children with ADHD: Effects on reading and mathematics achievement. *Journal of Abnormal Child Psychology, 34*(5), 633–646.

Durkin, M. (2002). The epidemiology of developmental disabilities in low-income countries. *Developmental Disabilities Research Reviews, 8*(3), 206–211.

Emerson, E., & McVilly, K. (2004). Friendship activities of adults with intellectual disabilities in supported accommodation in Northern England. *Journal of Applied Research in Intellectual Disabilities, 17*(3), 191–197.

Fiorello, C. A., & Jenkins, T. K. (2018). Best practices in intellectual disability identification. *Communiqué, 46*(5), 18–20.

Frankel, F., Myatt, R., Cantwell, D. P., & Feinberg, D. T. (1997). Parent-assisted transfer of children's social skills training: Effects on children with and without attention deficit hyperactivity disorder. *Journal of the American Academy of Child and Adolescent Psychiatry, 36*, 1056–1064.

Friedberg, R. D., & McClure, J. M. (2015). Working with disruptive children and adolescents. In *Clinical practice of cognitive therapy with children and adolescents: The nuts and bolts* (2nd ed., pp. 316–360). Retrieved from https://ebookcentral-proquest-com.ezproxy.lib.ucalgary.ca

Fuller, C. G., & Sabatino, D. A. (1998). Diagnosis and treatment considerations with comorbid developmentally disabled populations. *Journal of Clinical Psychology, 54*(1), 1–10.

Gaastra, G. F., Groen, Y., Tucha, L., & Tucha, O. (2016). The effects of classroom interventions on off-task and disruptive classroom behavior in children with symptoms of attention-deficit/hyperactivity disorder: A meta-analytic review. *PLoS One, 11*(2), 1–19.

Gresham, F. M., Sugai, G., & Horner, R. H. (2001). Interpreting outcomes of social skills training for students with high-incidence disabilities. *Exceptional Children, 67*(3), 331–344.

Harris, J. C. M. (2006). *Intellectual disability: Understanding its development, causes, classification, evaluation, and treatment*. Retrieved from http://ebookcentral.proquest.com

Harvey, E. A., Breaux, R. P., & Lugo-Candelas, C. I. (2016). Early development of comorbidity between symptoms of attention-deficit/hyperactivity disorder (ADHD) and oppositional defiant disorder (ODD). *Journal of Abnormal Psychology, 125*(2), 154–167.

Hässler, F., & Thome, J. (2012). Mental retardation and ADHD. *Zeitschrift für Kinder- und Jugendpsychiatrie und Psychotherapie, 40*(12), 83–93.

Hoza, B. (2007). Peer functioning in children with ADHD. *Academic Pediatrics, 7*(1), 101–106.

Johnson, C. R., Lubetsky, M. J., & Sacco, K. A. (1995). Psychiatric and behavioral disorders in hospitalized preschoolers with developmental disabilities. *Journal of Autism and Developmental Disorders, 25*(2), 169–182.

Kirk, H. E., Gray, K., Riby, D. M., & Cornish, K. M. (2015). Cognitive training as a resolution for early executive function difficulties in children with intellectual disabilities. *Research in Developmental Disabilities, 38*, 145–160.

Kirk, H. E., Gray, K. M., Ellis, K., Taffe, J., & Cornish, K. M. (2016). Computerised attention training for children with intellectual and developmental disabilities: A randomised controlled trial. *Journal of Child Psychology and Psychiatry, 57*(12), 1380–1389.

Koziol, L. F., & Budding, D. (2012). Requiem for a diagnosis: Attention-deficit hyperactivity disorder. *Applied Neuropsychology: Child, 1*(1), 2–5.

Kozma, A., Mansell, J., & Beadle-Brown, J. (2009). Outcomes in different residential settings for people with intellectual disability: A systematic review. *American Journal on Intellectual and Developmental Disabilities, 114*(3), 193–222.

Krain, A. L., & Castellanos, F. X. (2006). Brain development and ADHD. *Clinical Psychology Review, 26*(4), 433–444.

Kuntsi, J., Eley, T. C., Taylor, A., Hughes, C., Asherson, P., Caspi, A., & Moffitt, T. E. (2004). Co-occurrence of ADHD and low IQ has genetic origins. *American Journal of Medical Genetics Part B: Neuropsychiatric Genetics, 124*(1), 41–47.

Lambek, R., Tannock, R., Dalsgaard, S., Trillingsgaard, A., Damm, D., & Thomsen, P. H. (2011). Executive dysfunction in school-age children with ADHD. *Journal of Attention Disorders, 15*(8), 646–655.

Laugeson, E. A., Frankel, F., Gantman, A., Dillon, A. R., & Mogil, C. (2012). Evidence-based social skills training for adolescents with autism spectrum disorders: The UCLA PEERS program. *Journal of Autism and Developmental Disorders, 42*(6), 1025–1036.

Laugeson, E. A., Frankel, F., Mogil, C., & Dillon, A. R. (2009). Parent-assisted social skills training to improve friendships in teens with autism spectrum disorders. *Journal of Autism and Developmental Disorders, 39*(4), 596–606.

Lim, H. (2009). *The effectiveness of relaxation exercises for persons with mild intellectual disability*. Paper presented at the 19th Asian Conference on Mental Retardation, Singapore.

Lindsey, M. (2002). Comprehensive health care services for people with learning disabilities. *Advances in Psychiatric Treatment, 8*(2), 138–147.

Lysaght, R. M., & Larmour-Trode, S. (2008). An exploration of social support as a factor in the return-to-work process. *Work, 30*(3), 255–266.

Lysaght, R., Cobigo, V., & Hamilton, K. (2012). Inclusion as a focus of employment-related research in intellectual disability from 2000 to 2010: A scoping review. *Disability and Rehabilitation, 34*(16), 1339–1350.

Magnin, E., & Maurs, C. (2017). Attention-deficit/hyperactivity disorder during adulthood. *Revue Neurologique, 173*(7-8), 506–515.

Mahour, P., & Panday, S. (2015). Change in intellectual disability in DSM-5. *International Journal of Contemporary Issues in Behavioural Sciences, 1*(2), 46–50.

Matson, J. L. (1982). Treating obsessive-compulsive behavior in mentally retarded adults. *Behavior Modification, 6*(4), 551–567.

Maulik, P. K., Mascarenhas, M. N., Mathers, C. D., Dua, T., & Saxena, S. (2011). Prevalence of intellectual disability: A meta-analysis of population-based studies. *Research in Developmental Disabilities, 32*(2), 419–436.

Mautone, J. A., DuPaul, G. J., & Jitendra, A. K. (2005). The effects of computer-assisted instruction on the mathematics performance and classroom behavior of children with ADHD. *Journal of Attention Disorders, 9*(1), 301–312.

Merrell, K. W., & Wolfe, T. M. (1998). The relationship of teacher-rated social skills deficits and ADHD characteristics among kindergarten-age children. *Psychology in the Schools, 35*(2), 101–110.

Miltenberger, R. G. (2001). *Behavior modification: Principles and procedures* (2nd ed.). Belmont, CA: Wadsworth.

Miyake, A., Friedman, N. P., Emerson, M. J., Witzki, A. H., Howerter, A., & Wager, T. D. (2000). The Unity and diversity of executive functions and their contributions to complex "frontal lobe" tasks: A latent variable analysis. *Cognitive Psychology, 41*(1), 49–100.

Mortweet, S. L., Utley, C. A., Walker, D., Dawson, H. L., Delquadri, J. C., Reddy, S. S., … Ledford, D. (1999). Classwide peer tutoring: Teaching students with mild mental retardation in inclusive classrooms. *Exceptional Children, 65*(4), 524–536.

Mouzakitis, G. S. (2012). Intellectual disability management, causes, and social perceptions towards well-defined situations. In D. F. Mancini & C. M. Greco (Eds.), *Intellectual disability: Management, causes and social perceptions*. New York: Nova Science Publishers.

Mueller, A., Hong, D. S., Shepard, S., & Moore, T. (2017). Linking ADHD to the neural circuitry of attention. *Trends in Cognitive Sciences, 21*(6), 474–488.

Murray, D. W., & Rabiner, D. L. (2014). Teacher use of computer-assisted instruction for young inattentive students: Implications for implementation and teacher preparation. *Journal of Education and Training Studies, 2*(2), 58–66.

Neece, C. L., Baker, B. L., Blacher, J., & Crnic, K. A. (2011). Attention-deficit/hyperactivity disorder among children with and without intellectual disability: An examination across time. *Journal of Intellectual Disability Research, 55*(7), 623–635.

Nigg, J. T., & Barkley, R. A. (2014). Attention-deficit/hyperactivity disorder. In E. J. Mash & R. A. Barkley (Eds.), *Child psychopathology* (pp. 75–144). New York: Guildford Press.

Pearson, D. A., Lachar, D., Loveland, K. A., Santos, C. W., Faria, L. P., Azzam, P. N., … Cleveland, L. A. (2000). Patterns of behavioral adjustment and maladjustment in mental retardation: Comparison of children with and without ADHD. *American Journal on Mental Retardation, 105*(4), 236–251.

Pfiffner, L. J., & Haack, L. M. (2014). Behavior management for school aged children with ADHD. *Child and Adolescent Psychiatric Clinics of North America, 23*(4), 731–746.

Psychological Corporation. (2008). *WAIS-IV technical and interpretive manual*. San Antonio, TX: Author.

Raggi, V. L., & Chronis, A. M. (2006). Interventions to address the academic impairment of children and adolescents with ADHD. *Clinical Child and Family Psychology Review, 9*(2), 85–111.

Ramdoss, S., Lang, R., Fragale, C., Britt, C., O'Reilly, M., Sigafoos, J., … Lancioni, G. E. (2012). Use of computer-based interventions to promote daily living skills in individuals with intellectual disabilities: A systematic review. *Journal of Developmental and Physical Disabilities, 24*(2), 197–215.

Reiter, S., Bryen, D. N., & Shachar, Y. (2007). Adolescents with intellectual disabilities as victims of crimes. *Journal of Intellectual Disability Research, 11*, 1–17.

Richardson, M., Moore, D. A., Gwernan-Jones, R., Thompson-Coon, J., Ukoumunne, O., Rogers, M., … Taylor, E. (2015). Non-pharmacological interventions for attention-deficit/hyperactivity disorder (ADHD) delivered in school settings: systematic reviews of quantitative and qualitative research. *Health Technology Assessment, 19*(45), 1.

Rohrbeck, C. A., Ginsburg-Block, M. D., Fantuzzo, J. W., & Miller, T. R. (2003). Peer-assisted learning interventions with elementary school students: A meta-analytic review. *Journal of Educational Psychology, 95*(2), 240–257.

Roid, G. H. (2003). *Stanford-Binet Intelligence Scales: Technical manual* (5th ed.). Itasca, IL: Riverside Publishing.

Roording-Ragetlie, S., Klip, H., Buitelaar, J., & Slaats-Willemse, D. (2017). Working memory training in children with neuropsychiatric disorders and mild to borderline intellectual functioning, the role of coaching; a double-blind randomized controlled trial. *BMC Psychiatry, 17*(1), 1–10.

Seidman, L. J. (2006). Neuropsychological functioning in people with ADHD across the lifespan. *Clinical Psychology Review, 26*(4), 466–485.

Shaw, K., Cartwright, C., & Craig, J. (2011). The housing and support needs of people with an intellectual disability into older age. *Journal of Intellectual Disability Research, 55*(9), 895–903.

Shogren, K. A., & Plotner, A. J. (2012). Transition planning for students with intellectual disability, autism, or other disabilities: Data from the National Longitudinal Transition Study-2. *Intellectual and Developmental Disabilities, 50*(1), 16–30.

Sibley, M. H., Kuriyan, A. B., Evans, S. W., Waxmonsky, J. G., & Smith, B. H. (2014). Pharmacological and psychosocial treatments for adolescents with ADHD: An updated systematic review of the literature. *Clinical Psychology Review, 34*(3), 218–232.

Snodgrass, M. R., Israel, M., & Reese, G. C. (2016). Instructional supports for students with disabilities in K-5 computing: Findings from a cross-case analysis. *Computers & Education, 100*, 1–17.

Sparrow, S. S., Cicchetti, D. V., & Balla, D. A. (2005). *Vineland adaptive behavior scales* (2nd ed.). Circle Pines, MN: American Guidance Service.

Spence, S. H. (2003). Social skills training with children and young people: Theory, evidence and practice. *Child and Adolescent Mental Health, 8*(2), 84–96.

Steinberg, E. A., & Drabick, D. A. (2015). A developmental psychopathology perspective on ADHD and comorbid conditions: The role of emotion regulation. *Child Psychiatry & Human Development, 46*(6), 951–966.

Sturmey, P., Lee, R., Reyer, H., & Robek, A. (2003). Assessing preferences for staff: Some pilot data. *Behavioural and Cognitive Psychotherapy, 31*, 103–107.

Sturmey, P., Lindsay, W. R., Vause, T., & Neil, N. (2014). Anxiety disorders. In P. Sturmey & R. Didden (Eds.), *Evidence-based practices and intellectual disabilities*. West Sussex: John Wiley & Sons.

Takeda, T., Ambrosini, P. J., deBerardinis, R., & Elia, J. (2012). What can ADHD without comorbidity teach us about comorbidity? *Research in Developmental Disabilities, 33*(2), 419–425.

Taylor, E. (2011). Antecedents of ADHD: A historical account of diagnostic concepts. *ADHD Attention Deficit and Hyperactivity Disorders, 3*(2), 69–75.

Thompson, A. L., Molina, B. S., Pelham, W., Jr., & Gnagy, E. M. (2007). Risky driving in adolescents and young adults with childhood ADHD. *Journal of Pediatric Psychology, 32*(7), 745–759.

Van Laere, E., & van Braak, J. (2017). Auditory support in linguistically diverse classrooms: Factors related to bilingual text-to-speech use. *Journal of Computer Assisted Learning, 33*(1), 79–95.

Voigt, R. G., Barbaresi, W. J., Colligan, R. C., Weaver, A. L., & Katusic, S. K. (2006). Developmental dissociation, deviance, and delay: Occurrence of attention-deficit–hyperactivity disorder in individuals with and without borderline-to-mild intellectual disability. *Developmental Medicine and Child Neurology, 48*(10), 831–835.

Wehmeyer, M. L., & Garner, N. W. (2003). The impact of personal characteristics of people with intellectual and developmental disability on self-determination and autonomous functioning. *Journal of Applied Research in Intellectual Disabilities, 16*(4), 255–265.

Weng, P. L., Maeda, Y., & Bouck, E. C. (2014). Effectiveness of cognitive skills-based computer-assisted instruction for students with disabilities: A synthesis. *Remedial and Special Education, 35*(3), 167–180.

World Health Organization. (2009). *International statistical classification of diseases and health related problems (10th revision)*. Geneva: World Health Organization.

49

Developmental Coordination Disorder and Intellectual Disabilities

Bouwien Smits-Engelsman, Niri Naidoo,
Wendy Aertssen, and Reint Geuze

Introduction

The Importance of Motor Development in ID

Imagine humans without a voluntary motor system. We would not be able to communicate or interact with the environment. How would we ever function? This illustrates that movement—being able to act and react—is a basic capacity of human functioning. Very early in development, around the eighth week of gestation, the first movements of the fetus may be observed. Gradually the movement repertoire is extended and becomes more complex (Timor-Tritsch et al., 1988). The developing nervous system contacts the contracting tissues and help shape the bones and joints in a self-organized way. At some stage during later pregnancy, functional connections in the nervous system are strengthened, whereas the nonfunctional ones are removed again. After birth development continues. Major changes are the exposure to the environment (gravity, sounds, visual stimuli, object characteristics) and the possibility to explore and interact with the environment as far as the sensory and motor systems of the newborn allow. Interaction with the environment through movement elicits sensory consequences, facilitating calibration of the motor system with the varied sensory systems. Learning by active exploration of the environment is a major drive for cognitive development. Thus, cognitive and motor development should be seen as a dynamic interactive process (Crone & Ridderinkhof, 2011). Another domain of functioning where cognitive and motor capacities meet is planning and control of complex motor tasks. For example, in learning to write, language needs to be transferred to finger movements that produce a written text (Van Galen & Morasso, 1998).

What will happen in case of nonoptimal development of the nervous system? Lesions in the motor areas of the brain will likely result in reduced motor skills [e.g., children with cerebral palsy or developmental coordination disorder (DCD) with normal IQ], and lesions in the cognitive areas in reduced cognitive function (e.g., children with ID). However, developmental disorders often cannot be localized so clearly and functions are likely to be sub-served by parts of overlapping brain networks. If the brain networks are nonoptimal and involved in several functions, overlapping

B. Smits-Engelsman (✉) · N. Naidoo
Faculty of Health Sciences, Department of Health and Rehabilitation Sciences, University of Cape Town, Cape Town, South Africa

W. Aertssen
Avans+, University for Professionals,
Breda, The Netherlands

R. Geuze
Developmental and Clinical Neuropsychology,
University of Groningen,
Groningen, The Netherlands

J. L. Matson (ed.), *Handbook of Intellectual Disabilities*, Autism and Child Psychopathology Series, https://doi.org/10.1007/978-3-030-20843-1_49

functional limitations may occur such as ADHD and autism, or ID and poor motor skills.

The message to be taken from this part of the introduction is that although motor capacities and ID seem separate domains of functioning, they are related in varied ways during development and in cognitive-motor tasks that require cognitive planning and control.

Importantly, motor coordination problems cause individuals to struggle with everyday functional tasks and to participate less in physical activity (Mandich, Polatajko, & Rodger, 2003). These motor problems make participating in sport and daily outdoor activities more difficult (Barr & Shields, 2011) leading to more sedentary lifestyle, lower levels of physical activity and poor fitness (Lin & Wuang, 2012; Philips & Holland, 2011). Consequently, it impacts their capacity to execute everyday activities and their health-related fitness. Intellectual disability provides an additional barrier to an active lifestyle and this may lead to the underdevelopment or loss of independence, increased risk of noncommunicable diseases, and health complications later in adulthood (Ells et al., 2006). This suggests that designing specific interventions to address motor impairments, activity limitations and participation restrictions are needed to avert the adverse effect of poor motor coordination and its health complications in ID.

Developmental Coordination Disorder (DCD)

Children develop a range of movement skills through various forms of playful activities and exploration. However, not every child is able to develop adequate motor skills required for everyday activities. Difficulties acquiring movement skills is one of the most common issues in children (Henderson & Henderson, 2002). This group of children has been identified with different kinds of names in the literature. These included clumsiness, motor dyspraxia, minimal brain damage, and perceptual-motor dysfunction. If the motor difficulties are evident from an early age, affect activities of daily living, and are not associated with a known medical condition (cerebral palsy, muscular dystrophy, etc.) the preferred term today is Developmental Coordination Disorder (DCD) (DSM-5, American Psychiatric Association, 2013). DCD is a condition that affects around 5–6 percent of all children (Smits-Engelsman, Magalhães, Oliveira, & Wilson, 2015), with more boys than girls being affected. DCD is characterized by motor difficulties in performing essential movement-based activities. Movements seem clumsy, slow and lack accuracy and balance may be poor. Performing age appropriate activities of daily living often is difficult for children with DCD. However, the manifestation of symptoms tends to vary from one child to another and also across the developmental time, demonstrating heterogeneity. In addition, the signs and symptoms of DCD can show persistence from childhood through adulthood when treatment is not provided.

Skilled motor function is a prerequisite for the performance of activities of daily living (ADL) and (pre)vocational activities. While most children learn seamlessly the motor skills that prepare them well for later school achievement and participation, a significant proportion do not, which places them at a significant disadvantage relative to their peers (Bult, Verschuren, Jongmans, Lindeman, & Ketelaar, 2011). Hence DCD, affects their participation in daily activities (Barnhart, Davenport, Epps, & Nordquist, 2003) Moreover, motor skill difficulties have developmental consequences beyond just motor function and place the child at a significant risk for social, psychological, and health-related issues, even into adulthood (Hellgren, Gillberg, Gillberg, & Enerskog, 1993; Rasmussen & Gillberg, 2000). Thus, there is a pressing need to optimize therapeutic approaches such that motor function and participation of this group of children is promoted (Smits-Engelsman et al., 2018).

It is currently unclear if children with DCD have a pure motor coordination deficit or a motor learning deficit or "only" a motor developmental delay. Most children do not "grow out of it," but some 30–50% do (Geuze, 2007, p. 18), suggesting a developmental delay, whereas the others may have deficit or even a genetic basis for their motor problems (Martin, Piek, & Hay, 2006). Thus, as mentioned, children with DCD constitute a very heterogeneous group (Wilson et al., 2017).

DCD is classified in the *Diagnostic and Statistical Manual for Mental Disorders Fifth Edition* (DSM-5) and is subcategorized within the broader category of "neuro-developmental disorders." Table 49.1 lists the criteria for the classification of DCD in DSM-5.

The criteria for DCD are quite clear. However, DSM does not provide an operationalization for the criteria (Smits-Engelsman, Schoemaker, Delabastita, Hoskens, & Geuze, 2015). For example, for criterion A, it is not specified how to assess "the acquisition and execution of coordinated motor skills is substantially below that expected given the individual's chronological age and opportunity for skill learning and use." Operationalization of the criteria should therefore follow the protocol developed by an international working group that developed the International Clinical Practice Recommendations of Developmental Coordination Disorder (Blank et al., 2019), based on extensive reviews of the literature and consensus between experts.

Table 49.1 Diagnostic criteria specified in the Diagnostic and Statistical Manual of Mental Disorders, fifth edition (DSM-5) of the American Psychiatric Association, (2013)

A. The acquisition and execution of coordinated motor skills is substantially below that expected given the individual's chronological age and opportunity for skill learning and use. Difficulties are manifested as clumsiness (e.g., dropping or bumping into objects) as well as slowness and inaccuracy of performance of motor skills (e.g., catching an object, using scissors or cutlery, handwriting, riding a bike, or participating in sports)
B. The motor skills deficit in Criterion A significantly and persistently interferes with activities of daily living appropriate to chronological age (e.g., self-care and self-maintenance) and impacts academic/school productivity, prevocational and vocational activities, leisure, and play
C. Onset of symptoms is in the early developmental period
D. The *motor skills deficits are not better explained by intellectual disability* (intellectual developmental disorder) or visual impairment and are not attributable to a neurological condition affecting movement (e.g., cerebral palsy, muscular dystrophy, degenerative disorder)

International Clinical Practice Recommendations of Developmental Coordination Disorder

The Clinical Practice Recommendations (CPR) for DCD decomposes each of the criteria into parts, and recommendations are provided on how to operationalize these. For example, for the assessment of motor skills (Criterion A), the CPR recommends the use of reliable and valid motor tests that have standardized norms derived from the population; and for excluding the neurological conditions specified in Criterion D, the CPR recommends a medical assessment preferably by a pediatrician.

Interestingly, the DSM in Criterion D (Table 49.1) suggests that intellectual disability is expected to have an impact on the motor skills deficit. Therefore, in most studies children with an IQ below 70 have been excluded. Indeed, the study by Smits-Engelsman and Hill (2012) shows a linear relationship between IQ and motor skill in a group of 460 children identified with/without motor difficulties from both clinical and educational settings.

On the Criteria for DCD

As for criterion A, "… expected given the individual's chronological age and opportunity for skill learning and use," two cutoff values have been internationally accepted: the fifth and the 15/16th percentile on a standardized motor test (See Subheading Assessment of Primary and Secondary Movement-Related Problems). Few of these tests have been widely accepted or have good validity and reliability. With respect to the opportunities for skill-learning, these may have been less in children with DCD and even more so in children with ID, due to cognitive and social restrictions, making it difficult to estimate how much of the poor coordinated motor skills are to be ascribed to lack of experience. In addition, cognition and social restriction of children with ID also play a role in skill acquisition and when testing a child with ID.

With respect to criterion B, the impact of the motor problems on daily life and academic achievement may be different between children with DCD and DCD-ID. In children with DCD it is already difficult to disentangle language, comprehension and motor contributions in the handwriting difficulties, in children with DCD and mild ID this will be even harder.

Criterion C refers to onset in the developmental period. For older children, adolescents and adults this may be checked retrospectively. Very early diagnosis—say under the age of 6 years—is, however, not recommended because of the instability of early motor development. Young children who may seem atypical may catch up a few months later. Therefore, the CPR recommends to retest young children (<5 years) who seem atypical after half a year.

Criterion D refers to ID. In the working group for the International Clinical Practice Recommendations of Developmental Coordination Disorder (DCD) (Blank et al., 2019) the problem of diagnosing DCD in children with severe cognitive delays or intellectual disorder (ID) has been discussed thoroughly. It was recognized that defining a specific IQ score below which the diagnosis of DCD is precluded seems artificial. Given the complexities of arbitrating between cutoffs and determining discrepancy scores, it is recognized that categorical decisions (above or below a specific IQ score) may be extremely difficult. Looking at the meta-analysis on underlying mechanisms of DCD, a specific IQ score does not seem to be helpful to distinguish between children with DCD and children with coordination problems due to ID, as the construct of DCD is nowadays increasingly supported by neurobiological and neurophysiological findings and not restricted to global intellectual abilities (Wilson et al., 2017). Hence, it was agreed that the motor dysfunction in ID should be defined as DCD if the other criteria are fulfilled and if clinical history and examination cannot explain the motor problems and their impact on daily activities by cognitive status. It is recommended that the complete DCD assessment considers ADL (i.e., self-care and self-maintenance), academic/school productivity, prevocational or vocational activities, leisure, sports, and play.

It is also recommended that the child be checked for visual impairment and neurological conditions affecting movement. For DCD-ID this will exclude all children with such conditions. A future problem may be that refined brain imaging will show that all children will exhibit subtle to more substantial degrees of abnormalities in brain structure and activity, thus rendering this part (no neurological condition) of the criteria questionable. Frequently occurring health problems, such as a congenital heart defects or visual defects, can also influence motor development.

Co-occurring Developmental Disorders

Children with DCD often have co-occurring disorders such as ADHD (Loh, Piek, & Barrett, 2011), and children with developmental disorders such as specific language disorders often have co-occurring motor problems (Pieters et al., 2012). It is known that comorbidity in children with ID is large for disorders such as anxiety, depression, schizophrenia, epilepsy, obesity, chronic pain, ADHD, autism, and cardiovascular and muscular anomalies. These have to be taken into account in diagnosing the co-occurrence of ID and DCD. More specifically, when diagnosing DCD one should check for coexisting disorders and set priorities for treatment and support. In case of ID as a coexisting condition, the measured delay in motor development should not be attributable to a neurological condition affecting movement.

Assessment of Primary and Secondary Movement-Related Problems

Motor Competence

Motor competency should be measured by using an appropriate motor test that measures the different areas of motor competence, and have good reliability and validity and a population-based standardization (appropriately norm-referenced for that specific population). Motor performance tests recommended in the international CPR to diagnose DCD "Criterion A" are the Movement Assessment for Children—second edition (MABC2; Henderson, Sugden, & Barnett, 2007) and the Bruininks–Oseretsky test—second edition (BOT-2; Bruininks & Bruininks, 2005). Most of the tests have not been investigated for children with mild ID (IQ = 50–70) and may only be partly suitable. It follows that reference norms and cut-off points for children with ID are lacking.

A test for motor performance that has been used in children with ID is the Bruininks–Oseretsky test—second edition (BOT-2). The BOT-2 consists of eight subsets (fine motor skills, manual dexterity, bilateral coordination, balance, strength, upper limb coordination, running speed and agility) and is a reliable and valid measure in children with mild ID (Wuang & Su, 2009a, b).

The Test of Gross Motor Development-II (TGMD-2) is an assessment of locomotor function and object control that has been reported to be a reliable and valid measure in children with ID (Capio, Eguia, & Simons, 2016; Simons et al., 2008; Ulrich, 2000). Reference norms are available for the TGMD-2 for children with Down Syndrome. It is important to mention that validity of the BOT2 and the TGMD for these target groups was only investigated by group and age comparison and not compared with another measure.

The Developmental Test of Visual-Motor Integration (VMI) has also been adapted for children with ID and is a reliable and valid measure for identifying perceptual-fine motor deficits (Wuang & Su, 2009a, b).

To measure functional mobility, balance, and postural control, the timed up and go test (TUG) is a commonly used test in elderly, but it has also been investigated in children with ID. In the adapted version for children, children must stand up from a chair, walk as quickly as possible across a 3-meter distance, touch a target on a wall, then walk back and sit down on the chair. Research has shown that the TUG is valid and reliable tool for children with ID and there are norms available for TD children (Martin, Natarus, Martin, & Henderson, 2017; Nicolini-Panisson & Donadio, 2014).

The Gross Motor Function Measure-88 (GMFM-88) is a tool for motor function and consists of five domains: lying and rolling, sitting, crawling and kneeling, standing, walking–running–jumping. It was developed for children with cerebral palsy and adapted for children with Down Syndrome (DS). There is evidence to show that it is a reliable and valid tool for children with DS (Russel, Palisano, & Walter, 1998; Russel, Rosenbaum, Avery, & Lane, 2002).

To assess DCD criterion B, questionnaires are used to determine problems in daily living activities and school performance, (pre)vocational activities, leisure and play. The DCD-Q (Wilson, Crawford, Roberts, & Kaplan, 2006) is the most commonly used questionnaire for this purpose, but has not been investigated in children with mild ID. The Pediatric Evaluation Disability Inventory (PEDI) is a parent interview and consists of three domains (self-care, mobility, and social functioning) for which scale scores can be calculated. The PEDI is a reliable and valid tool and is able to discriminate between disabled children and TD children (Custers et al., 2003; Helders, 2003; Volman, Visser, & Lensvelt-Mulders, 2007).

Physical Fitness

The American College of Sports Medicine (2014) divides physical fitness into five different components: body composition, muscle strength, muscle endurance, cardiorespiratory fitness, and flexibility.

Body Composition

It is known that high BMI has a negative impact on physical fitness and motor abilities in children (Graf et al., 2004). Obesity in children with ID is frequently seen (O'Shea, O'Shea, Gibson, Leo, & Carty, 2018; Wang et al., 2018). For body composition, BMI and skinfold measures are reliable and valid measures in children with mild ID (Bandini, Fleming, Scampini, Gleason, & Must, 2013; MacDonncha, Watson, McSweeney, & O'Donovan, 1999; Tejero-Gonzalez et al., 2013). Waist circumference is also a reliable measure, but information concerning validity is lacking (Wouters, Evenhuis, & Hilgenkamp, 2017).

Muscle Strength and Muscle Endurance

Children with ID are known to have lower levels of muscle strength and muscle endurance (Hartman, Smith, Westendorp, & Visscher, 2015;

Hassani et al., 2014; Tejero-Gonzalez et al., 2013). In addition to the pathophysiological aspects (Dey et al., 2013), the lower level of physical activity and the fact that these children participate less in outdoor activities and sports (Barr & Shields, 2011) are seen as important factors for the lower level of muscle fitness. Few measures that evaluate muscle strength and muscle endurance in children with mild ID have been well investigated on psychometrics (Wouters et al., 2017). Grip strength, hand-held dynamometry (HHD), standing long jump and arm hang are seen as reliable muscle strength measures, but information concerning validity is lacking. The subset strength of the BOT-2 is reported to be a reliable and valid measure for children with ID (Wuang & Su, 2009a, b) and consists of five items for functional strength, mostly core muscles (long jump, sit-up, push up, wall sit and v-up). Similar tests considered valid and reliable in youth with and without ID are the 1 min sit-up and the isometric push-up protocol.

Also the Functional Strength Measurement for children with ID (FSM-ID) is a reliable and valid measure in this group of children and consist eight items measuring explosive power and muscle endurance in upper and lower extremities (overarm throwing, standing long jump, underarm throwing, lateral step up, chest pass, sit to stand, lifting a box and stair climbing) (Aertssen, Steenbergen, & Smits-Engelsman, 2018).

Cardiorespiratory Fitness

Children with ID have lower levels of cardiorespiratory fitness with lower VO2max and peak heart rate (Hartman et al., 2015; Oppewal, Hilgenkamp, van Wijck, & Evenhuis, 2013). The lower level of physical activity is mentioned as one of the explanations, but the pathophysiological features (e.g., chronotropic incompetence) also contribute to decreased cardiorespiratory fitness.

To measure cardiorespiratory fitness in children with mild ID several tools are available. Treadmill testing is a reliable and valid way to test cardiorespiratory fitness in children with mild ID (Oppewal et al., 2013). Additionally, field-based tests are available to estimate the cardiorespiratory fitness, measuring performance on a fixed running distance such as 300, 500, 880 yards and 1 mile run. These measures are seen as reliable and valid. The 6 min walk test measures cardiorespiratory fitness with a fixed time and measured distance, which is a reliable and valid measure in children with mild ID (Casey, Wang, & Osterling, 2012; Elmahgoub, Van de Velde, Peersman, Cambier, & Calders, 2012). Casey et al. (2012) found a learning effect after two practice trials in children with DS. Two types of shuttle run tests have been investigated in children with mild ID. The 16-meter Shuttle run and the 20-meter Shuttle Run and both are seen as reliable and valid measures (Agiovlasitis, Pitetti, Guerra, & Fernhall, 2011; Beets, Pitetti, & Fernhall, 2005; Fernhall et al., 1998).

(Hyper)flexibility

For flexibility, the Beighton score is a feasible and reliable measure in children with mild ID (Pitetti, Miller, & Beets, 2015). The Beighton test consists of five items (dorsal flexion of the fifth MCP, opposition of the thumb, hyperextension of elbow, hyperextension of the knees, hands flat on floor with straight knees).

Motor Fitness

The EUROFIT test battery is a worldwide used test battery to assess physical fitness in youth and adolescents. It consists of items for endurance, muscle strength, muscle endurance, speed, flexibility and balance. It was adapted for youth and adolescents with ID by removing or replacing items. The adapted version, EUROFIT-Special, consists of items for strength (long jump, pushing a medicine ball), muscle endurance (30 s sit up), speed (25 m run), flexibility (sit and reach test), and balance (balance walk) (Skowronski, Horvat, Nocera, Roswal, & Croce, 2009).

Physical Activity

Children and adolescents with mild ID are known to have lower levels of physical activity (PA) (Wouters, Evenhuis, & Hilgenkamp, 2018). Objective PA measurement may be helpful in providing a more accurate profile of PA levels in children with ID and in assessing effectiveness of intervention programs. To measure levels of physical activity questionnaires, pedometers and other accelerometers have been used. The Physical Activity Questionnaire for older children (PAQ-C) and the Physical Activity Questionnaire for adolescents (PAQ-A) are questionnaires regarding sports, games and physical activity during school time, and leisure time. These questionnaires have been used in different studies in children and adolescents with ID with findings of decreased levels of physical activity (Wyszyńska, Podgórska-Bednarz, Dereń, & Mazur, 2017). Although these questionnaires are used in children with ID, information regarding reliability and validity is lacking.

The pedometer measures the number of steps per day. It has also been used to investigate the level of physical activity in children with ID. Research has shown a lower amount of steps in children with Down Syndrome compared to the TD children (Alhusaini, Ali Al-Walah, Melam, & Buragadda, 2017; Beets & Pitetti, 2011). Also different kinds of accelerometers were used in children with ID (Izquierdo-Gomez et al., 2017; Pitchford, Adkins, Hasson, Hornyak, & Ulrich, 2018), but standardized protocol, cut-off points or information regarding reliability and validity in children with ID is lacking (Leung, Siebert, & Yun, 2017).

ID and Motor Performance

ID and Fundamental Motor Skills

Motor behavior of children with ID differs substantially from that of non-disabled children. It is evident that their motor ability develops relatively slowly and that motor milestones are achieved later (Peuschel, 1987; Ulrich, Ulrich, & Collier, 1992). Many children with intellectual disability have been reported to have deficiencies in fundamental motor skills and physical activity behaviors, which are essential requirements to lead healthy active lifestyles. The high rates of inactivity (Hinckson & Curtis, 2013) are likely to lead to obesity (Ells et al., 2006) and declining physical fitness levels (Wouters et al., 2018; Wyszyńska et al., 2017). New programs and interventions are being implemented to promote active behavior. Enriched environment can create the opportunity for motor learning given the capacity of the child. Aiming at optimal fundamental motor skills or movement literacy (the ability to plan and execute basic motor skills—walk, run, jump, catch, kick, and throw with agility, balance, and coordination) is important for ADL and participation in play and recreational activities.

Fundamental motor skills are related to physical activity (PA) (Cheng et al., 2016; Larouche, Boyer, Tremblay, & Longmuir, 2014). PA is related to physical fitness (Tsiros et al., 2014; Vasconcellos et al., 2014) and motor skills are likewise related to physical fitness (Carroll-Scott et al., 2013; Lai et al., 2014).

The relationship between motor proficiency and physical fitness requires consideration as children with motor deficiencies are less likely to participate in PA (Cairney, Rigoli, & Piek, 2013). It would appear that the reduced motor skills may be one of the characteristics of children with ID that early interventions should focus on.

ID and Physical Fitness

Low levels of motor development are associated with lower levels of physical activity. A systematic review of PA measures in children with ID was undertaken using the Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA) guidelines (Hinckson & Curtis, 2013). The review identified deficiencies on a number of valid and reliable tools used to quantify PA. Despite the differences in study design and methodological quality, there was agreement among studies that children with ID were significantly less active compared to children without ID.

Regular participation in PA enhances body composition (LeMura & Maziekas, 2002), bone health (Morris, Naughton, Gibbs, Carlson, & Wark, 1997), psychological health (Wilkinson, 1983), and promotes social engagement (Jobling, 2001). Key facilitators for PA participation are enjoyment and unstructured activities, social interactions, parental support and increasing level of independence. Such facilitators may have beneficial effects for PA participation in later life (Downs, Boddy, Knowles, Fairclough, & Stratton, 2013). However, children with ID may enjoy PA less, as they have less proficiency in balance and coordination and poor cardiovascular fitness compared to their TD peers (Heah, Case, McGuire, & Law, 2007) The barriers for participation include lack of knowledge and skills, fear, non-stimulating parental behavior, adverse attitudes to disability, scarce facilities, lack of transport, deficient programs and staff capacity, increased cost, functional limitations, unsafe environment, and lack of facilities or programs at close proximity to facilitate access (Stanish et al., 2016).

Environmental Factors

Environmental factors are those aspects that surround children and influence their food intake and physical activity, and include home, school, and community (Karnik, 2015; Sehm, Warschburger, & Abnorm, 2015). It is therefore important to identify environmental conditions that may be optimized to support development and participation of children with ID and promote a healthy life style, within the limits of parental and professional support. The environment should also be supportive so that associated problems are effectively managed. Where there is decreased energy demand (with increased motorized transport to school, etc.) and increased consumption of energy (such as snacks, fast food, and soft drinks), the setting is often referred to as the "obesogenic environment" (Shrivastava, Shrivastava, & Ramasamy, 2016) (p. 430). A recent systematic review supports the vital role of the microenvironment created at home, where the household setting is of significant importance in determining the risk of parental PA levels, eating habits and television viewing behavior. This would apply to all children (with DCD, ID, and TD children) associated with obesity (Kaushal & Rhodes, 2014).

Home Environment

Home is the primary environment for motor development and skill-acquisition for all children. Parents offer the child opportunities for learning and improving skills. The child learns by exploration, trial and error, imitation, feedback and instruction during play and ADL tasks. Parents are the first to note if their child's motor development seems different.

Role-modelling and parental support was the strongest predictor of PA levels in TD children and PA levels of children with ID were highest when parents participated in PA with their children, provided transport (to spaces for recreation, when neighborhoods were non-conducive to PA) or gave them encouragement and support (Fitzgerald, Fitzgerald, & Aherne, 2012). One of the identified barriers to participation was that parents did not value sport and PA for their children (Syrad et al., 2015). The children themselves cited barriers such as time constraints to complete their school homework, lack of physical skills and equipment, and the fact that PA made them sweat, while enjoyment and recognizing health benefits were positively associated with PA (Kubbayi, Jooste, Toriola, & Paul, 2014).

School Environment

Schools offer a stimulating environment for structured learning new motor skills such as writing and physical education (PE) for TD and ID children. Schools have facilities that differ from home, such as proper furniture, objects and instruments for PE and sports grounds, and playgrounds. The children should learn and improve not only by instruction and feedback from the teacher but also by interaction with and imitation of peers in the playground. This provides the extremely important chance to exercise and improve their fine and gross motor skills. Teachers will often note if a child's fine (writing or drawing) or gross motor (playground, PE) are awkward. Teachers play an important role negotiating extra help when the child has special needs.

Additionally, by implementing health and physical education (PE) in school-settings children become aware of the importance of PA and healthy food consumption (Heelan, Bartee, & Nihiser, 2015). These studies support the school environment as a model environment to teach children how to espouse and maintain a healthy and active lifestyle. However, the extent of the successful implementation between schools may vary and school category (with reference to socio-economic status and resources) was found to be significant in the prediction of childhood obesity (Centers for Disease Control, 2013). Emerging research has documented the associations between PA, physical education (PE) and nutrition programs, and academic performance (Muthuri, Wachira, Onywera, & Tremblay, 2014).

Several small-scale studies evaluated enhanced PE interventions or whole-of-school curriculum-based programs, with mixed results. In some cases PA was shown to increase (Minnaar, Grant, & Fletcher, 2016), whereas in other studies, PA and physical fitness remained largely unchanged (Tian, du Toit, & Toriola, 2015). There is a need to further explore the relationship between socioeconomic status (SES), BMI and motor skills, as this would assist in appropriate targeting of school-based interventions. Resources to promote healthy behaviors at schools have also been cited to positively influence implementation of interventions.

Socioeconomic Status

Living conditions and parental behavior affect children's opportunities for (motor) learning, activity and health. Family household factors including SES are associated with parental activity levels and weight status (de Villiers et al., 2012). Parental obesity is a principal risk factor for childhood obesity and poor motor skills. Children raised in socioeconomically disadvantaged conditions are at an increased risk of the development of obesity (Keane, Layte, Harrington, Kearney, & Perry, 2012) and this is attributed mainly to decreased PA and poor-quality diet (Igel, Romppel, Baar, Brähler, & Grande, 2016). In general, the increased accessibility of SES groups to energy-dense foods (including low-SES groups in high-income countries and high-SES groups in low-to-middle-income countries), carry a higher risk of obesity than their counterparts (Mchunu & Le Roux, 2010). More research is needed to understand why children with ID, even more so than TD children, experience such disparities in BMI status and to develop health promotion initiatives targeted at this population.

Role of Physical Activity Participation in ID and DCD

Physical activity is an essential component in the physical, mental, social, and cognitive development of children (Steyn, Nel, Labadarios, Maunder, & Kruger, 2014). Children with ID, being less active, do not engage in PA for the recommended PA duration (Draper et al., 2017). A large proportion of children with DCD also avoid PA because they are excluded by peers, frustrated because they do not master the necessary skills for participation.

The family environment plays an important role and may have significant impact on self-esteem, PA, self-efficacy and physical fitness. The barriers most frequently identified to the active participation in sports and physical recreation of children with disabilities are functional limitations, high costs, unsafe environment and the lack of nearby facilities or programs (Brown et al., 2016). Additional barriers to PA participation are perceived self-competence, time, societal sustenance from schools and communities, and family demographics (Nock et al., 2016).

Intervention and Support

Once the diagnosis DCD has been confirmed and treatment is indicated several options for intervention are available aimed at improving motor

Table 49.2 Main characteristics of task-oriented interventions (TOI)

Characteristics of task-oriented interventions (TOI)
1. Client oriented (meaningful for the client)
2. Goal oriented (aiming at activities and participation as described ICF-CY)
3. Task and context-specific (similar to the goal)
4. Active role of the client
5. Aiming at functionality not at normality (participation is the aim)
6. Active involvement of parent(s)/caretakers to enable transfer and maintenance

performance. For children with DCD without ID task-oriented intervention methods are supported by many review publications (see Table 49.2). Most of these evidence-based methods have the potential to be applied in the treatment of children with ID diagnosed with a motor delay that is larger than expected for their ID (DCD-ID). Such intervention plans should take into account not only the ID but also comorbidities since they have major consequences for the *instruction* (hearing deficit, severe refractive errors and comprehension), the ways to demonstrate and motivate (communication skills), and the *intensity* of exercise (heart conditions, obesity, cervical spine abnormalities, ligamentous laxity joint instability, hypotonia, increased risk of thyroid disease, type 2 diabetes, obesity, seizures). Although treatment programs, so called task-oriented intervention, for children with motor difficulties are available (Preston et al., 2017) it is important to be aware that these children constitute a very heterogeneous group. Hence tailored intervention in addition to and in conjunction with general stimulation for participation in PA is often needed.

Three main goals for intervention are important: (1) Movement literacy, which is the ability to plan and execute basic motor skills—run, jump, catch, kick and throw with agility, balance and coordination; (2) ADL and practical life skills; and (3) to create opportunities for physical activity to develop and maintain physical skills, self-confidence, socialization and prevent non-communicable diseases.

A recent example of a task-oriented intervention is neuromotor task training (NTT). NTT is based upon motor learning theory and an ecological approach. The first step in this method is the identification of those tasks and activities related to participation, which are of greatest concern to the child and his family—these are then the targets of treatment. NTT uses several motor teaching strategies such as implicit learning and external focus of attention suited for children with ID. By using motor teaching strategies therapists guide children through the different phases of motor skill learning by gradually changing task demands, which avoids failure and keeps the children motivated. Tasks and environmental constraints that impede successful task performance are identified and adapted. Task constraints refer to aspects of the task that restrain successful completion of a motor activity, such as when a child cannot catch a tennis ball or cannot close a shirt with very small buttons. Environmental constraints refer to aspects of the environment that impede performance, for instance when a child tries to tricycle on rough terrain. These task and environmental constraints are manipulated in the NTT intervention sessions to provide ample opportunity to practice and improve the deficient motor skills. In the early phase of learning, providing simple demonstration, exploration, and verbal instruction as to the intended outcome of the skill may be adequate to stimulate practice. Next, by repeating the task the child is provided with feedback (information about their performance from the therapist or from other external sources, from the success or failure of the attempt and how to adapt to the different contexts (action-perception coupling) so that they can improve performance on subsequent practice attempts. Techniques such as guided discovery—limiting the exploration space so they find the optimal solution sooner—are applied to promote efficient learning, to ensure development of the skill, and to motivate the child to persist with practice (Smits-Engelsman et al., 2018).

It is important to know that task-oriented training aims at training skills toward the levels and in actual context the task will be performed in later. This is of extra value in children with ID since transfer of skills to other contexts may be limited. Motor tasks in NTT are defined as movements or actions that are executed in order to achieve a goal. These tasks differ with respect to the number of actions needed to complete the task, as well as the degree to which the motor actions need to be integrated with events occurring in the environment. Many tasks require adapting task execution to either static or changing events in the environment. When the people or objects in the room are static, task execution involves integrating information about size, shape, and relative position to plan the movements/actions. When the people or objects are moving, motor planning also involves predicting how and where they will change position in the environment. Tasks, which comprise several separate subtasks, make additional demands on knowledge of the sequence, working memory, ability to persist and stay on task and completing the task within a reasonable/specified time frame. So the fact that a child with ID can walk or catch does not mean that the motor task will be performed adequately under more complicated circumstances, for instance in a game. Especially in children with ID, training the task in the actual context it will be performed in later is needed.

Once a skill is mastered to a certain extent it needs to be integrated into activities of daily living. In several studies, factors are mentioned that facilitate participation in physical activities related to personal aspects (e.g., social interaction, rewarded for participation) and environmental aspects (fun level of the activities, activity program meeting individual need, family support). Also barriers for participating in physical activities are described related to personal aspects (health issues, lack of motivation for physical activity, inadequate level of motor skills) and environmental aspects (financial aspects, limited options for physical activities, transportation, time limit parents, negative social aspect) (Alwhaibi & Aldugahishem, 2018; Bossink, van der Putten, & Vlaskamp, 2017). The importance of developing general motor skills in children with borderline and mild ID, especially with regard to ball skills might contribute positively to their sports participation (Westendorp, Houwen, & Hartman, 2011). Acquiring motor skills is also important in development due to the influence on social and emotional skills.

Stable and competent performance of general motor skills forms the basis for more complex motor skills control (Ulrich, Burghardt, Lloyd, Tiernan, & Hornyak, 2011). These skills are utilized by children when interacting with other children while playing and in other activities. Children with deficient motor skills are often ostracized or they may avoid play in school and neighborhood settings (Pienaar, van Reenen, & Weber, 2016). Play and recreational sports in children with and without disabilities are vital enablers in acquiring functional skills (Lee et al., 2015). The data from limited research studies suggest that deficiencies in the development of motor skills are more likely to be accountable for low PA levels, which are often demonstrated in later life, than is an inherent tendency toward physical inactivity (Lee et al., 2015). There may be a positive influence from early interventions to facilitate the development of competent functional motor control, which will in turn facilitate motor skills.

The physical activity habits of children with intellectual disabilities have been studied far less meticulously than those of typically developing children. There is evidence that children with disabilities are less active because they lack social skills and have inefficient motor skills to support physical activity (Eguia, Capio, & Simons, 2015; Westendorp et al., 2011). Interventions to improve this situation should target these areas. There is a scarcity of published literature on ways to improve physical activity in children with ID. Those studies that have been completed generally lack rigor with small sample sizes and short duration of intervention. Conducting research on children with ID is challenging, and it is recommended that large-scale, multi-site studies be conducted to support increased sample size and adequate study design. Including family support structures and teacher support with regards to education of both teachers and family on health and nutrition, and the inclusion of the benefits of exercise with the participation of families as motivation should be included in intervention programs.

Conclusion

Children with ID may have motor symptoms similar to those of children with DCD. The motor problems are likely to aggravate their lesser opportunities for participation and may lead to developmental and health issues due to inactivity. Gross motor skill proficiency is essential for engagement in sports and physical play and for the development of a healthy lifestyle. Their motor skills may be tested using the validated instruments for children with DCD, adapting the instruction and other cognitive demands if needed, but normative data for this population is lacking. There is real need to improve the motor skill deficiencies in children with ID; task-oriented interventions provide the best-researched opportunity to improve motor skills in DCD. It is recommended that intervention programs also include aspects to influence weight status in ID children. Existing intervention programs to improve selected motor skills and to promote physical activity and health may be adapted from evidence-based programs for children with motor problems. Future research should validate these.

References

Aertssen, W. F. M., Steenbergen, B., & Smits-Engelsman, B. C. M. (2018). The validity and reliability of the Functional Strength Measurement (FSM) in children with intellectual disabilities. *Journal of Intellectual Disability Research, 62*(8), 719–729.

Agiovlasitis, S., Pitetti, K. H., Guerra, M., & Fernhall, B. (2011). Prediction of VO2peak from the 20-m shuttle-run test in youth with Down syndrome. *Adapted Physical Activity Quarterly, 28*(2), 146–156.

Alhusaini, A. A., Ali Al-Walah, M., Melam, G. R., & Buragadda, S. (2017). Pedometer-determined physical activity levels of healthy children and children with Down's syndrome. *Somatosensory & Motor Research, 34*(4), 219–225.

Alwhaibi, R. M., & Aldugahishem, H. M. (2018). Factors affecting participation in physical activities in Saudi children with Down syndrome: mothers' perspectives. *Disability and Rehabilitation, 30*, 1–12.

American Psychiatric Association. (2013). *Diagnostic and statistical manual of mental disorders* (5th ed.). Washington, DC: American Psychiatric Association.

Bandini, L. G., Fleming, R. K., Scampini, R., Gleason, J., & Must, A. (2013). Is body mass index a useful measure of excess body fatness in adolescents and young adults with Down syndrome. *Journal of Intellectual Disability Research, 57*(11), 1050–1057.

Barnhart, R. C., Davenport, M. J., Epps, S. B., & Nordquist, V. M. (2003). Developmental coordination disorder. *Physical Therapy, 83*(8), 722–731.

Barr, M., & Shields, N. (2011). Identifying the barriers and facilitators to participation in physical activity for children with Down syndrome. *Journal of Intellectual Disability Research, 55*(11), 1020–1033.

Beets, M. W., & Pitetti, K. H. (2011). Using pedometers to measure moderate-to-vigorous physical activity for youth with an intellectual disability. *Disability and Health Journal, 4*(1), 46–51.

Beets, M. W., Pitetti, K. H., & Fernhall, B. (2005). Peak heart rates in youth with mental retardation: pacer vs. treadmill. *Pediatric Exercise Science, 17*(1), 51–61.

Blank, R., et al. (2019). International clinical practice recommendations on the definition, diagnosis, assessment, intervention and psycho-social aspects of Developmental Coordination Disorder (DCD). *Developmental Medicine and Child Neurology, 61*(3), 242–285.

Bossink, L., van der Putten, A. A., & Vlaskamp, C. (2017). Understanding low levels of physical activity in people with intellectual disabilities: A systematic review to identify barriers and facilitators. *Research in Developmental Disabilities, 68*, 95–110.

Brown, H., Atkin, A., Panter, J., Wong, G., Chinapaw, M., & Sluijs, E. (2016). Family-based interventions to increase physical activity in children: a systematic review, meta-analysis and realist synthesis. *Obesity Reviews, 17*(4), 345–360.

Bruininks, R. H., & Bruininks, B. D. (2005). *Bruininks-Oseretsky Test of Motor Proficiency* (2nd ed.). Minneapolis: Pearson Assessment.

Bult, M. K., Verschuren, O., Jongmans, M. J., Lindeman, E., & Ketelaar, M. (2011). What influences participation in leisure activities of children and youth with physical disabilities? A systematic review. *Research in Developmental Disabilities, 32*(5), 1521–1529.

Cairney, J., Rigoli, D., & Piek, J. (2013). Developmental coordination disorder and internalizing problems in children: The environmental stress hypothesis elaborated. *Developmental Review, 33*(3), 224–238.

Capio, C. M., Eguia, K. F., & Simons, J. (2016). Test of gross motor development-2 for Filipino children with intellectual disability: validity and reliability. *Journal of Sports Sciences, 34*(1), 10–17.

Carroll-Scott, A., Gilstad-Hayden, K., Rosenthal, L., Peters, S., McCaslin, C., Joyce, R., & Ickovics, J. (2013). Disentangling neighborhood contextual associations with child body mass index, diet, and physical activity: The role of built, socioeconomic, and social environments. *Social Science and Medicine, 95*, 106–114.

Casey, A. F., Wang, X., & Osterling, K. (2012). Test-retest reliability of the 6-minute walk test in individuals with Down syndrome. *Archives of Physical Medicine and Rehabilitation, 93*(11), 2068–2074.

Centers for Disease Control. (2013). Make a difference at your school. *Chronic Disease*, 31.

Cheng, J., East, P., Blanco, E., Kang Sim, E., Castillo, M., Lozoff, B., & Gahagan, S. (2016). Obesity leads to declines in motor skills across childhood. *Child: Care, Health and Development, 42*(3), 343–350.

Crone, E. A., & Ridderinkhof, K. R. (2011). The developing brain: from theory to neuroimaging and back. *Developmental Cognitive Neuroscience, 1*(2), 101–109.

Custers, J. W., van der Net, J., Hoijtink, H., Wassenberg-Severijnen, J. E., Vermeer, A., & Helders, P. J. (2003). Discriminative validity of the Dutch Pediatric Evaluation of Disability Inventory. *Archives of Physical Medicine and Rehabilitation, 83*(10), 1437–1441.

de Villiers, A., Steyn, N., Draper, C., Fourie, J., Barkhuizen, G., Lombard, C., … Lambert, E. (2012). "HealthKick": Formative assessment of the health environment in low-resource primary schools in the Western Cape Province of South Africa. *BMC Public Health, 12*(1), 794.

Dey, A., Bhowmik, K., Chatterjee, A., Chakrabarty, P. B., Sinha, S., & Mukhopadhyay, K. (2013). Down Syndrome Related Muscle Hypotonia: Association with COL6A3 Functional SNP rs2270669. *Frontiers in Genetics, 4*, 57.

Downs, S., Boddy, L., Knowles, Z., Fairclough, S., & Stratton, G. (2013). Exploring opportunities available and perceived barriers to physical activity engagement in children and young people with Down syndrome. *European Journal of Special Needs Education, 28*(3), 270–287.

Draper CE, Tomaz SA, Stone M, Hinkley T, Jones RA, Louw J, Twine R, Kahn K, SA. N. (2017) Developing intervention strategies to optimise body composition in early childhood in South Africa. Biomedical Research International 2017: 5283457.

Eguia, K., Capio, C., & Simons, J. (2015). Object control skills influence the physical activity of children with intellectual disability in a developing country: The Philippines. *Journal of Intellectual and Developmental Disability, 40*(3), 265–274.

Ells, L. J., Lang, R., Shield, J. P., Wilkinson, J. R., Lidstone, J. S., Coulton, S., & Summerbell, C. D. (2006). Obesity and disability—A short review. *Obesity Reviews, 7*(4), 341–345.

Elmahgoub, S. S., Van de Velde, A., Peersman, W., Cambier, D., & Calders, P. (2012). Reproducibility, validity and predictors of six-minute walk test inoverweight and obese adolescents with intellectual disability. *Disability and Rehabilitation, 34*(10), 846–851.

Fernhall, B., Pitetti, K. H., Vukovich, M. D., Stubbs, N., Hensen, T., & Winnick, J. P. (1998). Validation of cardiovascular fitness field tests in children with mental retardation. *American Journal of Mental Retardation, 102*, 602–612.

Fitzgerald, A., Fitzgerald, N., & Aherne, C. (2012). Do peers matter? A review of peer and/or friends' influence on physical activity among American adolescents. *Journal of Adolescence, 35*(4), 941–958.

Geuze, R. H. (2007). Characteristics of DCD: on problems and diagnosis). In R. H. Geuze (Ed.), *Developmental Coordination Disorder. A review of current approaches* (pp. 9–25). Marseille: Solal Éditeurs.

Graf, C., Koch, B., Kratschmann-Kandel, E., Falkowski, G., Christ, H., & Coburger, S. (2004). Correlation between BMI, leisure habits and motor abilities in childhood (CHILT-project). *International Journal of Obesity and Related Metabolic Disorders, 28*, 22–26.

Hartman, E., Smith, J., Westendorp, M., & Visscher, C. (2015). Development of physical fitness in children with intellectual disabilities. *Journal of Intellectual Disability Research, 59*(5), 439–449.

Hassani, A., Kotzamanidou, M. C., Tsimaras, V., Lazaridis, S., kotzamanidis, C., & Patikas, D. (2014). Differences in counter-movement jump between boys and girls with and without intellectual disability. *Research in Developmental Disabilities, 35*(7), 1433–1438.

Heah, T., Case, T., McGuire, B., & Law, M. (2007). Successful participation: The lived experience among children with disabilities. *Canadian Journal of Occupational Therapy, 74*(1), 38–47.

Heelan, K. A., Bartee, R. T., & Nihiser, A. (2015). Healthier school environment leads to decreases in childhood obesity: the Kearney Nebraska story. *Childhood Obesity, 11*(5), 600–607.

Helders, P. J. (2003). Reliability of the Dutch Pediatric Evaluation of Disability Inventory (PEDI). *Clinical Rehabilitation, 17*(4), 457–462.

Hellgren, L., Gillberg, C., Gillberg, I. C., & Enerskog, I. (1993). Children with deficits in attention, motor control and perception (DAMP) almost grown up: general health at 16 years. *Developmental Medicine & Child Neurology, 35*(10), 881–892.

Henderson, S. E., & Henderson, L. (2002). Toward an understanding of developmental coordination disorder. *Adapted Physical Activity Quaterly, 19*(1), 11–31.

Henderson, S. E., Sugden, D. A., & Barnett, A. L. (2007). *Movement assessment battery for children-2 [Movement ABC-2]* (2nd ed.). London: The Psychological Corporation.

Hinckson, E., & Curtis, A. (2013). Measuring physical activity in children and youth living with intellectual disabilities: A systematic review. *Research in Developmental Disabilities, 34*(1), 72–86.

Igel, U., Romppel, M., Baar, J., Brähler, E., & Grande, G. (2016). The association between area-level socioeconomic status and childhood overweight and the role of urbanicity. *Obesity Medicine, 2*, 13–18.

Izquierdo-Gomez, R., Martinez-Gómez, D., Esteban-Cornejo, I., Hallal, P. C., García-Cervantes, L., Villagra, A., & Veiga, O. L. (2017). Changes in objectively measured physical activity in adolescents with Down syndrome: the UP&DOWN longitudinal study. *Journal of Intellectual Disability Research, 61*(4), 363–372.

Jobling, A. (2001). Life be in it: Lifestyle choices for active leisure. *Down Syndrome Research and Practice, 6*(3), 117–122.

Karnik S, A. K. (2015) Childhood obesity: A global public health crisis. International Journal of Preventative Medicine, 3(1), 1–7.

Kaushal, N., & Rhodes, R. (2014). The home physical environment and its relationship with physical activity and sedentary behavior: A systematic review. *Preventive Medicine, 67*, 221–237.

Keane, E., Layte, R., Harrington, J., Kearney, P., & Perry, I. (2012). Measured parental weight status and familial socio-economic status correlates with childhood overweight and obesity at age 9. *PLoS One, 7*(8), e43503.

Kubbayi, N., Jooste, J., Toriola, A., & Paul, Y. (2014). Familial and peer influences on sport participation among adolescents in rural South African secondary schools. *Mediterranean Journal of Social Sciences, 5*(20), 1305–1308.

Lai, S., Costigan, S., Morgan, P., Lubans, D., Stodden, D., Salmon, J., & Barnett, L. (2014). Do school-based interventions focusing on physical activity, fitness, or fundamental movement skill competency produce a sustained impact in these outcomes in children and adolescents? A systematic review of follow-up studies. *Sports Medicine, 44*(1), 67–79.

Larouche, R., Boyer, C., Tremblay, M., & Longmuir, P. (2014). Physical fitness, motor skill, and physical activity relationships in grade 4 to 6 children. *Applied Physiology, Nutrition, and Metabolism, 39*(5), 553–559.

Lee, H., Tamminen, K., Clark, A., Slater, L., Spence, J., & Holt, N. (2015). A meta-study of qualitative research examining determinants of children's independent active free play. *International Journal of Behavioral Nutrition and Physical Activity, 12*(1), 5.

LeMura, L., & Maziekas, M. (2002). Factors that alter body fat, body mass, and fat-free mass in pediatric obesity. *Medicine and Science in Sports and Exercise, 34*(3), 487–496.

Leung, W., Siebert, E. A., & Yun, J. (2017). Measuring physical activity with accelerometers for individuals with intellectual disability: A systematic review. *Research in Developmental Disabilities, 67*, 60–70.

Lin, H. C., & Wuang, Y. P. (2012). Strength and agility training in adolescents with Down syndrome: a randomized controlled trial. *Research in Developmental Disabilities, 33*(6), 2236–2244.

Loh, P. R., Piek, J. P., & Barrett, N. C. (2011). Comorbid ADHD and DCD: Examining cognitive functions using the WISC-IV. *Research in Developmental Disabilities, 32*, 1260–1269.

MacDonncha, C., Watson, A. W. S., McSweeney, T., & O'Donovan, D. J. (1999). Reliability of Eurofit physical fitness items for adolescent males with and without mental retardation. *Adapted Physical Activity Quarterly, 16*(1), 86–95.

Mandich, A. D., Polatajko, H. J., & Rodger, S. (2003). Rites of passage: understanding participation of children with developmental coordination disorder. *Human Movement Science, 22*(4–5), 583–595.

Martin, K., Natarus, M., Martin, J., & Henderson, S. (2017). Minimal detectable change for TUG and TUDS tests for children with down syndrome. *Pediatric Physical Therapy, 29*(1), 77–82.

Martin, N. C., Piek, J. P., & Hay, D. (2006). DCD and ADHD: A genetic study of their shared aetiology. *Human Movement Studies, 25*, 110–124.

Mchunu, S., & Le Roux, K. (2010). Non-participation in sport by Black learners with special reference to gender, grades, family income and home environment. *The South African Journal for Research in Sport, Physical Education and Recreation, 32*(1), 85–98.

Minnaar, E., Grant, C., & Fletcher, L. (2016). Physical activity of children from a small rural town, South Africa. *Journal of South African Family Practice, 58*(2), 68–73.

Morris, F., Naughton, G., Gibbs, J., Carlson, J., & Wark, J. (1997). Prospective ten-month exercise intervention in pre-menarcheal girls: positive effects on bone and lean mass. *Journal of Bone and Mineral Research, 12*(9), 1453–1462.

Muthuri, S., Wachira, L.-J., Onywera, V., & Tremblay, M. (2014). Correlates of objectively measured overweight/obesity and physical activity in Kenyan school children: results from ISCOLE-Kenya. *BMC Public Health, 14*(1), 1.

Nicolini-Panisson, R. D., & Donadio, M. V. (2014). Normative values for the Timed 'Up and Go' test in children and adolescents and validation for individuals with Down syndrome. *Developmental Medicine & Child Neurology, 56*(5), 490–497.

Nock, N., Ievers-Landis, C., Dajani, R., Knight, D., Rigda, A., Narasimhan, S., & Uli, N. (2016). Physical activity, self-efficacy and fitness: Family environment relationship correlates and self-esteem as a mediator among adolescents who are overweight or obese. *Childhood Obesity, 12*(5), 360–367.

O'Shea, M., O'Shea, C., Gibson, L., Leo, J., & Carty, C. (2018). The prevalence of obesity in children and young people with Down syndrome. *Journal of Applied Research in Intellectual Disabilities, 31*(6), 1225–1229.

Oppewal, A., Hilgenkamp, T. I., van Wijck, R., & Evenhuis, H. M. (2013). Cardiorespiratory fitness in individuals with intellectual disabilities—A review. *Research in Developmental Disabilities, 34*(10), 3301–3316.

Peuschel, S. M. (1987). Clinical aspects of Down syndrome from infancy to adulthood. *American Journal of Medical Genetics*. Suppl*, 7*, 52–56.

Philips, A. C., & Holland, A. J. (2011). Assessment of objectively measured physical activity levels in individuals with intellectual disabilities with and without Down's syndrome. *PLoS One, 6*(12), e28618.

Pienaar, A., van Reenen, I., & Weber, A. (2016). Sex differences in fundamental movement skills of a selected group of 6-year-old South African children. *Early Child Development and Care, 186*(12), 1–15.

Pieters, S., De Block, K., Scheiris, J., Eyssen, M., Desoete, A., Deboutte, D., … Roeyers, H. (2012). How common are motor problems in children with adevelopmental disorder: rule or exception. *Child Care Health Development, 38*(1), 139–145.

Pitchford, E. A., Adkins, C., Hasson, R. E., Hornyak, J. E., & Ulrich, D. A. (2018). Association between physical activity and adiposity in adolescents with Down Syndrome. *Medicine & Science in Sports & Exercise, 50*(4), 667–674.

Pitetti, K., Miller, R. A., & Beets, M. W. (2015). Measuring joint hypermobility using the Beighton scale in children with intellectual disability. *Pediatric Physical Therapy, 27*(2), 143–150.

Preston, N., Magallon, S., Hill, L. J., Andrews, E., Ahern, S. M., & Mon-Williams, M. (2017). A systematic review of high quality randomized controlled trials investigating motor skill programmes for children with developmental coordination disorder. *Clinical Rehabilitation, 31*(7), 857–870.

Rasmussen, P., & Gillberg, C. (2000). Natural outcome of ADHD with developmental coordination disorder at age 22 years: a controlled, longitudinal, community-based study. *American Academy of Child and Adolescent Psychiatry, 39*(11), 1424–1431.

Russel, D., Palisano, R., & Walter, S. (1998). Evaluating motor function in children with Down syndrome: validity of GMFM. *Developmental Medicine & Child Neurology, 40*, 693–701.

Russel, D. J., Rosenbaum, P. L., Avery, L. M., & Lane, M. (2002). *Gross Motor Function Measure (GMFM-6 & GMFM-88) user's manual*. London: Mac Keith Press.

Sehm, M., Warschburger, P., & Abnorm, P. (2015). The specificity of psychological factors associated with binge eating in adolescent boys and girls. *Journal of Abnormal Child Psychology, 43*(8), 1563–1571.

Shrivastava, S., Shrivastava, P., & Ramasamy, J. (2016). Neutralizing the contribution of obesogenic environment in the development of childhood obesity. *Annals of Tropical Medicine and Public Health, 9*(6), 430–431.

Simons, J., Daly, D., Theodorou, F., Caron, C., Simons, J., & Andoniadou, E. (2008). Validity and reliability of the TGMD-2 in 7-10-year-old Flemish children with intellectual disability. *Adapted Physical Activity Quarterly, 25*(1), 71–82.

Skowronski, W., Horvat, M., Nocera, J., Roswal, G., & Croce, R. (2009). Eurofit special: European fitness battery score variation among individuals with intellectual disabilities. *Adapted Physical Activity Quarterly, 26*(1), 54–67.

Smits-Engelsman, B., & Hill, E. L. (2012). The relationship between motor coordination and intelligence across the IQ range. *Pediatrics, 130*(4), e950-6.

Smits-Engelsman, B., Schoemaker, M., Delabastita, T., Hoskens, J., & Geuze, R. (2015). Diagnostic criteria for DCD: Past and future. *Human Movement Science, 42*, 293–306.

Smits-Engelsman, B., Vinçon, S., Blank, R., Quadrado, V. H., Polatajko, H., & Wilson, P. H. (2018). Evaluating the evidence for motor-based interventions in developmental coordination disorder: A systematic review and meta-analysis. *Research in Developmental Disabilities, 74*, 72–102.

Smits-Engelsman, B. C., Magalhães, L. C., Oliveira, M. A., & Wilson, P. H. (2015). DCD research: How are we moving along? *Human Movement Science, 42*, 289–292.

Stanish, H., Curtin, C., Must, A., Phillips, S., Maslin, M., & Bandini, L. (2016). Physical activity enjoyment, perceived barriers and beliefs among adolescents with and without intellectual disabilities. *Journal of Physical Activity and Health, 13*(1), 102–110.

Steyn, N., Nel, J., Labadarios, D., Maunder, E., & Kruger, H. (2014). Which dietary diversity indicator is best to assess micronutrient adequacy in children 1 to 9 y. *Nutrition, 30*(1), 55–60.

Syrad, H., Falconer, C., Cooke, L., Saxena, S., Kessel, A., Viner, R., … Croker, H. (2015). 'Health and happiness is more important than weight': A qualitative investigation of the views of parents receiving written feedback on their child's weight as part of the National Child Measurement Programme. *Journal of Human Nutrition and Dietetics, 28*(1), 47–55.

Tejero-Gonzalez, C. M., Martinez-Gomez, D., Bayon-Serna, J., Izquierdo-Gomez, R., Castro-Pinero, J., & Veiga, O. L. (2013). Reliability of the alpha health-related fitness test battery in adolescents with Down syndrome. *Journal of Strength and Conditioning Research, 27*(11), 3221–3224.

Tian, H., du Toit, D., & Toriola, A. (2015). The effects of an enhanced quality Physical Education programme on the physical activity levels of Grade 7 learners in Potchefstroom, South Africa. *Physical Education and Sport Pedagogy, 2015*, 1–16.

Tsiros, M., Coates, A., Howe, P., Walkley, J., Hills, A., Wood, R., & Buckley, J. (2014). Are obese children really less fit? Influences of body composition and physical activity on cardiorespiratory fitness in obese and healthy-weight children. *Obesity Research and Clinical Practice, 8*, 106.

Ulrich, B. D., Ulrich, D. A., & Collier, D. H. (1992). Alternating stepping patterns: hidden abilities of 11-month-old infants with Down syndrome. *Developmental Medicine & Child Neurology, 34*(3), 233–239.

Ulrich, D., Burghardt, A., Lloyd, M., Tiernan, C., & Hornyak, J. (2011). Physical activity benefits of learning to ride a two-wheel bicycle for children with Down syndrome: A randomized trial. *Physical Therapy, 91*(10), 1463–1477.

Ulrich, D. A. (2000). *Test of Gross Motor Development*. Austin, TX: Pro-ed Publishers.

van Galen, G. P., & Morasso, P. G. (1998). Neuromotor control in handwriting and drawing: introduction and overview. *Acta Psychologica (Amst), 100*(1–2), 1–7.

Vasconcellos, F., Seabra, A., Katzmarzyk, P., Kraemer-Aguiar, L., Bouskela, E., & Farinatti, P. (2014). Physical activity in overweight and obese adolescents: Systematic review of the effects on physical fitness components and cardiovascular risk factors. *Sports Medicine, 44*(8), 1139–1152.

Volman, M. J., Visser, J. J., & Lensvelt-Mulders, G. J. (2007). Functional status in 5 to 7-year-old children with Down syndrome in relation to motor ability and performance mental ability. *Disability and Rehabilitation, 29*(1), 25–31.

Wang, J., Gao, Y., Kwok, H. H. M., Huang, W. Y. J., Li, S., & Li, L. (2018). Children with intellectual disability are vulnerable to overweight and obesity: A cross-sectional study among Chinese children. *Childhood Obesity, 14*(5), 316–326.

Westendorp, M., Houwen, S., & Hartman, E. (2011). C. V: Are gross motor skills and sports participation related in children with intellectual disabilities? *Research in Developmental Disabilities, 32*(3), 1147–1153.

Wilkinson, P. (1983). Disabled children and integrated play environments. *Recreation Research Review, 10*(1), 20–28.

Wilson, B. N., Crawford, S. G., Roberts, G., & Kaplan, B. J. (2006). *Further validation and revision of the developmental coordination disorder questionnaire*. Calgary, AB: University of Calgary and Calgary Health Region.

Wilson, P. H., Smits-Engelsman, B., Caeyenberghs, K., Steenbergen, B., Sugden, D., ClarkJ, M. N., & Blank, R. (2017). Cognitive and neuroimaging findings in developmental coordination disorder: new insights from a systematic review of recent research. *Developmental Medicine & Child Neurology, 59*(11), 1117–1129.

Wouters, M., Evenhuis, H. M., & Hilgenkamp, T. I. (2017). Systematic review of field-based physical fitness tests for children and adolescents with intellectual disabilities. *Research in Developmental Disabilities, 61*, 77–94.

Wouters M, Evenhuis HM, Hilgenkamp TI (2018) Physical activity levels of children and adolescents with moderate-to-severe intellectual disabilities. Journal of Applied Research in Intellectual Disabilities 2017: 1–12.

Wuang, Y. P., & Su, C. Y. (2009a). Rasch analysis of the developmental test of visual-motor integration in children with intellectual disabilities. *Research in Developmental Disabilities, 30*(5), 1044–1053.

Wuang, Y. P., & Su, C. Y. (2009b). Reliability and responsiveness of the Bruininks-Oseretsky Test of Motor Proficiency-Second Edition in children with intellectual disability. *Research in Developmental Disabilities, 30*(5), 847–855.

Wyszyńska, J., Podgórska-Bednarz, J., Dereń, K., & Mazur, A. (2017). The relationship between physical activity and screen time with the risk of hypertension in children and adolescents with intellectual disability. *BioMed Research International, 2017*, 8.

Substance Use Disorders

50

Joanne E. L. VanDerNagel, Neomi van Duijvenbode,
and Robert Didden

Introduction

Substance use disorders (SUD) are among the most common mental disorders in the adult population (Substance Abuse and Mental Health Services Administration [SAMHSA], 2017). Results from a national survey in the USA indicate that 7.5% of the people aged 12 and older have had an SUD in the past year. Individuals with intellectual disabilities (ID) are considered at risk for developing SUD. In this first paragraph, we discuss the core features of SUD, and its diagnostic classification according to the DSM-5. In the remainder of this chapter, we discuss the prevalence of SUD among individuals with ID, its risk factors, and methods for screening, assessment, and treatment of SUD among individuals with ID.

Substance Use and Substance Use Disorders

Substances are being used by many individuals and they are part of their daily lives. Indeed, according to a national survey in the USA (SAMHSA, 2017), in 2016 28.6 million people (10.6%) aged 12 or older used an illicit drug in the past 30 days, 136.7 million used alcohol (50.7%), and 51.3 million used tobacco (19%). For some, the use of substances becomes problematic and they develop an SUD. Results from the same national survey indicate that there were 15.1 million individuals with an alcohol use disorder and 7.4 million individuals with an illicit drug use disorder (SAMHSA, 2017).

According to the American Psychiatric Association (2013), a substance use disorder (SUD) is characterized by a persistent desire to use and the inability to cut down or control substance use, even in the face of severe negative consequences (i.e., physical, psychological, social, or interpersonal problems). This "paradox of addiction" is explained by disruptions in

---

J. E. L. VanDerNagel (✉)
Tactus Addition Institute, Deventer, The Netherlands

Radboud University Nijmegen, Nijmegen Institute for Scientist-Practitioners in Addiction, Nijmegen, The Netherlands

Aveleijn, Borne, The Netherlands
e-mail: j.vandernagel@tactus.nl

N. van Duijvenbode
Tactus Addition Institute, Deventer, The Netherlands

Radboud University Nijmegen, Nijmegen Institute for Scientist-Practitioners in Addiction, Nijmegen, The Netherlands

Radboud University Nijmegen, Behavioural Science Institute, Nijmegen, The Netherlands
e-mail: n.vanduijvenbode@tactus.nl

R. Didden
Radboud University Nijmegen, Behavioural Science Institute, Nijmegen, The Netherlands

Trajectum, Zwolle, The Netherlands
e-mail: r.didden@pwo.ru.nl

J. L. Matson (ed.), *Handbook of Intellectual Disabilities*, Autism and Child Psychopathology Series, https://doi.org/10.1007/978-3-030-20843-1_50

motivational, reward, and inhibitory control brain regions. These disruptions result in the over-evaluation of substance use-related stimuli, and reduction of inhibitory control. In other words, substance use-related stimuli “grab” the attention, seem attractive, and elicit approach behavior. At the same time, executive functions such as working memory capacity, the ability to delay gratification and behavioral control is deteriorating. Behavior of individuals with SUD is therefore increasingly influenced by automatic processes, indicating a growing loss of control over substance use (Koob, 2013; Volkow, Wang, Tomasi, & Baler, 2013).

In the Diagnostic and Statistical Manual of mental disorders, fifth edition (DSM-5; American Psychiatric Association [APA], 2013), an SUD is assessed on a continuum from mild to severe, using 11 criteria related to (1) impaired control over quantity and duration of substance use (criteria 1–4); (2) social impairment due to substance use (criteria 5–7); (3) risky substance use (criteria 8–9); and (4) tolerance and withdrawal (criteria 10–11). SUD severity is mild when two to three symptoms are met, moderate if four to five symptoms are met, or severe if six or more symptoms are met. It should be noted that severity can change over time. The DSM-5 identifies 10 classes of substances that are commonly found to have addictive potential for a large number of individuals of the general population. These substances are alcohol; caffeine; cannabis; hallucinogens (such as LSD); inhalants; opioids; sedatives, hypnotics, and anxiolytics; stimulants (e.g., amphetamine and cocaine); tobacco; and other (or unknown) substances. Although all of these types have common effects on the brain systems mentioned above, they may differ in other effects (see Box 50.1).

Box 50.1: Types of Substances

There are different methods to classify substances into groups. These methods coexist and serve different purposes, such as diagnosis, legal purposes, and clinical use.

A first distinction is legal versus illegal, or scheduled/non-scheduled. Within the latter system, schedule I drugs are regarded as the most harmful, with high abuse potential and no registered medical use. These drugs include for instance heroin, cocaine, and LSD. Schedule II drugs also have a high abuse and dependency potential, but with medical uses. These drugs include opioids such as methadone and morphine but also methamphetamine. Schedule III–V drugs mainly have medical use, with lower risks for abuse or dependency. Alcohol and tobacco—despite their highly addictive qualities and severe medical, mental, and social consequences—are not classified within this system, which is a downside to this classification system.

A second distinction can be made based to the psychoactive effects. Substances can be stimulating, depressant, hallucinogenic, or a combination of these. Stimulants (“uppers”) increase the activity of the central nervous system and body, and are widely used (prescribed or non-prescribed, legal and illicit) as performance enhancing, mood elevating drugs. Stimulants include substances such as caffeine and tobacco but also methylphenidate, amphetamines, and cocaine. When overdosed, these substances cause symptoms of physical and mental overstimulation, such as restlessness, sleeplessness, tachycardia, hypertension, muscle spasms, irritability, and psychosis. Depressants (“downers”) are substances that reduce arousal and activity, and cause anxiolysis, sedation, pain relief, muscle relaxation, but also somnolence, cognitive impairment, and complete anesthesia or even coma. These substances include alcohol but also opioids and sedatives/anxiolytics. Hallucinogens can cause hallucinations and other changes in cognition. A classic example of hallucinogens is LSD. Cannabis has both hallucinogenic and depressant effects. The distinction between stimulant, depressant, and hallucinogenic properties provides the most help for clinical management during intoxication or withdrawal, even when the exact type of substance that was used remains unclear.

Substance Use (Disorder) in Individuals with Intellectual Disabilities

Prevalence of SUD in Individuals with ID

In 2003, based on the limited number of studies at that time, Sturmey, Reyer, Lee, and Robek (2003) estimated the overall prevalence rate of SUD to be around 0.5–2% of the total population of individuals with ID. In 2010, using a large sample of Medicaid data, Slayter (2010a) found that individuals with ID and SUD constituted 2.6% of all people with ID. Also, using data from the Health Care Access Research and Developmental Disabilities (H-CARDD) cohort, Lin et al. (2016) found 6.4% SUD in adults with ID. It should be mentioned that rates of SUD vary between samples, with higher prevalence rates in populations with psychiatric comorbidity (e.g., Didden, Embregts, Van der Toorn, & Laarhoven, 2009; Plant, McDermott, Chester, & Alexander, 2011) and forensic populations (e.g., Lindsay et al., 2013). As expected, the prevalence rates among individuals with mild ID are much higher compared to those with more severe ID (e.g., Slayter, 2010a). However, given the scarcity of large-scale studies, it remains unclear what "the" prevalence of SUD is in individuals with ID. Given the variety in methodology, results from different studies are difficult if not impossible to compare. At present, we therefore are unable to substantiate (or falsify) the observation that the prevalence of SUD in individuals with ID is rising.

Despite a scarcity of studies, it now is recognized that individuals with ID do use substances, that this substance use (SU) can lead to SUD and that this warrants attention from clinicians and researchers. Several European studies showed that alcohol is the main substance used in adults and adolescents with ID, although prevalence rates are generally lower compared to those in the general population (Emerson & Turnbull, 2005; Pacoricona Alfaro et al., 2017; Reis, Wetzel, & Haßler, 2017; To, Neirynck, Vanderplasschen, Vanheule, & Vandevelde, 2014; VanDerNagel et al., 2017; VanDerNagel, Kiewik, Buitelaar, & De Jong, 2011). Smoking rates, however, are higher in adults and adolecents with ID than those in the general population (Emerson & Turnbull, 2005; Pacoricona Alfaro et al., 2017; Swerts et al., 2017; VanDerNagel et al., 2017). Regarding illicit drug use, studies provide conflicting evidence, with lower prevalence in some studies (e.g., Swerts et al., 2017) and higher prevalence in others (e.g., Pacoricona Alfaro et al., 2017; VanDerNagel et al., 2017). Overall, it can be concluded that substances are used by many individuals with ID, especially those with mild ID and those with comorbid problems.

Prevalence of ID Within Addiction Services

What is even much less known is the prevalence rate of individuals with ID within addiction services. In a survey among staff members, 5.7% of the patients in a Dutch addiction facility were identified as having a mild to borderline intellectual functioning (MID-BIF, IQ 50-85). The authors comment that this is probably an underestimation of the true prevalence of individuals with MID-BIF in SUD samples (VanDerNagel et al., 2014). Based on client related measurements, using the Wechsler Adult Intelligence Scale III (WAIS-III; Wechsler, 2004), To, Vanheule, Vanderplasschen, Audenaert, and Vandevelde (2015) found that the average IQ in a sample of 90 clients with SUD was 89, with 11 clients (12%) having a total IQ score below 70. In another study, an almost identical average IQ (88) was found in a sample of forensic addiction inpatients, with 39% of the sample being identified as having MID-BIF (Luteijn, Didden, & VanDerNagel, 2017).

Challenges in Prevalence Estimations

As illustrated above, it is challenging to provide reliable SU and SUD prevalence rates in individuals with ID (also see Van Duijvenbode et al., 2015). Differences between samples (e.g., community based, institutionalized), subgroups (e.g., forensic, psychiatric), definitions of SU(D), and methodological and measurement issues will result in different rates of SU(D). In addition, the rates may vary between countries, cultures, and regions. Another explanation is that the majority of individuals with mild ID—one of the groups at risk for SU and SUD—is generally housed within the community and may not be known to ID services. Some of these individuals may be known to addiction services, even without being identified as a person with an ID. In fact, adaptive skills deficits in this group may be (wrongfully) ascribed to the SUD instead of to the ID. Last but not least, SU and SUD are under recognized. Both client factors (such as reluctance to admit having a SU related problem), and non-client factors (preconceptions about individuals with ID not using substances, lack of screening) contribute to this lack of timely case identification. Thus, representative and large-scale studies on SU(D) in ID are hindered by practical and methodological challenges. Those identified with SUD are probably the "tip of the iceberg" and may not be representative for the total population (Van Duijvenbode et al., 2015).

Risk Factors for SUD

In addiction medicine, SUD is regarded as a complex and multifaceted brain disorder, with biological, psychological and social risk- and protective factors. These factors and their relationships to SUD have been mostly investigated in the general population. However, these is no reason to assume that the relationships and mechanisms do not apply to individuals with ID. In fact, social and psychological risk factors for SU(D) seem to accumulate in individuals with ID and which may put them at increased risk for SUD (see for example Van Duijvenbode et al., 2015). For example, general risk factors for SUD such as adverse life events, inadequate coping strategies, social isolation, and a low socioeconomical status are often more prevalent in individuals with ID.

In addition to these general risk factors, factors related to the ID, such as lack of basic substance (risk) knowledge and insight in its consequences, lack of delay gratification ability, the desire to "fit in," increased susceptibility to peer pressure, and low self-esteem, may further contribute to SU initiation and the development of SUD in individuals with ID. For instance, individuals with ID are reported to misinterpret the rather "liberal" cannabis policy in the Netherlands and (wrongly) conclude that cannabis use is both legal and safe (VanDerNagel et al., 2017). The relatively high prevalence of co-occurring use of prescribed psychotropic medication and psychiatric disorders further increase this risk. Also, a lack of awareness of SU risks, and recognition of early signs and symptoms of SU by caregivers, as well as barriers in access to preventative and treatment services, may further contribute to the progression of SU into SUD (e.g., Slayter, 2010b; VanDerNagel, Kemna, & Didden, 2013).

Screening and Assessment

Issues in Screening and Assessment

Diagnosis of SUD in individuals with ID is not so much hindered by the complexity of the disorder, but more by the lack of reliable information regarding quantity and duration of SU and its consequences (Mikkelsen, VanDerNagel, & Lindsay, 2016). Both individuals with and without ID may be inclined to underreport their SU, because of fear of stigma and consequences. In individuals with ID, the lack of knowledge about SU risks, including misinterpretation of what can be regarded as safe SU practices, as well as their dependence on others who may judge their SU taking behavior, may contribute to their failure to be forthcoming.

In addition, signs and symptoms of SUD can remain undetected by caregivers for prolonged periods of time. As many of these signs are not specific for SUD, they can be easily misinterpreted as tokens of other problems such as mental or somatic comorbidity or those related to the ID itself (e.g., Van Duijvenbode et al., 2015; VanDerNagel et al., 2013). Such signs may include physical symptoms (e.g., altered consciousness, weight loss, poor general health), psychological symptoms (e.g., behavioural issues, mood swings, impulsiveness), and social problems (e.g., work- or school-related problems, forensic involvement, debts).

The lack of both validated questionnaires and systematic screening procedures further contributes to the underdiagnosis of SU(D) in individuals with ID (VanDerNagel et al., 2013). Widely used mainstream screening instruments for SU(D) such as the AUDIT (Alcohol Use Disorder Identification Test; Babor, Higgins-Biddle, Saunders, & Monteiro, 2001) may not be suitable for individuals with ID. They often rely on a relatively high level of self-reflection, recall, and cause-effect relationships, and may contain complex sentences and difficult wording. Items such as “How often during the last year have you failed to do what was normally expected of you because of drinking” (AUDIT, item 5), and their rating scales (never, monthly or less, 2–4 times a month, 2–3 times a week, and 4 or more times a week) may surpass the cognitive skills of the individual with ID. In addition, screening instruments are generally short, and therefore have a relatively straightforward focus that may evoke evasive responses. Using such a test in individuals with ID may result in both high levels of underreport and overreport (VanDerNagel et al., 2013).

Screening and Assessment Methods

As the diagnosis of SUD mostly relies on information on lack of control over use, social impairment due to SU, and risky use, self-reported information—even in the light of tendencies to underreport—is the cornerstone of SUD assessment. Collateral information, such as reports from family members or professional caregivers, as well as biomarker analysis (such as breath, blood, or urine analysis), can contributute to the assessment. Though the use of the latter is often found appealing (given the presumed “objective” diagnosis), biomarker analysis has some serious limitations, such as false-positive and false-negative testing, as well as a limited window of detection depending on the type of biomarker. In addition, biomarker testing is costly, and generally depends on the subject’s consent and cooperation (e.g., De Beaurepaire et al., 2007). But foremost, though biomarker testing can contribute to assessing SU (and to some extent to assessing amounts of use), it does not provide information on consequences of SU or control over SU.

Strategies that promote timely case-identification are discussing SU in an open, non-confrontational style, preferably before problems arise. Thus, SU should be the topic of conversation in a wide, non-selected population, and skills to adress it, should be present in professionals working with individuals with ID. In subgroups with a higher a priori risk for SUD, such as those with MID-BIF, and individuals with psychiatric, behavioural, or forensic comorbidity, periodically systematic screening should be in place (VanDerNagel et al., 2013). To do this, some authors (e.g., Pezzoni & Kouimtsidis, 2015; Van Duijvenbode, Didden, Korzilius, & Engels, 2017) have used mainstream screening and assessment instruments, that were adapted to the needs of individuals with ID. For example, by using an interview rather than self-report format or by using pictures to assist in differentiating between answer categories. To date, there is only one instrument specifically designed to assess SU(D) among individuals with ID (Box 50.2). Another line of research has been specifically focused on the potential use of implicit measures for the screening, assessment and treatment of SUD among individuals with ID (Box 50.3).

Box 50.2: The Substance Use and Misuse in Intellectual Disability Questionnaire

To our knowledge, the Substance use and misuse in Intellectual Disability Questionnaire (SumID-Q) is the first and only instrument specifically designed to assess SU(D) in individuals with ID (VanDerNagel, Kiewik, Van Dijk, De Jong,

(continued)

Box 50.2 (continued)

& Didden, 2011). It is used in an interview format with the client. The SumID-Q was developed and validated in Dutch, and translated in several other languages. The SumID-Q consists of several steps:

Step 0: Establish a good working relationship with the client, and be willing to discuss substance use in an open, empathetic way. Maintain this neutral stance and an inquisitive attitude during the whole interview.

Step 1: Assess the client's familiarity with substances, using pictures. In this step, the client is playfully confronted with a standardized set of pictures, depicting various products and types of use of tobacco, alcohol, cannabis, and other drugs. Key to this phase of the assessment is to elicit responses indicative for the client's familiarity with the substance, as well as his or her terminology for it. This may include common, but also peculiar slang words for various substances and methods of usage. In the remainder of the interview, the client's terminology is adopted and incorporated in the questions. Though this phase often elicits spontaneous responses indicating the client's SU (such as "I never smoke cigars, I prefer cigarettes"), the client is not pressed to elaborate on his or her experiences yet.

For each of the substances the client is familiar with, steps 2–4 are repeated.

Step 2: Discuss substances and SU in general. Assess the client's knowledge of and attitude towards each type of substance, using the predefined set of questions for each substance. To assist with answer categories, the SumID-Q contains pictures and symbols. During this phase, the interviewer should remain interested in the client's opinion and does not correct or confront him/her. Further assess SU in the client's social environment, by using the standardized questions on substance use among peers, family members, and staff members ("Do your friends smoke cigarettes?" etc.).

Step 3: Enquire about client's own experiences with substance use. After the set of questions regarding SU in the client's social environment, the question "Did you ever use … yourself?" can follow naturally. During this step, it is important to retain a neutral stance. Try to be interested without being inquisitive. Accept whatever answer was given, without questioning its validity.

Step 4: Further inquire about the use of this type of substance. Assess age of first use, patterns of SU (frequency, quantity), SU circumstances (alone/with others, at home or somewhere else), and SU effects (positive and negative). In this phase it remains important to keep a genuine interest in the client's story. Using pictures and focusing on the "how," "what," 'when," "with whom," and "to what effect" help to do so. Try to avoid "why" questions.

Box 50.3: Implicit Measurements

Another avenue that is currently explored to assess SU in individuals with ID, is the use of implicit measurements. SUD is characterized by disruptions in motivational, reward, and inhibitory control processes and subsequent deficiencies in information processing (Hyman, Malenka, & Nestler, 2006; Koob, 2013). More specifically, SUD has been associated with several cognitive biases, including biases in attention, action tendencies, and memory associations (Robinson & Berridge, 2008). This means that substance-related stimuli have gained "incentive salience" and, as a result, "grab the attention," seem attractive and elicit approach behavior. SUD has also been associated with executive dysfunctioning,

(continued)

Box 50.3 (continued)

reflected in a smaller working memory capacity, difficulties in delaying gratification, and less behavioral control in problematic substance users compared to non-problematic substance users (Hyman et al., 2006). Implicit measures are used to assess these neuropsychological changes. In theory, all of these neuropsychological changes can serve as markers for SUD and could therefore be incorporated in screening and assessment procedures.

However, a series of studies of cognitive biases in individuals with ID with alcohol use disorder provided inconclusive results (Van Duijvenbode et al., 2017). Overall, attentional and approach biases were not found. Problematic drinkers with ID did show a so-called interpretation or association bias. In a word association task, subjects were required to finish ambiguous scenarios such as "*Movie night at your friend's house. "One More?" one of your friends asks. You cannot resist the temptation and reach for ....*" Individuals with alcohol use disorders were more inclined to finish these scenarios with alcohol-related answers ("a beer") rather than neutral answers (e.g., "a new film").

The authors hypothesize that in individuals with ID and SUD, the same neuropsychological changes are present as in individuals with SUD without ID, but methodological challenges contributed to the lack of attentional and approach biases. These methodological challenges need to be addressed. The authors therefore discourage the use of currently available, indirect, reaction time-based cognitive bias measurements for clinical purposes but regard word-association tasks as an opportunity to identify high-risk situations for SU.

Diagnosis of SUD is relatively straightforward when basic information regarding SU patterns, behaviors and consequences is available. When an SUD is suspected or confirmed, it is important to assess usage of other substances (past and present) as well. Many substance users combine multiple substances, leading to increased somatic and psychiatric risks as well as potentially specific treatment needs. Smoking requires special attention, being a highly prevalent but often ignored addiction with severe physical and psychiatric complications. Past SU can be relevant both to assess the length of SU history, its complexity (e.g., with shifting combinations of SU), as well as potential risks for current health (including the risk of infections in former intravenous drug users and risks of neurological damage in former alcohol users). Differential diagnosis of a specific type of SUD consists mainly of 1. other SUD (i.e., symptoms ascribed to a stimulant use disorder, can also be caused by new substances such as "designer drugs"), and 2 of the same SUD with a different specifier for severity (e.g., "mild" versus "severe") or stage (e.g., "in early remission"). In addition, related to the direct effects of substances, SU intoxication or withdrawal can be scored (APA, 2013).

Screening and Assessment of Comorbidity

As SUD is often complicated by somatic and psychiatric comorbidity, as well as social issues, a wider assessment of problems is warranted. Instruments such as the Addiction Severity Index (ASI, McLellan, Luborsky, O'Brien, & Woody, 1980) addresses several problem areas such as medical status, employment and social support, drug and alcohol use, legal status, family/social status, and psychiatric status. The ASI requires an interview with a trained profession of about 1 h. For individuals with ID, these standardized questionnaires have the same issues as screening instruments for SUD, with the additional problem of their length. Therefore, screening for and assessing of comorbidity in individuals with ID and SUD often relies on non-standardized methods. In this process, several topics warrant attention: past and present medication use (including over the counter medication, as well as prescribed and non-prescribed opioids, benzodiazepines, and other psychoactive and non-psychoactive medication), medical history and current complaints, psychiatric history and current complaints, as well as assessment of psychosocial functioning. A full assessment should also include a physical and psychiatric examination, as well as a laboratory screening (hematology, liver- and kidney functioning, vitamin B1, B12, D, and thyroid functioning). Urine or saliva drug screening can be used to confirm and monitor (recent) SU but are not to be considered an alternative for an ongoing conversation with the individual about their substance use.

Prevention, Treatment, and Rehabilitation

As SU can have negative effects and may progress into SUD that warrants clinical attention, a variety of interventions is needed in prevention, treatment and rehabilitation. Unfortunately, there is a huge treatment gap: only about 10% of the people who needed substance use treatment received such treatment at a specialized facility in the past year (SAMSHA, 2017). For individuals with ID, access to such services may be even more limited for reasons we discussed earlier.

Prevention

Awareness of the risks for SU(D) in individuals with ID is a prerequisite for adequate preventative interventions. Such interventions may include identifying and reducing risk factors for SU (e.g., addressing mental health issues, inadequate coping, stress), promotion of a positive, healthy and socially inclusive lifestyle (adequate daytime activities, sufficient social and practical support), and reducing external risk factors (e.g., providing a smoke-free living environment, including regulations to prevent caregivers from using substances such as alcohol or tobacco in the presence of clients). Prohibition of and punishment for SU (including withholding care or limiting personal freedom) may have some temporary effects, but in the long run such measures causes serious and unwanted side-effects (e.g., secretive use). As the prison system and the “war on drugs” have proven: eradication of SU through restrictive measurements is unfeasible at best, and comes at a high cost.

Effects of education about the risks of SU as a sole intervention is not an effective strategy to prevent SU, neither in those with or in those without ID (see Kerr, Lawrence, Darbyshire, Middleton, & Fitzsimmons, 2013; Kiewik, VanDerNagel, Engels, & De Jong, 2017). However, both users and nonusers may have a need for factual information on SU to make informed decisions and to communicate with their caregivers. Direct caregivers (including family members and professional staff) are centerpiece in both primary prevention (prevention of SU), secondary prevention (timely case identification), and tertiary prevention (preventing further harm from SU). Therefore, one important preventative intervention is not so much to educate clients with ID but to educate their caregivers.

Treatment of SUD

In less progressed cases, SUD can be treated with relatively short interventions, such as the advice of a general practitioner (or family member/professional caregiver) to cut down on alcohol or benzodiazepine use or to quit smoking. Unfortunately, many clients with SUD are unable to control or cease their SU, even while they are aware of its risks and consequences (e.g., Van Duijvenbode et al., 2015). In those instances, SUD is a complex, chronic, relapsing, and remitting condition that warrants more complex and often multimodal interventions. These may include a combination of motivational strategies, detoxification, cognitive behavioral therapy, relapse prevention, and treatment of comorbidity.

Though SUD poses a threat to the health and well-being of the individual, treatment can be complicated with severe negative health effect. For instance, the detoxification of gamma hydroxy butyric acid (GHB) is associated with serious agitation and delirium, even in clients in specialized addiction services. Alcohol withdrawal symptoms (when not properly managed by benzodiazepine prescription) can lead to seizures and delirium. And restoration of normal dietary intake in malnourished alcoholics can lead to acute and severe neurological problems (Wernicke's syndrome) due to acute vitamin B1 deficiency (as vitamin B1 is utilized in digesting carbohydrates). All of these complications of SUD treatment can be avoided with appropriate medical interventions. Thus, treatment of SUD often requires specialized addiction knowledge and services. Inpatient treatment is needed when the patient requires permanent supervision and when complications within the treatment process are expected. These problems could include the aforementioned medical complications during detoxification but also behavioral or psychological issues, somatic or psychiatric comorbidity, or the inability of the client to complete an outpatient treatment.

SUD Treatment in Individuals with ID

In individuals with ID, a close collaboration between ID service providers and SUD treatment providers is warranted (see Slayter, 2010b; Van Duijvenbode et al., 2015; 2017). Ideally, both types of services know each other's qualities and limitations, and work collaboratively to provide a treatment program tailored to the needs of the individual patient.

General Adaptations

One of the main and common adaptations is the use of a trusted proxy (e.g., family member or professional caregiver) within the treatment program. Such a person can be of utmost value to reassure the client with SUD and ID, to help overcome misunderstandings, to follow through with the program, and to assist practicing newly learned behavior in daily life. Treatment providers in addiction medicine who treat clients with ID need to adapt their language and treatment protocol to the cognitive and adaptive abilities of these clients. Use of pictures, drawings and props as well as real-life examples are helpful. Generally, the client with SUD and ID will require more, but shorter sessions, a more prolonged treatment plan, and—last but not least—a more supportive than confrontational approach.

Motivational Interviewing

Motivational interviewing can help the client to consider the benefits of changing SU, and to explore his or her options how to achieve this change. Several studies suggest that this strategy is feasible for individuals with ID and SUD (e.g., Frielink & Embregts, 2013; Kouimtsidis et al., 2017; Mendel & Hipkins, 2002).

Cognitive Behavioral Therapy

Cognitive behavioral therapy (CBT) is the core of many addiction programs, aiming to increase self-control and improve alternative behavior. Preliminary data suggest that CBT can be sucessfully used in individuals with ID, either in brief extended interventions (Kouimtsidis et al., 2017) or in full CBT protocols (VanDerNagel & Kiewik, 2016). In addiction treatment centers, CBT often is offered in groups. This may not always be suitable for individuals with ID, unless they are placed in more or less homogeneous groups, in which communication and group interactions are correspondent to their abilities.

Inpatient Treatment

Inpatient detoxification may be required more often in individuals with ID, given their higher rates of comorbidity, and their limited cognitive and adaptive skills that may hinder treatment. As detoxification generally is a rather short, medically oriented intervention, providing this type of care to individuals with ID does not imply major adaptations.

Prolonged inpatient treatment may last up to several months, and often uses group therapy as a major intervention. As mentioned above, this may be a hindrance for individuals with ID. Also, individuals with ID may not profit from this type of intervention if they are mixed in mainstream groups. Group interactions—even outside therapy sessions—may be too complicated, the level of staff support in day to day functioning may be too little and vulnerable individuals with ID may be exploited or negatively influenced by group members.

To overcome these issues, several clinics in the Netherlands have established specialized wards for SUD treatment for individuals with ID. Within these wards, the focus is not only on the SUD but also on the development of daily living skills, restoration of daily structure and activities, as well as gradual rehabilitation into outpatient or community facilities.

Relapse Prevention

Given the chronic character of SUD and its associated changes in brain functioning, relapse prevention needs to be integral part of any SUD treatment program. Aim of this type of prevention is not so much eradication of relapse (which is probably unfeasible), but reducing the number and severity of relapses, and to prevent complications. Some clients with SUD in remission and their caregivers may not be very willing to engage in such a program, as they may regard SUD as a problem of the past. A program that focuses on positive goals for the future may be more acceptable to these groups, and achieve the same results as a relapse prevention program per se.

Medication

Prescribed medication can be used during detoxification (to prevent withdrawal), during maintenance treatment and relapse prevention, and to treat somatic and psychiatric comorbidity. Generally, treatment strategies, their indications and contraindications, do not differ between individuals with and without ID. However, clinicians should be aware that individuals with ID may respond differently (i.e., with an increase, decreased, or “paradoxical” effect) to psychoactive medication. Also, the use of aversive drugs (that induce very unpleasant and even dangerous effects when taken together with alcohol), warrant close scrutiny. Though clients with ID may benefit from such drugs (knowing they cannot use alcohol, for the risk of getting unwell), their safe use requires both a level of insight and self-control that may not be present in all individuals with ID.

Rehabilitation

Some clients may not be able to stop or control their SU for a prolonged time. In this group, tertiary prevention interventions (also called “harm reduction”) are needed. These include prevention of somatic complications (e.g., in needle exchange programs), monitoring of general and mental health, and providing social support (including housing, daytime activities and counselling). In all of these interventions, knowledge and skills of ID service providers is needed, as withholding these services may contribute to deterioration of the client’s condition.

Treatment of Comorbidity

Treatment of comorbid somatic and mental health conditions may contribute to the reduction of risk for (prolonged) SU. However, comorbid disorders generally require treatment of both conditions. In the case of substance use induced mental health disorders, symptomatic treatment may be required for the psychiatric symptoms, even though recovery after prolonged abstinence is to be expected. Special consideration should be given to the risk of increasing symptoms of one of the conditions as a result of the treatment of the other condition. Examples of these include relapse or worsening of symptoms of posttraumatic stress disorder (such as irritability or sleeplessness) after abstinence of cannabis, and increasing risks of SU by treating ADHD or pain with poorly monitored prescription medication. However, as mentioned earlier, withholding adequate treatment and support for comorbidity will not help to reduce symptoms and consequences of SUD.

Conclusion

Substance use disorders (SUD) are among the most common mental disorders in the adult population. Substance use (disorders) among individuals with intellectual disabilities (ID) has gained increasing attention, with a growing number of studies on prevalence, assessment, prevention, and treatment of SUD in this population. Although it is still challenging to provide reliable SU and SUD prevalence rates in individuals with ID, it is generally accepted that individuals with ID are at risk for developing SUD and experience more severe negative consequences from SU.

A number of suggestions for policy and practice to further improve the care for individuals with SUD and ID can be made. First, systematic screening and assessment of SU(D) among individuals with ID is essential for timely case identification and increasing the opportunities to intervene in an early phase. Second, professionals need to be aware of possible comorbidity with other medical, mental health, and psychosocial problems. These issues should be included in the assessment and treatment. Third, individuals with ID require specialized care from multidisciplinary teams in which knowledge and expertise regarding both ID and SUD is present. Collaboration and fertilization between addiction medicine and ID service providers are key factors in achieving this. Initiatives such as adapted assessment and treatment protocols (see for example Box 50.2 and 50.3) are therefore welcomed and are in fact becoming increasingly available.

References

American Psychiatric Association. (2013). *Diagnostic and statistical manual of mental disorders* (5th ed.). Washington, DC: American Psychiatric Association.

Babor, T. F., Higgins-Biddle, J. C., Saunders, J. B., & Monteiro, M. G. (2001). *AUDIT the alcohol use disorders identification test, guidelines for use in primary care*. Geneva: World Health Organization.

De Beaurepaire, R., Lukasiewicz, M., Beauverie, P., Castéra, S., Dagorne, O., Espaze, R., … Molimard, R. (2007). Comparison of self-reports and biological measures for alcohol, tobacco, and illicit drugs consumption in psychiatric inpatients. *European Psychiatry, 22*, 540–548.

Didden, R., Embregts, P., Van der Toorn, M., & Laarhoven, N. (2009). Substance abuse, coping strategies, adaptive skills and behavioral and emotional problems in clients with mild to borderline intellectual disability admitted to a treatment facility: A pilot study. *Research in Developmental Disabilities, 30*, 927–932.

Emerson, E., & Turnbull, L. (2005). Self reported smoking and alcohol use, among adolescents with intellectual disabilities. *Journal of Intellectual Disabilities, 9*, 58–69.

Frielink, N., & Embregts, P. (2013). Modification of motivational interviewing for use with people with mild intellectual disability and challenging behaviour. *Journal of Intellectual and Developmental Disabilities, 38*, 279–291.

Hyman, S. E., Malenka, R. C., & Nestler, E. J. (2006). Neural mechanisms of addiction: The role of reward-related learning and memory. *Annual Review of Neuroscience, 29*, 565–598.

Kerr, S., Lawrence, M., Darbyshire, C., Middleton, A. R., & Fitzsimmons, L. (2013). Tobacco and alcohol-related interventions for people with mild/moderate intellectual disabilities: A systematic review of the literature. *Journal of Intellectual Disability Research, 57*, 393–408.

Kiewik, M., VanDerNagel, J. E. L., Engels, R. C. M. E., & De Jong, C. A. J. (2017). The efficacy of an e-learning prevention program for substance use among adolescents with intellectual disabilities: A pilot study. *Research in Developmental Disabilities, 63*, 160–166.

Koob, G. F. (2013). Addiction is a reward deficit and stress surfeit disorder. *Frontiers in Psychiatry, 4*, 1–18.

Kouimtsidis, C., Bosco, A., Scior, K., Baio, G., Hunter, R., Pezzoni, V., … Hassiotis, A. (2017). A feasibility randomized controlled trial of extended brief intervention for alcohol misuse in adults with mild to moderate intellectual disabilities living in the community: The EBI-LD study. *Trials, 18*, 1–12.

Lin, E., Balogh, R., McGarry, C., Selick, A., Dobranowski, K., Wilton, A. S., & Lunsky, Y. (2016). Substance-related and addictive disorders among adults with intellectual and developmental disabilities (IDD): An Ontario population cohort study. *BMJ Open, 6*, e011638.

Lindsay, W. R., Carson, D., Holland, A. J., Taylor, J. L., O'Brien, G., Wheeler, J. R., & Steptoe, L. (2013). Alcohol and its relationship to offence variables in a cohort of offenders with intellectual disability. *Journal of Intellectual and Developmental Disability, 38*, 325–331.

Luteijn, I., Didden, R., & VanDerNagel, J. E. L. (2017). Individuals with Mild Intellectual Disability or Borderline Intellectual Functioning in a Forensic Addiction Treatment Centre: Prevalence and Clinical Characteristics. *Advances in Neurodevelopmental Disorders, 1*, 240–251.

McLellan, A. T., Luborsky, L., O'Brien, C. P., & Woody, G. E. (1980). An improved diagnostic instrument for substance abuse patients: The Addiction Severity Index. *Journal of Nervous & Mental Diseases, 168*, 26–33.

Mendel, E., & Hipkins, J. (2002). Motivating learning disabled offenders with alcohol-relatedproblems: A pilot study. *British Journal of Learning Disabilities, 30*, 153–158.

Mikkelsen, E. J., VanDerNagel, J. E. L., & Lindsay, W. R. (2016). Substance related and addictive disorders. In R. Fletcher, J. Barnhill, & S.-A. Cooper (Eds.), *Diagnostic manual—intellectual disability DM-ID-II. A textbook of diagnosis of mental disorders in persons with intellectual disability* (pp. 561–572). Washington: NADD Press.

Pezzoni, V., & Kouimtsidis, C. (2015). Screening for alcohol misuse within people attending a psychiatric intellectual disability community service. *Journal in Intellectual Disability Research, 4*, 353–359.

Pacoricona Alfaro, D. L., Ehlinger, V., Spilka, S., Ross, J., Sentenac, M., & Godeau, E. (2017). Alcohol, tobacco and cannabis use: Do students with mild-intellectual disability mimic students in the general population? *Research in Developmental Disabilities, 63*, 118–131.

Plant, A., McDermott, E., Chester, V., & Alexander, R. T. (2011). Substance misuse among offenders in a forensic intellectual disability service. *Journal of Learning Disabilities and Offending Behaviour, 2*, 127–135.

Reis, O., Wetzel, B., & Haßler, F. (2017). Mild or borderline intellectual disability as a risk for alcohol consumption in adolescents—A matched-pair study. *Research in Developmental Disabilities, 63*, 132–141.

Robinson, T. E., & Berridge, K. C. (2008). The incentive sensitization theory of addiction: Some current issues. *Philosophical Transaction of the Royal Society of London: Series B, Biological Sciences, 363*, 3137–3146.

Slayter, E. M. (2010a). Demographic and clinical characteristics of people with intellectual disabilities with and without substance abuse disorders in a medicaid population. *Intellectual and Developmental Disabilities, 48*, 417–431.

Slayter, E. M. (2010b). Disparities in access to substance abuse treatment among people with intellectual disabilities and serious mental illness. *Health and Social Work, 35*, 49–59.

Sturmey, P., Reyer, H., Lee, R., & Robek, A. (2003). *Substance-related disorders in persons with mental retardation*. Kingston: NADD Press.

Substance Abuse and Mental Health Services Administration [SAMHSA]. (2017). *Key substance use and mental health indicators in the United States: Results from the 2016 National Survey on Drug Use and Health*. Rockville: SAMHSA.

Swerts, C., Vandevelde, S., VanDerNagel, J. E. L., Vanderplasschen, W., Claes, C., & DeMayer, J. (2017). Substance use among persons with intellectual disabilities (ID) living independently in the community in Flanders. *Research in Developmental Disabilities, 63*, 107–117.

To, W. T., Neirynck, S., Vanderplasschen, W., Vanheule, S., & Vandevelde, S. (2014). Substance use and misuse in persons with intellectual disabilities (ID): Results of a survey in ID and addiction services in Flanders. *Research in Developmental Disabilities, 35*, 1–9.

To, W. T., Vanheule, S., Vanderplassschen, W., Audenaert, K., & Vandevelde, S. (2015). *Screening for intellectual disability in persons with a substance abuse problem: Exploring the validity of the Hayes Ability Screening Index in a Dutch-speaking sample*. *Research in Developmental Disabilities* (Vol. 36, pp. 498–504).

Van Duijvenbode, N., VanDerNagel, J. E. L., Didden, R., Engels, R., Buitelaar, J., Kiewik, M., & de Jong, C. A. (2015). Substance use disorders in individuals with mild to borderline intellectual disabilities: Current status and future directions. *Research in Developmental Disabilities, 38*, 319–328.

Van Duijvenbode, N., Didden, R., Korzilius, H., & Engels, R. (2017). The usefulness of implicit measures for the screening, assessment and treatment of problematic alcohol use in individuals with mild to borderline intellectual disability. *Advances in Neurodevelopmental Disorders, 1*, 42–51.

VanDerNagel, J., Kemna, L., & Didden, R. (2013). Substance use among persons with mild intellectual disability: Approaches to screening and interviewing. *The NADD Bulletin, 16*, 87–92.

VanDerNagel, J. E. L., & Kiewik, M. (2016). *CGT+, Cognitieve gedragstherapeutische behandeling voor problematisch middelengebruik bij mensen met een Lichte Verstandelijke Beperking [CBT+, Cognitive behavioral therapy for substance use disorder in individuals with mild intellectual disability]*. Utrecht: Perspectief.

VanDerNagel, J. E. L., Kiewik, M., Buitelaar, J. K., & De Jong, C. A. J. (2011). Staff perspectives of substance use and misuse among adults with intellectual disabilities enrolled in Dutch disability services. *Journal of Policy and Practice in Intellectual Disabilities, 8*, 143–149.

VanDerNagel, J. E. L., Kiewik, M., Van Dijk, M., De Jong, C. A. J., & Didden, R. (2011). *Handleiding SumID-Q, Meetinstrument voor het in kaart brengen van Middelengebruik bij mensen met een lichte verstandelijke beperking [Manual of the SumID-Q. An instrument to assess substance use in individuals with a mild intellectual disability]*. Deventer: Tactus.

VanDerNagel, J. E. L., Kiewik, M., Postel, M. G., Van Dijk, M., Didden, R., Buitelaar, J. K., & de Jong, C. A. (2014). Capture recapture estimation of the prevalence of mild intellectual disability and substance use disorder. *Research in Developmental Disabilities, 35*, 808–813.

VanDerNagel, J. E. L., Kiewik, M., Didden, R., Korzilius, H. P. L. M., Van Dijk, M., Van der Palen, J., … de Jong, C. A. J. (2017). Substance use in individuals with mild to borderline intellectual disability in the Netherlands: An exploration of risks and rates. *Advances in Neurodevelopmental Disorders, 1*, 283–293.

Volkow, N. D., Wang, G.-J., Tomasi, D., & Baler, R. D. (2013). Unbalanced neuronal circuits in addiction. *Current Opinion in Neurobiology, 23*, 639–648.

Wechsler, D. (2004). *WAIS-III, Nederlandstalige bewerking*. Lisse: Swets Test Publishers.

51

Dementia and Intellectual Disability: Prevalence, Assessment and Post-Diagnostic Support

Eimear McGlinchey, Evelyn Reilly,
Philip McCallion, Pamela Dunne, Niamh Mulryan,
Rachael Carroll, and Mary McCarron

Chapter Overview

This chapter will focus on dementia in people with an intellectual disability. Because for this group a genetic risk factor for Alzheimer's disease leads to higher and earlier incidence, the majority of research to date into dementia in people with an intellectual disability has focused on Alzheimer's disease in those with Down syndrome.

This chapter will discuss the prevalence of dementia in people with Down syndrome, and for people with ID and without Down syndrome. The presentation of dementia will be discussed, along with the assessment of dementia and challenges to assessment due to pre-existing intellectual disability. Post-diagnostic support will be illustrated using a case study highlighting best practice dementia standards in an intellectual disability specialised memory clinic.

Throughout the chapter, key points will be supported using data from the Intellectual Disability Supplement to the Irish Longitudinal Study on Ageing (IDS-TILDA) and from a 20-year longitudinal follow-up study on incidence of dementia in people with Down syndrome.

The Intellectual Disability Supplement to the Irish Longitudinal Study on Ageing (IDS-TILDA) is a unique nationally representative longitudinal study. It is the only study of its kind where people with an intellectual disability over the age of 40 are followed longitudinally in tandem with the general population. Both studies gather data on a wide range of issues, including, social participation and connection, health, mental health, health care utilisation, cognitive health and level of functioning. This allows for direct comparison of the ageing experiences of people with an intellectual disability to those of the general ageing population. Data collected includes dementia diagnosis, ADLs, chronic health conditions, and objective measures of cognitive function. The study has been ongoing since 2009, and has published three waves of data to date. In Wave 1 of IDS-TILDA, there were 753 people with an intellectual disability, of which 149 had Down

---

E. McGlinchey (✉) · M. McCarron
Trinity Centre for Ageing and Intellectual Disability, Trinity College Dublin, The University of Dublin, Dublin, Ireland
e-mail: mcgline@tcd.ie

E. Reilly · P. Dunne · N. Mulryan
Daughters of Charity Disability Support Service, Dublin, Ireland

P. McCallion
School of Social Work, Temple University, Philadelphia, PA, USA

R. Carroll
IDS TILDA and School of Nursing and Midwifery, Trinity College Dublin, The University of Dublin, Dublin, Ireland

J. L. Matson (ed.), *Handbook of Intellectual Disabilities*, Autism and Child Psychopathology Series, https://doi.org/10.1007/978-3-030-20843-1_51

syndrome. In Wave 3, there were 609 individuals, of which 109 had Down syndrome, with the majority of attrition attributed to deaths.

The second study is a prospective 20-year longitudinal study on the incidence of dementia, co morbidities, and mortality, in 77 people with Down syndrome, in a dementia specific service (McCarron, McCallion, Reilly, & Mulryan, 2014a, McCarron et al., 2017). For the 77 individuals enrolled in the study, there was a comprehensive baseline screening, with annual follow-up of both cognitive and physical functioning.

Dementia and Alzheimer's

As much of the research focuses on people with Down syndrome, so too does much of it focus on Alzheimer's disease rather than other types of dementia.

Dementia in the general population given it coincides with increases in longevity and reductions of premature deaths (Livingston et al., 2017) is increasingly reported to be the greatest global challenge for health and social care, with the number of people with dementia expected to triple by 2050. The estimated global cost of dementia in 2015 was $818 billion dollars (Prince et al., 2017), a 35% increase over 2010 estimates (Wimo et al., 2017). It should be noted that a progressive decline in the incidence of dementia since the 1970s in the general population has been noted (Satizabal et al., 2016). The reasons for this are not fully understood; however, the earlier diagnosis and more effective treatment of stroke and heart disease may have played a role (Satizabal et al., 2016).

Alzheimer's Disease

Alzheimer's disease is the most common form of dementia, and is the cause of approximately 50–70% of all dementias (Alzheimer's Association, 2018). In the 1984 diagnostic guidelines, to be diagnosed with Alzheimer's disease, symptoms of dementia must be present. This was changed in the 2011 guidelines, where Alzheimer's disease now includes the continuum from initial changes in brain pathology, with or without symptoms, through to dementia. What was previously classified as Alzheimer's disease is now referred to as Alzheimer's dementia, or dementia due to Alzheimer's, and is the phase in the continuum where symptoms of dementia are present (Alzheimer's Association, 2018).

Neuropathology of Alzheimer's and Specific Risk for People with Down Syndrome

Since the discovery of Alzheimer's disease, the only definitive diagnosis was and is at autopsy. Alzheimer's disease is characterised by positive B amyloid neuritic plaques and phosphorylated-tau neurofibrillary tangles (NFT), along with neuronal and synaptic loss (Theofilas et al., 2018).

People with Down syndrome have a genetic risk for developing Alzheimer's disease. A number of studies have shown that by the age of 40, all people with Down syndrome will have the neuropathology of Alzheimer's disease (Lamar et al., 2011; Schupf et al., 2010). The most common cause of Down syndrome is trisomy 21—a triplication of chromosome 21. The amyloid precursor protein (APP) gene is located on chromosome 21, and thus, due to trisomy 21, people with Down syndrome have 4–5 times overexpression of APP (Jennings et al., 2015). This increase in the APP expression leads to an increase in ß-amyloid deposition. The amyloid cascade hypothesis (Hardy & Higgins, 1992; Selkoe, 1991) suggests that the deposition of amyloid ß plays a critical role in the development of AD pathology, with Aß40 and Aß42 being the two major species generated by the amyloid precursor protein (APP) (Schupf et al., 2010). Plasma levels of Aß42 and Aß40 have been found to be higher at all ages in individuals with DS compared to age matched peers (Prasher, Sajith, Mehta, Zigman, & Schupf, 2010).

Tau protein has also been the focus of explanations for the neuropathology of Alzheimer's disease in people with Down syndrome possibly helping to explain neuronal death (Butterfield, Di Domenico, Tramutola, Head, &

Perluigi, 2017). Individuals with Alzheimer's disease are identifiable based on amounts of insoluble tau protein and phosphorylated tau species (Mukaetova-Ladinska, Harrington, Roth, & Wischik, 1993). Changes in tau pathology begin in the outer layer of the hippocampus, with neurofibrillary tangles in the hippocampal CA1 region then found with neuronal loss in the entorhinal cortex (Head et al., 2003). There is also a higher density of neurofibrillary tangles in those with Down syndrome compared to people in the general population (Head, Lott, Wilcock, & Lemere, 2016).

Prevalence, Incidence, Duration, Age of Onset of Dementia

Prevalence

A number of population based studies have focused on the prevalence of Alzheimer's dementia in people with Down syndrome, with the consensus that dementia is far more prevalent in people with Down syndrome than is found in people with intellectual disability from other aetiologies or for people in the general population.

A review of population prevalence based studies was conducted in 2010 (Strydom et al., 2010), with prevalence listed in Table 51.1.

A number of studies have also calculated the age specific risk of dementia in people with Down syndrome. Coppus et al. (2006) reported that, from the age of 45, dementia prevalence doubled with each 5-year interval, up to the age of 60. In this study, prevalence was then reported to decline after the age of 60, most likely due to the higher mortality rates in this age group, as there was no decrease in incidence of dementia over the age of 60.

Table 51.1 Prevalence of dementia in people with Down syndrome; review of studies

Author	Year	Percent
Cooper et al.	1997	21.6
Sekijima et al.	1998	15.1
Holland et al.	1998	24
Tyrell et al.	2001	13.3
Coppus et al.	2006	16.8
Strydom et al.	2007	13.1
Nieuwenhuis-mark	2009	15

In a 20-year longitudinal study in Ireland, age specific risk of developing dementia in a group of women with Down syndrome using incidence rates (McCarron et al., 2017) was calculated as 23% at age 50 years, 45% at age 55 years and 88% at age 65 years. Over 20 years, 97.4% of participants with Down syndrome developed dementia.

In Ireland in Wave 1 of IDS-TILDA, reported overall prevalence of dementia for participants with ID was 5.8%. As the sample aged, this had risen to 9% in Wave 2 and 9.1% in Wave 3. The graph below shows the prevalence of dementia for the overall sample in IDS-TILDA compared to the sub sample with Down syndrome. In Wave 1, 15.8% of those with Down syndrome over the age of 40 had a diagnosis of dementia, compared to 2% of those with non-DS-ID. By Wave 3, this had more than doubled, with 35.5% of those with Down syndrome reporting a diagnosis of dementia; this compared to 3.4% of those with an intellectual disability but without Down syndrome. These figures, however, are likely to be an underestimation, as it was found of those without a diagnosis of dementia, 47% had never had a dementia assessment (McCarron et al., 2017) (Fig. 51.1).

Mean Age of Onset and Duration of Dementia

Understanding the age at which dementia is likely to occur and the number of years with which one will live with dementia has serious implications for individual quality of life, service provision, planning and training needs.

In the same 20-year follow-up study, all 77 participants were screened annually for dementia, and when changes were noted, they were formally assessed with a multidisciplinary team arriving at a consensus diagnosis. The average age of onset in the 20-year study was 55 years, with an average duration of 5.6 years and a median duration of 7 years (McCarron et al., 2017). For those with pre-existing severe or profound ID, survival post diagnosis was 8.6 years.

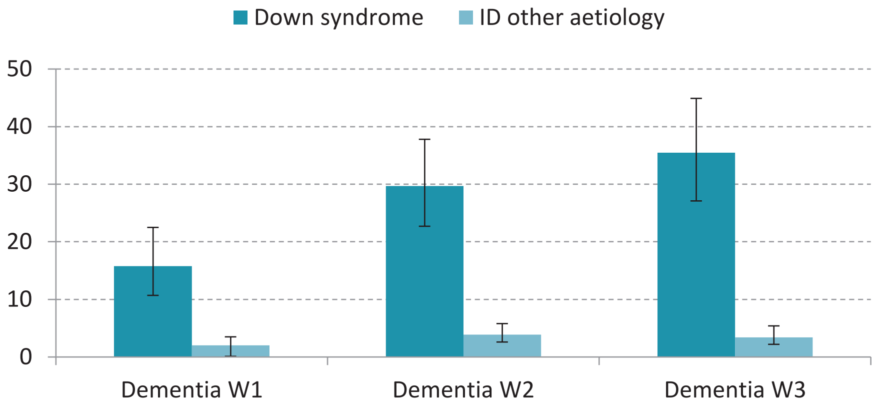


Fig. 51.1 Prevalence of dementia in IDS-TILDA. Wave 1 *n* = 752; Missing Obvs = 1; Wave 2 *n* = 705; Missing Obvs = 3; Wave 3 *n* = 604; Missing obvs = 5; Error bars correspond to 95% confidence intervals

Findings here were similar to those from a study of data extracted from clinical records of 839 individuals (254 with dementia) referred for memory screening. Here the average age of diagnosis was 55.8 years with a duration of 3.78 years and approximately 50% were diagnosed in their sixth decade (Sinai et al., 2017). Those living with family were diagnosed 4.5 years earlier than those who were living away from family.

Despite differences in methodology, there is agreement on age of diagnosis at 55 years in both studies, confirming previous findings of average age of onset of between 51 and 55 years (Lai & Williams, 1989; Prasher & Krishnan, 1993). No significant difference was noted in age of onset for those with mild/moderate or severe/profound intellectual disability (McCarron et al., 2017; Strydom, Hassiotis, King, & Livingston, 2009).

Prevalence of Alzheimer's in Non-DS ID

Prevalence rates for people with intellectual disability but without Down syndrome are not as well established. There has been no consensus on the prevalence with one study showing that prevalence was higher than the general population at 21.6% (Cooper, 1997), and others showing that it was similar to what is found in the general population at 9% in those aged 65 and over (Zigman et al., 2004). In the most recent study to examine prevalence in people with intellectual disability without Down syndrome, it was reported that incidence of dementia is up to five times higher for people with intellectual disability than is found in the general population (Strydom, Chan, King, Hassiotis, & Livingston, 2013).

Presentation of Dementia

Presentation in People with Down Syndrome

Previous literature has shown that all individuals with Down syndrome will have neuropathological and neuro-imaging features of Alzheimer's disease by the age of 40 (Lamar et al., 2011; Roizen & Patterson, 2003; Schupf et al., 2010). Furthermore, it has been shown that age related atrophy in Down syndrome is similar to the atrophy observed in early stages of Alzheimer's disease in those without Down syndrome (Carr & Collins, 2018; Teipel & Hampel, 2006). Nevertheless, presentation can vary largely between individuals and the subsequent rate of decline, although the reasons for this are not fully understood (Lautarescu, Holland, & Zaman, 2017).

Despite the neuropathology being present, it is important to understand that dementia and presentation of clinical symptoms is not inevitable for everyone with Down syndrome. It is essential therefore to understand the unique presentation and progression of dementia in this population so that dementia is not diagnosed when there are other causes of the symptoms observed that may be manageable or treatable (O'Caoimh, Clune, & Molloy, 2013).

The presentation of the later stages of dementia is reportedly similar to that seen in the general population (McCarron et al., 2018; Strydom et al., 2010). The early stages of dementia in people with Down syndrome, however, are believed to be atypical (Lautarescu et al., 2017). While there is some overlap in symptoms such as memory loss, confusion, deterioration in speech, the earliest changes noticed appear to be personality and behavioural changes associated with executive dysfunction, rather than the short-term memory loss that is seen in the general population (Ball et al., 2006). In a systematic review of the early presentation of dementia in people with Down syndrome, 9 of the 15 studies identified "frontal-like symptoms" as the earliest change (Lautarescu et al., 2017).

Behavioural changes may be misdiagnosed as new onset behavioural disorder or depression. Yet, Holland, Hon, Huppert, Stevens, and Watson (1998) reported that behavioural changes noted by carers retrospectively such as apathy, stubbornness, withdrawal were the earliest changes noticed prior to a clinical diagnosis. Behavioural changes have been described by Ball et al. (2006) as being more typical of dementia of the frontal type, and may be seen as preclinical Alzheimer's, which occurs before the continued progression to clinical dementia. For 48% of persons observed in one study, the deterioration of activities of daily living (ADL) were noted as trigger symptoms, with deterioration first noted in personal hygiene and later in spatial disorientation (Cosgrave, Tyrrell, McCarron, Gill, & Lawlor, 2000).

Behavioural and Psychological Symptoms of Dementia (BPSD) are defined as a "heterogeneous range of psychological reactions, psychiatric symptoms and behaviours resulting from the presence of dementia" (Finkel, 2001). BPSD have been studied intensively over the past two decades in the general population, using scales such as the Behavioural Pathology in AD Rating Scale (BEHAVE-AD) (Reisberg, Auer, & Monteiro, 1997) and the Neuropsychiatric Inventory (Cummings et al., 1994). However, despite increased reported prevalence of psychological and behavioural symptoms of dementia, these scales have not been adapted or validated for people with an intellectual disability (Dekker et al., 2015).

Presentation in People with Non-DS ID

For people with intellectual disability without Down syndrome, decline in general functioning along with emotional and behavioural changes are frequently reported with aggression seen more often in those with intellectual disability without Down syndrome, and a higher rate of other behavioural changes noted for those with Down syndrome (Strydom et al., 2010).

Assessment and Assessment Challenges

In this section, assessment for dementia and the challenges in assessment and diagnosis will be highlighted. In discussing the cognitive assessments available and the challenges associated with diagnosis, no differentiation will be made between those with and those without Down syndrome as the same issues appear to apply to all people with intellectual disability regardless of aetiology. However, the use of neuroimaging in diagnosis will be discussed for those with Down syndrome.

In the general population, the diagnosis of dementia depends on reliable clinical history (Bayer, 2018) and includes assessment of core clinical criteria that include

- Interference with ability to work or function.
- Decline from previous function.
- Not explained by major psychiatric episode.
- Cognitive impairment detected and diagnosed through.
  - History taking from patient and reliable informant.
  - Objective cognitive assessment.
- Impairment includes two or more of the following.
  - Impaired ability to acquire or learn new information.
  - Impaired reasoning or handling of complex tasks.
  - Impaired visuospatial abilities.
  - Impaired language function.
  - Changes in personality or behaviour.

    (McKhann et al., 2011)

The International Classification of Diseases (ICD-10) (World Health Organization, 2004) and the Diagnostic and Statistical Manual of Mental Disorders (DSM-V) (American Psychiatric Association, 2013) lay out criteria that need to be met in order to dementia to be diagnosed. There are some differences in the criteria depending on which is used, where the ICD-10 puts emphasis on emotional changes, where the DSM-V does not.

Usefulness of Criteria for Dementia and Mild Cognitive Impairment for People with Intellectual Disability

There are no specific guidelines in the ICD-10 (World Health Organization, 2004) or the DSM-V (American Psychiatric Association, 2013) for diagnosing dementia in people with intellectual disability (O'Caoimh et al., 2013). The same criteria, however, can be used for people with intellectual disability as are used in the general population, as long as it is understood that any observed decline is relative to baseline functioning levels, rather than impairment to "normal" functioning (Ball et al., 2004). The Diagnostic Criteria for use with adults with Learning Disabilities (DC-LD) was developed in 2003 to compliment the ICD-10 and to improve accuracy of diagnosis in people with an intellectual disability (Royal College of Psychiatrists, 2001). It was found that there was some discrepancy between rates of diagnosis using the DC-LD compared to previous diagnoses using the ICD-10, including discrepancy in prevalence of dementia, but that the DC-LD was useful in aiding the diagnosis in this population (Tully, Schirliu, & Moran, 2012).

It is essential that criteria for diagnosing dementia are able to differentiate between deficits due to dementia and those due to other cognitive deficits, such as is found in intellectual disability. One study examining the predictive validity and reliability of the DSM-IV and the ICS-10 criteria when used in a one-off assessment with people with intellectual disability (Strydom et al., 2013) found that, although validity and reliability were satisfactory, results were less stable than reported in the general population. This was particularly true for people with severe or profound intellectual disability.

There is no consensus on whether the ICD-10 or the DSM-IV criteria are more useful for people with intellectual disability (Aylward, Burt, Thorpe, Lai, & Dalton, 1997). Prior to the DSM IV, the ICD-10 criteria was recommended for use with people with an intellectual disability as there is greater emphasis on the emotional, or "non-cognitive" aspects of dementia, which are often the earliest signs reported by people with an intellectual disability. In a more recent study, the DSM-IV criteria were found to be more inclusive of people with an intellectual disability with moderate to severe dementia (Strydom et al., 2007). More work is needed to see if this also holds for use of DSM V criteria.

Challenges in Assessment

The presentation of the early stages of dementia in people with intellectual disability can differ from early symptoms in the general population. Difficulties in diagnosis may be due to the pre-existing intellectual disability (Aylward et al., 1997) and the fact that individuals do not reach the same cognitive ability as individuals without an intellectual disability, thus it may be more difficult to identify decline due to dementia, rather than pre-existing intellectual disability (Rowe, Lavender, & Turk, 2006). For people with Down syndrome, it has also been found that by age 50, scores decline on tests even for those without dementia—suggesting there is a confounding age related decline in people with Down syndrome that is not due to dementia (Carr & Collins, 2018). Non-dementia age-related cognitive decline has similarly been documented in the ageing general population (Grady, 2008). For instance, older individuals regularly perform at a lower level than their younger counterparts when engaged in cognitive tests that require working memory, task switching and attention, all of which are largely associated with executive functioning. It should be noted, however, that these effects seem to occur at a later stage in the general population, with most research in this field tending to focus on those in their seventh decade.

In the general population, dementia diagnosis relies on the integration of history, physical examination and cognitive testing (Feldman et al., 2008). While this is true also for those with intellectual disability, a number of challenges pose unique difficulties for diagnosis:

- Difficulties in communication.
- Lack of baseline measurements.
- Difficulties in using standardised tests.
- High staff turnover.
- Lack of awareness of caregivers of the significance of decline.

(McCallion & McCarron, 2004; McCarron & Lawlor, 2003).

A Model of Best Practice for Diagnosis

An international consensus group developed a model of best practice for the diagnosis of people with intellectual disability in order to overcome these challenges (Burt & Aylward, 2000). They recommended a baseline assessment of people with Down syndrome >35 and for others with an intellectual disability >50 and an annual follow-up assessment with agreed and validate measures. This model continues to be refined with guidance for a comprehensive diagnostic workup if an initial decline is noted and a person centred approach to subsequent care including specialist staff training, standardised assessments, and further research (McCarron et al., 2014a; McCarron et al., 2017; Wilkinson & Janicki, 2002). These recommendations continue to be endorsed, with the British Psychological Society continuing to recommend reactive screening and prospective monitoring (British Psychological Society and Royal College of Psychiatrists, 2015; Jokinen, Janicki, Keller, McCallion, & Force, 2013).

Assessment Process

Based on the model of best practice, the recommended assessment process is:

Baseline Assessment

A baseline assessment should be completed for adults with Down syndrome by age 35 and for adults with intellectual disability from other aetiologies by age 50 (Aylward et al., 1997). It is more straightforward to measure pre-morbid level of functioning in the general population through self-report or employment history; however, this is not always the case with an individual with intellectual disability, making baseline screening more important in this population. Measurement of a pre-dementia baseline is needed against which deterioration later in life may be measured avoiding reliance on a "normative" level for an assessment instrument, which likely does not apply for a person with an intellectual disability.

Data from the IDS-TILDA study shows that the percentage of the overall intellectual disability population in Ireland who had ever had a dementia assessment increased from 14.3% in wave 1–30% in Wave 3 (6 years later), and from 13.9% to 61.4% for those with Down syndrome.

While this increase in screening is positive, the data also shows that 47% of those with Down syndrome who did not have a diagnosis of dementia at wave 3 had never had a dementia assessment. This finding highlights that prevalence figures may be underestimated due to lack of baseline screening.

Monitoring

Annual screening after baseline screening is recommended so that any change is picked up at the earliest possible stage. Annual screening should also include information on any other changes noted in everyday functioning.

Regular monitoring ensures that (1) changes due to dementia are noted early in the course of the illness, (2) any other health conditions that may mimic symptoms of dementia, but are treatable are noted and addressed and (3) care plans are adapted if medical, psychosocial or environmental interventions need to be put in place.

Dementia Assessment and Diagnostic Work Up

If changes are noted in the screening process, a dementia assessment is recommended. The initial step is to rule out other physical causes that could mimic the symptoms of dementia. Physical, psychiatric, neurological and audiology assessments are needed (McCarron & Lawlor, 2003).

Recommended physical checks include

- Full blood count.
- Urea and electrolytes.
- Blood sugar.
- Thyroid function tests.
- Liver function tests.
- Folate and B12.
- Lipid levels.
- Sensory screening—vision and hearing.

(British Psychological Society & Royal College of Psychiatrists, 2015)

A dementia diagnosis should result from a multidisciplinary team coming to a consensus agreement on diagnosis after considering assessment data on the individual's physical, cognitive, emotional and mental state, along reviewing possible environmental influences and external life events that may have contributed to the change.

Assessment Instruments

Assessment of cognitive function is essential in the field of dementia study, both for timely diagnosis, and for symptom measurement following treatment using anti-dementia drugs. While the Montreal Cognitive Assessment (MOCA) (Nasreddine et al., 2005) and the Dementia Rating Scale (DRS) (Mattis, 1988) are standardised and validated in the general population, they are not suitable for use with people with an intellectual disability, particularly for those with moderate or more severe level of intellectual disability. For example, it has been found in one study that three quarters of people with ID who might be classified as having dementia based on Mini Mental State Examination (MMSE) score did have a diagnosis of dementia when other criteria were applied (Deb & Braganza, 1999).

One difficulty in diagnosing dementia in this population is the lack of an agreed battery of instruments. The benefits of having such standardised tests for use with a population with intellectual disability would include results being directly comparable, and it would facilitate communication between clinicians (Zeilinger, Stiehl, & Weber, 2013). In a systematic review on assessment instruments for dementia for people with an intellectual disability, 114 different tools were identified by Zeilinger et al. (2013).

Administration of neuropsychological assessments can be challenging to administer, due to communication difficulties and pre-existing intellectual impairment, particularly for those with severe or profound intellectual disability (McCarron et al., 2017). Due to these challenges, there is often a reliance on informant measures, where information on an individual's functioning

Table 51.2 Informant and objective measures of dementia for people with an intellectual disability (Strydom & Hassiotis, 2003; Zeilinger et al., 2013)

Informant		
Dementia Questionnaire for People with Learning Disability (DLD)	Evenhuis et al. (2007)	The *DLD*, an informant-based questionnaire, consists of 50 items and eight subscales. It measures specific cognitive deterioration as a result of dementia, as well as functional deterioration as a result of dementia severe sensory or psychiatric problems
IQ Code	Jorm (1994)	An informant tool that determines whether there is a decline in cognitive functioning
Dementia Scale for Down syndrome (DSDS)	Gedye (1995)	Test to determine the presence of dementia. Can only be administered by psychologists or psychometrists
Cambridge examination for Mental Disorders of the Elderly, modified for use for assessing people with Down syndrome (CAMDEX-DS)	Ball et al. (2004)	An informant assessment tool for diagnosis dementia in people with Down syndrome. Emphasis is placed on change from person's previous best of functioning
Dementia Screening Questionnaire for Individuals with intellectual Disability (DSQIID)	Deb et al. (2007)	The 53 items of the DSQIID cover areas such as loss of memory, confusion, loss of skills, social withdrawal, behavioural changes, psychological symptoms, physical symptoms, sleep logical symptoms, physical symptoms, sleep disturbance and speech abnormalities
Adaptive Behaviour Dementia Questionnaire (ABDQ)	Prasher et al. (2004)	15-item questionnaire to detect change in adaptive behaviour
Objective		
Test for Severe Impairment	Albert and Cohen (1992)	The Test for Severe Impairment (TSI) is a 24-item scale that measures a range of cognitive functions including motor performance, language production, language comprehension, memory, conceptualisation, and general knowledge
Down syndrome mental status examination (DSMSE)	Haxby (1989)	Objective measure of memory, orientation, language, visual perception, spatial ability, praxis
Neuropsychological Assessment of Dementia in Adults with intellectual Disability (NAID)	Crayton et al. (1998)	Simple battery of tests covering memory, orientation, language and praxis
Brief Praxis Test	Dalton and Fedor (1997)	The BPT is a 24-item non-verbal cognitive test. It requires simple behavioural responses with minimal language demands
Modified FULD object memory Test	Seltzer (1997)	Memory is one of the key changes in dementia. The Modified Fuld Object-Memory Test assesses recognition/naming and learning/recall. The test includes the use of objects rather than pictures to improve salience, and inclusion of multiple presentations as well as a recognition trial

over a period of time can be gathered by someone who knows the person well (McKenzie, Metcalfe, & Murray, 2018). In order to complete a dementia assessment in this population, it is recommended that that both objective and informant measures are used. Table 51.2 summarises some of the measures that have been found valid and useful for a population with an intellectual disability.

Evidence from Research

20-Year Follow-Up Study

In a longitudinal 20-year follow-up study in Ireland, at a specialist memory service for people with an intellectual disability, dementia assessments were completed for all 77 participants with Down syndrome on an annual basis. These assessments included the Test for Severe

Impairment (Albert & Cohen), The Down syndrome Mental State Examination (DSMSE) (Haxby, 1989), The Dementia Questionnaire for People with Learning Disability (DLD) (Evenhuis, Kengen, & Eurlings, 2007), and the Daily Living Skills Questionnaire (National Institute of Aging 1989). As of 2017, 75 of the 77 (97.4%) had developed dementia. Data is available on the annual cognitive assessments. The graph below shows scores on the four instruments 5 years prior to diagnosis and 5 years post diagnosis. An increase in scores on the DLD indicates lower level of functioning with DLD scores plotted on the secondary axis on the graph below (Fig. 51.2).

As can be seen on this graph, scores on the DLD show the earliest signs of decline, suggesting this is the most sensitive instrument used, with decline seen over a period of 5 years pre diagnosis. The DLSQ measures activities of daily living and identified changes 3–4 years prior to diagnosis. The TSI and DSMSE appeared less sensitive to change prior to diagnosis, (McCarron et al., 2017). When changes in scores were mapped by level of intellectual disability, greater change was seen for those with a moderate level of intellectual disability as compared to those with a severe level of intellectual disability; perhaps when scores are higher at baseline, greater change can be seen over time (McCarron et al., 2017)

IDS-TILDA

IDS-TILDA has cognitive scale data on 609 people who have completed the TSI for all three waves of the study to date. As is shown on the graph below, scores for those with a diagnosis of dementia in Wave 1 declined more over time than for those without a diagnosis, lending further confirmation to the sensitivity of the instrument (Fig. 51.3).

Neuroimaging and Assessment in People with Down Syndrome

There are findings that people with Down syndrome show accelerated ageing in the brain, with an adjusted brain predicted age increase of 7.69 years (Cole et al., 2017) and greater age related reduction in the frontal, temporal, and parietal lobes compared to the general ageing population (Beacher et al., 2009). The most consistent structural change seen early in Alzheimer's disease is atrophy of the temporal medial lobe. Neuroimaging studies have found that atrophy is present in the temporal medial lobe in people with Down syndrome even without a diagnosis if dementia and no normative values have been established for people with Down syndrome (British Psychological Society & Royal College of Psychiatrists, 2015). Some MRI studies with people with Down syndrome have shown signifi-

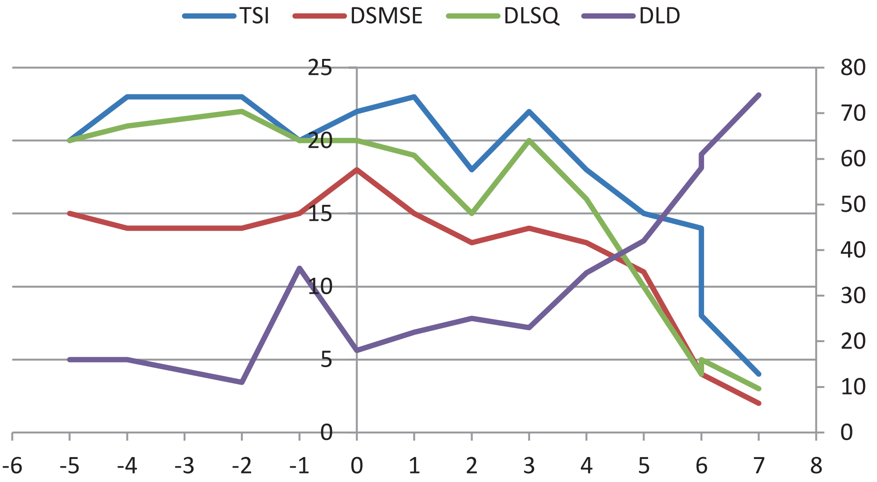


Fig. 51.2 Scores on cognitive assessments in 20-year follow-up study in people with Down syndrome

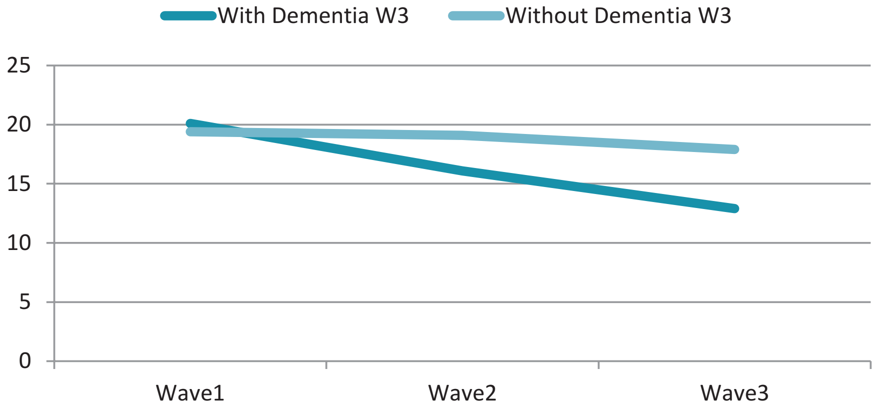


Fig. 51.3 Change in score in TSI for those with and without dementia diagnosis in IDS-TILDA

cant reduction in volume in the temporal lobe and hippocampus in individuals with Alzheimer's compared to those without Alzheimer's; however, the samples were small and those with Alzheimer's were significantly older than the controls without Alzheimer's (Kesslak, Nagata, Lott, & Nalcioglu, 1994; Pearlson et al., 1998).

In a more recent study, however, it was found that, when adjusted for head size, age and gender, volume size of the hippocampus, striatal structures and amygdala was significantly smaller in those with Down syndrome and Alzheimer's compared to those with Down syndrome without Alzheimer's. The volume of the hippocampus and the caudate nuclei (dorsal striatum) appeared to distinguish between people with Down syndrome with and without Alzheimer's with 90% specificity and 80% sensitivity (Beacher et al., 2009). This is an important finding but clinical use is limited, as cross sectional data does not address time course changes. Longitudinal studies are therefore needed to better distinguish brain changes associated with Alzheimer's from anomalies associated with Down syndrome (Neale, Padilla, Fonseca, Holland, & Zaman, 2018).

Co-morbidities

Co-morbidities in People with Down Syndrome

Frequency and severity of co-morbidities appear to be associated both with presence of Alzheimer's and with the stage of Alzheimer's in people with Down syndrome (McCarron, Gill, McCallion, & Begley, 2005). Among participants in the 20-year follow-up study in Ireland, co-morbidities were common (see Fig. 51.4) with vision impairment the most commonly reported, at 93.3%, and hearing impairment at 61.3%. Epilepsy was also common.

Epilepsy in People with Down Syndrome and Dementia

In the IDS-TILDA study, 77.9% of participants had epilepsy, and all participants with epilepsy had a diagnosis of dementia. Diagnoses of epilepsy and dementia were highly associated, and a diagnosis of epilepsy came on average 0.57 years after a diagnosis of dementia. This is in keeping with previous research on epilepsy and dementia in a population with Down syndrome. It has previously been reported that 80% of people with Down syndrome and dementia will develop epilepsy, and younger age of onset of dementia is associated with greater risk of seizures (Menéndez, 2005). Finding in the Irish longitudinal follow-up confirm these earlier reports.

Co-morbidities Common in All People with Intellectual Disability and Dementia

In addition to the co-morbidities found in the study above for people with Down syndrome, sleep disorders, gastrointestinal disorders, infec-

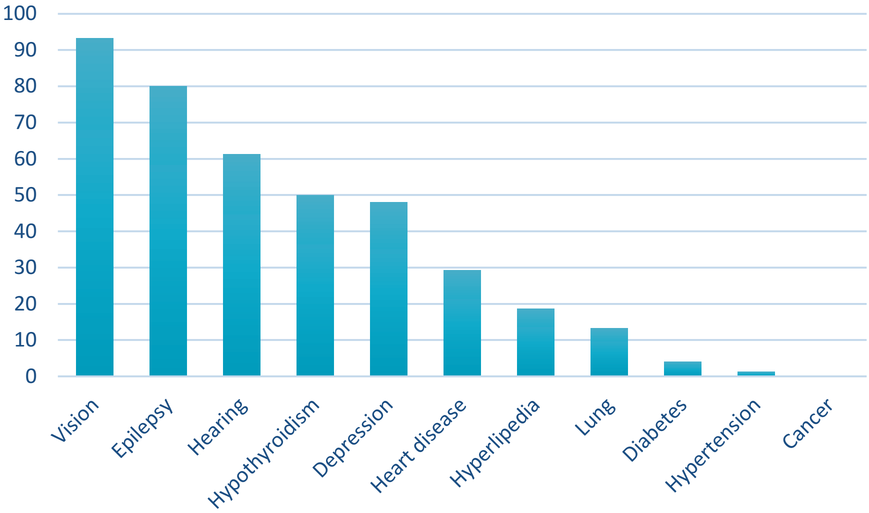


Fig. 51.4 Health co-morbidities of dementia in people with Down syndrome in 20-year follow-up study

tions and pain have been recognised as common co morbidities associated with dementia for people with an intellectual disability (British Psychological Society & Royal College of Psychiatrists, 2015).

It was been highlighted that pain can go unrecognised in this population. In one study examining staff knowledge and experience in working with people with intellectual disability and dementia, it was found that pain often went undetected and unmanaged, and where behaviours were assigned as "challenging" rather than associated with pain (Wilkinson, Kerr, & Cunningham, 2005).

Diagnosing pain in people with intellectual disability and dementia can also be challenging due to:

- Diagnostic overshadowing.
- Communication challenges.
- High turnover of staff.
- Carer beliefs about pain thresholds in people with ID.

(British Psychological Society & Royal College of Psychiatrists, 2015; McCarron et al., 2014b)

Post-diagnostic Support

Good quality post-diagnostic support is essential for people with intellectual disability who develop dementia. It is acknowledged that environmental features that do not meet the needs of older adults can trigger confusion and result in agitation and disruptive behaviours (Brawley, 2001). Also, in the general population, of the estimated $818 billion spent on dementia care, it is estimated that 85% of costs are related to family and social care rather than medical care (Livingston et al., 2017).

There are numerous definitions of post-diagnostic support where some place emphasis on planning, support, information and maintaining independence (World Health Organization, 2012), and others place more emphasis on psychosocial interventions (Watts, Cheston, Moniz-Cook, Burley, & Guss, 2013). The British Psychological Society focus the definition of post-diagnostic support on the person, rather than the disease and place emphasis on things that help the person to live well (British Psychological Society, 2015). This includes ensuring that the individual feels a sense of belonging to their family or community, and being able to contribute and participate meaningfully to their community. Post-diagnostic care refers to care from this time a dementia diagnosis is highly suspected through to end of life stage of dementia. Thus, there is a recognition also that any definition of post-diagnostic care needs to be flexible enough to adapt to the changing demands and needs of the individual as dementia progresses (Dodd et al., 2017).

The first stage of post-diagnostic support is helping the individual to understand and to come to terms with the diagnosis. As with all care for the individual with intellectual disability, the approach should be individualised and person centred, where the carer best known to the person and family members should lead in sharing information to share, and in understanding what the individual is experiencing (Jokinen et al., 2013; Tuffrey-Wijne & Watchman, 2014).

Stages of Dementia

Early Stage Dementia

In the early stages of dementia, subtle changes in behaviour and mood may be seen, with difficulties in memory for recent events, completing everyday tasks and learning new information, declines in social and daily living skills, and disorientation.

The emphasis at this stage is to support the individual to maintain their skills while gradually increasing support to the individual as needs change. This may also involve simplifying elements of support, such as communication, routines and tasks.

Suggested activities for this stage include arts and crafts, board games, discussion about people and places, use of visual planners (Jokinen et al., 2013; Watchman, 2014).

International guidelines recommend the use of non-pharmacological interventions at this stage, to deal behavioural or psychological symptoms of dementia; however, there is a lack of evidence base to suggest which non-pharmacological interventions to use (Kales, Gitlin, & Lyketsos, 2015).

Mid Stage Dementia

Mid stage dementia is characterised by further memory loss including of names of familiar people, declines in language, difficulties with self-care, personality or mood changes, problems with eating and drinking, disturbed sleep patterns and increased incontinence (British Psychological Society & Royal College of Psychiatrists, 2015).

The focus of care at this stage is to reduce stress and anxiety, interventions to preserve abilities while ensuring safety, comfort and dignity. Excessive demands on the individual should be reduced, with clear routines and with as little change as possible especially with respect to health and social care services (Dodd et al., 2017; Jokinen et al., 2013).

There should be a focus at this stage should focus on finding failure free activities; music therapy, aromatherapy, reminiscence, movement and exercise, one-step gardening such as watering plants, massage (Watchman, 2014).

Late Stage Dementia

Myoclonus and epilepsy are common in late stage dementia, incontinence is prevalent, along with loss of eating/drinking skills, difficulty recognising people, will require 24 h care, will be bedridden and there is greater risk of infections, particularly pneumonia (British Psychological Society & Royal College of Psychiatrists, 2015; McCarron et al., 2018).

Health monitoring is essential at this stage, with attention to weight, nutrition and hydration, continence, pain and epilepsy (British Psychological Society & Royal College of Psychiatrists, 2015; Jokinen et al., 2013).

Activities at this stage include smiling and laughing, singing, stroking, holding, gentle rocking (Watchman, 2014).

End of Life Care

Defining end of life is difficult in any situation, but is further complicated in people with an intellectual disability. Scales used in the general population such as the Global deterioration scale (Reisberg, Ferris, Leon, & Crook, 1982) or the Functional Assessment Staging Tool (FAST) scale (Reisberg, 1988) are of limited use with people with intellectual disability, as some people may not have had the stated capabilities to begin with. There are consensus recommendations that the FAST scale supplemented with findings of acute medical complications, recurrent infections, pneumonia, pressure sore and/or pressure sores is indicative that life expectancy is of 6 months or less, and care should move to specialised end of life care (McCallion et al., 2017).

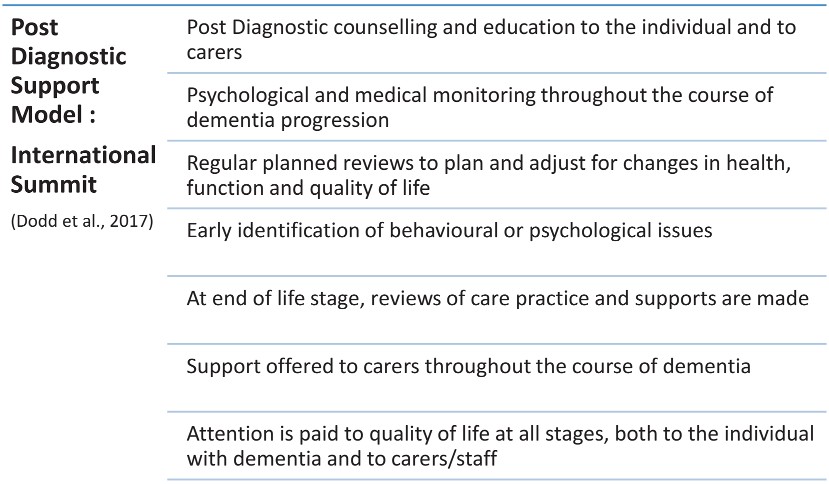
Understanding the characteristics of advanced dementia in people with Down syndrome and/or other ID is critical to understanding when transitions must be initiated from active curative care to supportive comfort care that is palliative care (McCarron et al., 2018). It is essential that person-centred approaches embedded in prior care are continued, and that there is genuine collaboration between ID services and hospice/palliative care with planning that involves the person with ID their families, other carers and their peers. The history and the voice of the person with an intellectual disability must continue to be heard (McCallion et al., 2017; McCarron et al., 2018).

A consensus statement of the international summit on intellectual disability and dementia recommended that post-diagnostic support needs to be individualised and adapted to the person's needs as dementia progresses (Dodd et al., 2017). This statement also highlighted that models of post-diagnostic support for people with an intellectual disability generally mirror those for the general population. The main differences noted were that many people with intellectual disability are part of a service prior to diagnosis, and thus post-diagnostic support tends to be an extension of this service, adapted to serve specific dementia needs. Specific consensus group recommendations (see Table 51.3) addressed care across the stages of dementia, regular reviews and attention to needs of carers.

It was also noted that because people with intellectual disability are often attached to formal services prior to the dementia diagnosis, approaches to post-diagnostic support tend to be more person centred than in general dementia care. Care goals are often to adapt the service that was already in place for the individual to ensure that changing needs are better supported (Dodd et al., 2017; McCallion et al., 2017; McCarron et al., 2018).

Supports and interventions are individualised and tailored to both the person's pre-existing level of intellectual disability and stage of dementia with an emphasis on the psychological wellbeing of the individual and the minimisation of the impact of changes for the individual with

Table 51.3 Model of post-diagnostic support for people with an intellectual disability and dementia



Post Diagnostic Support Model : **International Summit** (Dodd et al., 2017)	Post Diagnostic counselling and education to the individual and to carers
	Psychological and medical monitoring throughout the course of dementia progression
	Regular planned reviews to plan and adjust for changes in health, function and quality of life
	Early identification of behavioural or psychological issues
	At end of life stage, reviews of care practice and supports are made
	Support offered to carers throughout the course of dementia
	Attention is paid to quality of life at all stages, both to the individual with dementia and to carers/staff

dementia and their carers (British Psychological Society & Royal College of Psychiatrists, 2015; Jokinen et al., 2013).

These theories of post-diagnostic support have been brought together in a practical way in the Standards of dementia care (McCarron & Reilly, 2010) developed at the Daughters of Charity Service.

The Daughters of Charity Disability Support Services

The Daughters of Charity Disability Services provides a wide range of services to with intellectual disability in Ireland. A dementia specialist clinic was established to respond to

1. The changing demographics of the service users.
2. An ageing population with Down syndrome.
3. The need to restructure residential and day programmes.
4. The need to upskill staff at all levels of the organisation.
5. To address end of life care concerns.
   (McCarron, McCallion, Reilly, & Mulryan, 2014b)

The memory clinic is run by two clinical nurse specialists in dementia. Baseline screening for dementia is completed with all adults with Down syndrome over the age of 35 and with individuals with intellectual disability from other aetiologies from the age of 55. Annual screening is then completed, using a mixture of objective and informant measures.

In 2011, two purpose built dementia homes were built; an eight bedded home specifically to accommodate the needs of those with mid stage dementia and a six bedded home adapted to accommodate people with end stage dementia (McCarron & Reilly, 2010). The houses were designed to support the changing needs of individuals with intellectual disability across the continuum of dementia with features of the design selected to minimise stress, and to compensate for the loss of abilities with the aim of empowering the individual to use their remaining abilities to the greatest extent.

Features of the home include

- Familiar features and a homely style.
- Curved corridors with strategic seating to promote wandering and to cue to rest.
- Open plan kitchen to allow for involvement in cooking activities.
- Good signage and "cueing" with good visual access.
- Large central garden area to support gardening activities, found to be therapeutic.
- Supports unplanned activities between staff and service users, such as taking a walk in the garden.
- The ability for staff to be able to discreetly observe service users, without encroaching on their space.

Standards of Dementia Care

Within the context of a memory clinic, specialised residential facilities and ongoing support for ageing in place for many individuals with Down syndrome and dementia standards of dementia care were established to formalise the essential elements that are needed to provide a consistent quality approach to dementia care (McCarron & Reilly, 2010).

The six standards recognise the uniqueness of each person's experience of dementia (Fig. 51.5).

Standard 1: Appropriately Trained Staff and Service Development

It is essential that people with dementia receive care and support from staff that have sufficient and correct knowledge to care effectively. Education on dementia is also recommended for peers with an intellectual disability, which is tailored to their needs. Information is also offered to families of the individual with dementia in relation to dementia care.

Standard 2: Memory Assessment Service

All individuals with intellectual disability have access to baseline screening, following up assessment and timely diagnosis of dementia. The memory clinic model offers systematic screening, assessment, diagnosis, including a compre-

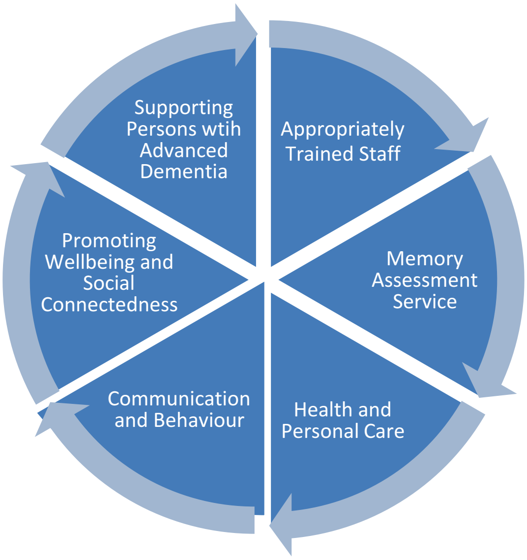


Fig. 51.5 Standards of dementia care

hensive diagnostic work-up and post-diagnostic support and treatment.

Standard 3: Health and Personal Care

Individuals are encouraged to continue with previous level of personal care to the greatest extent possible. Person centredness underpins all physical care activities with focus on dignity, privacy and the person's preferences. Optimum physical and mental care is essential using a multi-disciplinary approach and in partnership with the individuals with dementia, their carers, family and healthcare team.

Standard 4: Communication and Behaviour

Effective communication is essential in order to maintain the individual's personhood and well-being, and to minimise stress for the person with dementia. Non-cognitive symptoms that cause distress are assessed in a timely manner to alleviate aggravating factors. Every effort is made to support ageing in the home of choice for the individual, but that procedures are in place should the changing needs become too great where a change in living circumstances is recommended.

Standard 5: Promoting Well-Being and Social Connectedness

Relationships between the person with dementia and to those important to them are encouraged, and supported. Activities that reflect the person's lifelong interests are maintained as much as possible, and are adapted to support changing needs of the person.

Standard 6: Supporting Persons with Advanced Dementia: Addressing Palliative Care and End of Life Needs

Care is appropriate to the needs of the individual and adapted as necessary. Every effort is made to optimise the person's quality of life by providing the best end of life and palliative care. End of life plans take in to account the person's religious, cultural and spiritual needs. The family and staff who best know the person with dementia have the opportunity to discuss and partake in decisions relating to end of life.

Case Study of Best Practice in Dementia Care for individual with Down syndrome and Dementia

Paula∗ was born in 1962 with Down syndrome and a moderate intellectual disability. In her adult years, she lived in a community home with other people who had an intellectual disability within a large disability organisation in Dublin. Paula was an independent person. She mastered many home making skills, travelled independently, worked a few hours weekly in her local supermarket and had an active social life competing in swimming and bowling at Special Olympics both nationally and internationally.

The organisation Paula attended has a specialist Memory Clinic and in line with international best practice, Paula received a baseline neuropsychological screening at age 35 years. This screening entailed completing a variety of cognitive tests and with informants in her both community home and work placement, giving information on her functional, cognitive and social skills. The Clinical Nurse specialist (CNS) in dementia who completed the screening process documented in detail Paula's functional, cognitive and social abilities. Over the following years, these assessment scales were repeated and compared along with an overview of Paula's physical health profile.

In 2009 when Paula was aged 47 years, the first perceptible changes were noted at her annual screening. Staff in her home described that Paula was more irritable with her peers and she was more argumentative. They described "door banging" and episodes of weepiness. Paula had become more apathetic and was declining to participate in previously enjoyed activities. Staff were requested to seek a mental health review to rule out depression. There was no decline noted in Neuropsychological assessment scores and no change to functional skills. Staff had observed no changes to her short-term memory. Despite this, the CNS was alerted to the change in personality. Frontal temporal type changes are often first observed as prodromal dementia changes; however, it is vital at this stage to rule out environmental/mental health contributory factors as these can mimic early dementia type changes.

Over the following 18 months, the following changes were observed. Staff noted that they had to repeat their instructions more often and Paula started to require verbal prompts to keep her orientated to her weekly routine. She insisted on making her lunch for work at weekends and found it difficult to adapt to a change in routine. She misplaced her house keys and when something was misplaced, she blamed her peers, which escalated arguments within the home. In 2011, a consensus meeting with the Psychiatrist, CNS dementia and key support staff agreed the presentation typical of Alzheimer's type dementia in people with Down syndrome and a full physical work up to rule out differential diagnosis was undertaken. Following this work-up, a formal diagnosis of Alzheimer's dementia was made and a trial of cognitive medication was commenced.

Supports were put in place to enable Paula to continue to engage in her day-to-day activities of living and her diagnosis was shared with her in a way that was meaningful to her. The CNS spoke to her of having problems with her memory and encouraged her to use her diary and visual pictures to help keep her orientated to her daily activities. Family and staff also helped Paula to create her life story in a photo album and Paula found this very enjoyable. Staff training guided practice consistent with the Dementia Care standards (McCarron & Reilly, 2010).

The CNS continued to repeat the neuropsychological assessment tools to support Paula, her family, peers and staff through the stages of dementia.

In 2013, Paula had increased problems with her communication and she had great difficulty in expressing herself verbally. Her short-term memory was very impaired and she could not remember an instruction given moments before. Her cognitive test scores plummeted and she required support in all activities of daily living, dressing, showering and feeding. Staff provided an increased level of verbal and physical prompts and they endeavoured to support Paula to maintain each skill for as long as was possible. In this, the mid stage of dementia typical issues emerged as Paula tried to make sense of her world. With increased confusion and declining emotional control, she would hit out at peers and staff showing evidence of hyperacusis, an increased sensitivity to noise. Staff used her life biography to support their care practice and reminiscence provided a valuable tool for redirection when Paula became overwhelmed in day-to-day situations While supporting Paula to the extent possible in her existing home Paula was also introduced to a specialist home supporting people at mid stage dementia. In October 2013, she commenced respite there, initially for afternoon visits progressing to overnights and then weekly stays. This allowed a break for her peers who found it difficult living with their friend who had now become very vocal at times screaming at them for no apparent reason. It was important to engage her peers understanding of what was happening for Paula and through a series of peer support sessions using resources from Dementia Scotland the CNS offered education on supporting their friend with dementia.

Staff in her long-time home wished to continue to provide care for Paula but this became increasingly difficult when problems emerged with depth perception and mobility. Paula lived in a two storey home with no downstairs sleeping accommodation. In 2014, she started to have difficulty getting on and off the bus and staff was concerned in relation to her safety when using the stairs Family and key support people's views were considered and it was decided that the specialist home could best support her current and future care needs and a detailed transition plan was developed.

Behaviours that arose in her community home were less evident in specialist home for people with mid stage dementia and Paula was supported to maintain previously important relationships as is detailed in standard 5 of the dementia care standards.

In 2015, Paula developed new onset seizure activity. Despite being well managed through anticonvulsant medication, the development of seizure activity corresponded with a further decline in her dementia. Issues of incontinence and immobility became a further issue. Paula had access to an Occupational therapist, Physiotherapist, Speech and Language therapist and dietician and their combined skills provided expertise in managing her safety and comfort as dementia progressed. Paula's dementia assessment review pointed towards late stage dementia and as the year progressed, she had an increase in respiratory and urinary tract infections. Paula moved to a specialist home that supports the advanced palliative care needs of people with an advanced dementia in 2016. Its environment is quieter than her previous home and best supports the needs of a person with advanced dementia.

Advance care planning had been discussed with family and this conversation was further progressed when Paula moved to the specialist home for advanced dementia. Family, Paula's GP, CNS dementia and key support staff discussed what was in Paula's best interest and her prior expressed wishes in terms of supporting her health needs as her dementia further progressed. Resources from the Irish Hospice foundation and palliative care guidelines were used as a framework for discussions. Family had built a relationship of trust with Paula's care professionals and all decisions were made supporting a palliative approach to care.

In June 2017 surrounded by family and friends, Paula passed away peacefully in this home. Her life was celebrated and her loving memory lives on.

∗Real name not used.

References

Albert, M., & Cohen, C. (1992). The test for severe impairment: An instrument for the assessment of patients with severe cognitive dysfunction. *Journal of the American Geriatrics Society, 40*, 449–453.

Alzheimer's Association. (2018). 2018 Alzheimer's disease facts and figures. *Alzheimer's & Dementia, 14*, 367–429.

American Psychiatric Association. (2013). *Diagnostic and statistical manual of mental disorders (DSM-5®)*. Washington, DC: American Psychiatric Association.

Aylward, E., Burt, D., Thorpe, L. U., Lai, F., & Dalton, A. (1997). Diagnosis of dementia in individuals with intellectual disability. *Journal of Intellectual Disability Research, 41*, 152–164.

Ball, S., Holland, A., Hon, J., Huppert, F., Treppner, P., & Watson, P. (2006). Personality and behaviour changes mark the early stages of Alzheimer's disease in adults with Down's syndrome: Findings from a prospective population-based study. *International Journal of Geriatric Psychiatry, 21*, 661–673.

Ball, S., Holland, A., Huppert, F., Treppner, P., Watson, P., & Hon, J. (2004). The modified CAMDEX informant interview is a valid and reliable tool for use in the diagnosis of dementia in adults with Down's syndrome. *Journal of Intellectual Disability Research, 48*, 611–620.

Bayer, A. J. (2018). The role of biomarkers and imaging in the clinical diagnosis of dementia. *Age and Ageing, 47*(5), 641–643.

Beacher, F., Daly, E., Simmons, A., Prasher, V., Morris, R., Robinson, C., … Murphy, D. (2009). Alzheimer's disease and Down's syndrome: An in vivo MRI study. *Psychological Medicine, 39*, 675–684.

Brawley, E. C. (2001). Environmental design for Alzheimer's disease: A quality of life issue. *Aging & Mental Health, 5*, 79–83.

British Psychological Society. (2015). *Dementia and people with intellectual disabilities: Guidance on the assessment, diagnosis, interventions and support of people with intellectual disabilities who develop dementia*. Leicester: British Psychological Society.

British Psychological Society & Royal College of Psychiatrists. (2015). *Dementia and people with intellectual disabilities: Guidance on the assessment, diagnosis, interventions and support of people* *with intellectual disabilities who develop dementia*. Leicester: British Psychological Society.

Burt, D. B., & Aylward, E. H. (2000). Test battery for the diagnosis of demntia in individuals with intellectual disability. *Journal of Intellectual Disability Research, 44*, 175–180.

Butterfield, D. A., Di Domenico, F., Tramutola, A., Head, E., & Perluigi, M. (2017). Oxidative stress and mTOR activation in Down syndrome brain: Roles in Aβ42 and Tau neuropathology and transition to Alzheimer disease-like dementia. *Free Radical Biology and Medicine, 112*, 70.

Carr, J., & Collins, S. (2018). 50 years with Down syndrome: A longitudinal study. *Journal of Applied Research in Intellectual Disabilities., 31*(5), 743–750.

Cole, J. H., Annus, T., Wilson, L. R., Remtulla, R., Hong, Y. T., Fryer, T. D., … Menon, D. K. (2017). Brain-predicted age in Down syndrome is associated with beta amyloid deposition and cognitive decline. *Neurobiology of Aging, 56*, 41–49.

Cooper, S.-A. (1997). High prevalence of dementia among people with learning disabilities not attributable to Down's syndrome. *Psychological Medicine, 27*, 609–616.

Coppus, A., Evenhuis, H., Verberne, G. J., Visser, F., Van Gool, P., Eikelenboom, P., & Van Duijin, C. (2006). Dementia and mortality in persons with Down's syndrome. *Journal of Intellectual Disability Research, 50*, 768–777.

Cosgrave, M. P., Tyrrell, J., McCarron, M., Gill, M., & Lawlor, B. A. (2000). A five year follow-up study of dementia in persons with Down's syndrome: Early symptoms and patterns of deterioration. *Irish Journal of Psychological Medicine, 17*, 5–11.

Crayton, L., Oliver, C., Holland, A., Bradbury, J., & Hall, S. (1998). The neuropsychological assessment of age related cognitive deficits in adults with Down's syndrome. *Journal of Applied Research in Intellectual Disabilities, 11*(3), 255–272.

Cummings, J. L., Mega, M., Gray, K., Rosenberg-Thompson, S., Carusi, D. A., & Gornbein, J. (1994). The Neuropsychiatric Inventory comprehensive assessment of psychopathology in dementia. *Neurology, 44*, 2308–2308.

Dalton, A. J., & Fedor, B. L. (1997). *Dyspraxia scale for adults with Down syndrome*. New York State: Institute for Basic Research in Developmental Disabilities.

Deb, S., & Braganza, J. (1999). Comparison of rating scales for the diagnosis of dementia in adults with Down's syndrome. *Journal of Intellectual Disability Research, 43*(5), 400–407.

Deb, S., Hare, M., Prior, L., & Bhaumik, S. (2007). Dementia screening questionnaire for individuals with intellectual disabilities. *The British Journal of Psychiatry, 190*(5), 440–444.

Dekker, A. D., Strydom, A., Coppus, A. M., Nizetic, D., Vermeiren, Y., Naudé, P. J., … De Deyn, P. P. (2015). Behavioural and psychological symptoms of dementia in Down syndrome: Early indicators of clinical Alzheimer's disease? *Cortex, 73*, 36–61.

Dodd, K., Watchman, K., Janicki, M. P., Coppus, A., Gaertner, C., Fortea, J., … Strydom, A. (2017). Consensus statement of the International Summit on Intellectual Disability and Dementia related to post-diagnostic support. *Aging & Mental Health*, 1–10.

Evenhuis, H. M., Kengen, M. M., & Eurlings, H. A. (2007). *Dementia questionnaire for people with learning disabilities*. Pearson Assessment.

Feldman, H. H., Jacova, C., Robillard, A., Garcia, A., Chow, T., Borrie, M., … Chertkow, H. (2008). Diagnosis and treatment of dementia: 2. Diagnosis. *Canadian Medical Association Journal, 178*, 825–836.

Finkel, S. I. (2001). Behavioral and psychological symptoms of dementia: A current focus for clinicians, researchers, and caregivers. *The Journal of Clinical Psychiatry., 21*, 3–6.

Gedye, A. 1995. Dementia scale for Down syndrome: Manual.

Grady, C. L. (2008). Cognitive neuroscience of aging. *Annals of the New York Academy of Sciences, 1124*, 127–144.

Hardy, J. A., & Higgins, G. A. (1992). Alzheimer's disease: The amyloid cascade hypothesis. *Science, 256*, 184.

Haxby, J. V. (1989). Neuropsychological evaluation of adults with Down's syndrome: Patterns of selective impairment in non-demented old adults. *Journal of Intellectual Disability Research, 33*, 193–210.

Head, E., Lott, I. T., Hof, P. R., Bouras, C., Su, J. H., Kim, R., … Cotman, C. W. (2003). Parallel compensatory and pathological events associated with tau pathology in middle aged individuals with Down syndrome. *Journal of Neuropathology & Experimental Neurology, 62*, 917–926.

Head, E., Lott, I. T., Wilcock, D. M., & Lemere, C. A. (2016). Aging in Down syndrome and the development of Alzheimer's disease neuropathology. *Current Alzheimer Research, 13*, 18–29.

Holland, A. J., Hon, J., Huppert, F. A., Stevens, F., & Watson, P. (1998). Population-based study of the prevalence and presentation of dementia in adults with Down's syndrome. *The British Journal of Psychiatry, 172*, 493–498.

Jennings, D., Seibyl, J., Sabbagh, M., Lai, F., Hopkins, W., Bullich, S., … Stephens, A. (2015). Age dependence of brain β-amyloid deposition in Down syndrome An [18F] florbetaben PET study. *Neurology, 84*, 500–507.

Jokinen, N., Janicki, M., Keller, S., McCallion, P., & Force, L. (2013). National Task Group on Intellectual Disabilities and Dementia Care Practices. Guidelines for structuring community care and supports for people with intellectual disabilities affected by dementia. *Journal of Policy and Practice in Intellect Disabilities, 10*, 1–24.

Jorm, A. (1994). A short form of the Informant Questionnaire on Cognitive Decline in the Elderly (IQCODE): Development and cross-validation. *Psychological Medicine, 24*, 145–153.

Kales, H. C., Gitlin, L. N., & Lyketsos, C. G. (2015). Assessment and management of behavioral and psychological symptoms of dementia. *BMJ, 350*, h369.

Kesslak, J., Nagata, S., Lott, I., & Nalcioglu, O. (1994). Magnetic resonance imaging analysis of age-related changes in the brains of individuals with Down's syndrome. *Neurology, 44*, 1039–1039.

Lai, F., & Williams, R. S. (1989). A prospective study of Alzheimer disease in Down syndrome. *Archives of Neurology, 46*, 849–853.

Lamar, M., Foy, C. M., Beacher, F., Daly, E., Poppe, M., Archer, N., … Simmons, A. (2011). Down syndrome with and without dementia: an in vivo proton magnetic resonance spectroscopy study with implications for Alzheimer's disease. *NeuroImage, 57*, 63–68.

Lautarescu, B. A., Holland, A. J., & Zaman, S. H. (2017). The early presentation of dementia in people with Down syndrome: A systematic review of longitudinal studies. *Neuropsychology Review, 27*, 31–45.

Livingston, G., Sommerlad, A., Orgeta, V., Costafreda, S. G., Huntley, J., Ames, D., … Cohen-Mansfield, J. (2017). Dementia prevention, intervention, and care. *The Lancet, 390*, 2673–2734.

Mattis, S. (1988). *Dementia rating scale (DRS)*. Odessa, FL: Psychological Assessment Resources.

McCallion, P., Hogan, M., Santos, F. H., McCarron, M., Service, K., Stemp, S., … Watchman, K. (2017). Consensus statement of the International Summit on Intellectual Disability and Dementia related to end-of-life care in advanced dementia. *Journal of Applied Research in Intellectual Disabilities, 30*, 1160–1164.

McCallion, P., & McCarron, M. (2004). Ageing and intellectual disabilities: A review of recent literature. *Current Opinion in Psychiatry, 17*, 349–352.

McCarron, M., Gill, M., McCallion, P., & Begley, C. (2005). Health co-morbidities in ageing persons with Down syndrome and Alzheimer's dementia. *Journal of Intellectual Disability Research, 49*, 560–566.

McCarron, M., & Lawlor, B. (2003). Responding to the challenges of ageing and dementia in intellectual disability in Ireland. *Aging & Mental Health, 7*, 413–417.

McCarron, M., McCallion, P., Coppus, A., Fortea, J., Stemp, S., Janicki, M., & Watchman, K. (2018). Supporting advanced dementia in people with Down syndrome and other intellectual disability: Consensus statement of the International Summit on Intellectual Disability and Dementia. *Journal of Intellectual Disability Research., 62*(7), 617–624.

McCarron, M., Haigh, M., McCallion, P., McCallion, P., Carroll, R., Burke, E., … O'Dwyer, M. (2017). *Health, Wellbeing and Social Inclusion: Ageing with an Intellectual Disability in Ireland*. Dublin: School of Nursing and Midwifery, Trinity College Dublin.

McCarron, M., McCallion, P., Reilly, E., Dunne, P., Carroll, R., & Mulryan, N. (2017a). A prospective 20-year longitudinal follow-up of dementia in persons with Down syndrome. *Journal of Intellectual Disability Research, 61*, 843–852.

McCarron, M., McCallion, P., Reilly, E., & Mulryan, N. (2014a). A prospective 14-year longitudinal follow-up of dementia in persons with Down syndrome. *Journal of Intellectual Disability Research, 58*, 61–70.

McCarron, M., McCallion, P., Reilly, E., & Mulryan, N. (2014b). *Responding to the challenges of service development to address dementia needs for people with an intellectual disability and their caregivers. Intellectual disability and dementia: Research into practice* (pp. 241–269).

McCarron, M., & Reilly, E. (2010). *Supporting persons with intellectual disability and dementia: Quality Dementia Care Standards. A guide to practice.* San Antonio: Daughters of Charity Service.

McKenzie, K., Metcalfe, D., & Murray, G. (2018). A review of measures used in the screening, assessment and diagnosis of dementia in people with an intellectual disability. *Journal of Applied Research in Intellectual Disabilities., 31*(5), 725–742.

McKhann, G. M., Knopman, D. S., Chertkow, H., Hyman, B. T., Jack, C. R., Kawas, C. H., … Mayeux, R. (2011). The diagnosis of dementia due to Alzheimer's disease: Recommendations from the National Institute on Aging-Alzheimer's Association workgroups on diagnostic guidelines for Alzheimer's disease. *Alzheimer's & Dementia: The Journal of the Alzheimer's Association, 7*, 263–269.

Menéndez, M. (2005). Down syndrome, Alzheimer's disease and seizures. *Brain and Development, 27*, 246–252.

Mukaetova-Ladinska, E., Harrington, C., Roth, M., & Wischik, C. (1993). Biochemical and anatomical redistribution of tau protein in Alzheimer's disease. *The American Journal of Pathology, 143*, 565.

Nasreddine, Z. S., Phillips, N. A., Bédirian, V., Charbonneau, S., Whitehead, V., Collin, I., … Chertkow, H. (2005). The Montreal Cognitive Assessment, MoCA: A brief screening tool for mild cognitive impairment. *Journal of the American Geriatrics Society, 53*, 695–699.

National Institute of Aging, Laboratory of Neurosciences (1989). *The daily living skills questionnaire*. National Institute of Aging, US.

Neale, N., Padilla, C., Fonseca, L. M., Holland, T., & Zaman, S. (2018). Neuroimaging and other modalities to assess Alzheimer's disease in Down syndrome. *NeuroImage: Clinical, 17*, 263–271.

O'Caoimh, R., Clune, Y., & Molloy, D. (2013). Screening for Alzheimer's disease in Downs syndrome. *Journal of Alzheimers Disease and Parkinsonism*, 2161–0460.

Pearlson, G. D., Breiter, S. N., Aylward, E. H., Warren, A. C., Grygorcewicz, M., Frangou, S., … Pulsifer, M. B. (1998). MRI brain changes in subjects with Down syndrome with and without dementia. *Developmental Medicine and Child Neurology, 40*, 326–334.

Prasher, V., Farooq, A., & Holder, R. (2004). The Adaptive Behaviour Dementia Questionnaire (ABDQ): screening questionnaire for dementia in Alzheimer's disease in adults with Down syndrome. *Research in Developmental Disabilities, 25*(4), 385–397.

Prasher, V., & Krishnan, V. (1993). Age of onset and duration of dementia in people with Down syndrome: integration of 98 reported cases in the literature. *International Journal of Geriatric Psychiatry, 8*, 915–922.

Prasher, V., Sajith, S., Mehta, P., Zigman, W., & Schupf, N. (2010). Plasma β-amyloid and duration of Alzheimer's disease in adults with Down syndrome. *International Journal of Geriatric Psychiatry, 25*, 202–207.

Prince, M., Wimo, A., Guerchet, M., Ali, G., Wu, Y. & Prina, M. 2017. World Alzheimer Report 2015—The global impact of dementia: An analysis of prevalence, incidence, cost and trends. London, Alzheimer's Disease International (ADI).

Reisberg, B. (1988). Functional assessment staging (FAST). *Psychopharmacology Bulletin, 24*, 653–659.

Reisberg, B., Auer, S. R., & Monteiro, I. M. (1997). Behavioral pathology in Alzheimer's disease (BEHAVE-AD) rating scale. *International Psychogeriatrics, 8*, 301–308.

Reisberg, B., Ferris, S. H., de Leon, M. J. & Crook, T. 1982. The Global Deterioration Scale for assessment of primary degenerative dementia. The American Journal of Psychiatry, 139(9), 1136–1139.

Roizen, N. J., & Patterson, D. (2003). Down's syndrome. *The Lancet, 361*, 1281–1289.

Rowe, J., Lavender, A., & Turk, V. (2006). Cognitive executive function in Down's syndrome. *British Journal of Clinical Psychology, 45*, 5–17.

Royal College of Psychiatrists. (2001). *DC-LD: Diagnostic criteria for psychiatric disorders for use with adults with learning disabilities/mental retardation*. Berlin/Heidelberg: Springer Science & Business.

Satizabal, C. L., Beiser, A. S., Chouraki, V., Chêne, G., Dufouil, C., & Seshadri, S. (2016). Incidence of dementia over three decades in the Framingham Heart Study. *New England Journal of Medicine, 374*, 523–532.

Schupf, N., Zigman, W., Tang, M.-X., Pang, D., Mayeux, R., Mehta, P., & Silverman, W. (2010). Change in plasma Aβ peptides and onset of dementia in adults with Down syndrome. *Neurology, 75*, 1639–1644.

Seltzer, G. B. (1997). *Modified FULD object memory evaluation*. Madison, WI: Waisman Centre, University of Wisconsin.

Selkoe, D. J. (1991). The molecular pathology of Alzheimer's disease. *Neuron, 6*, 487–498.

Sinai, A., Mokrysz, C., Bernal, J., Bohnen, I., Bonell, S., Courtenay, K., … Hillier, R. (2017). Predictors of age of diagnosis and survival of Alzheimer's disease in Down syndrome. *Journal of Alzheimer's Disease*, 1–12.

Strydom, A., Chan, T., Fenton, C., Jamieson-Craig, R., Livingston, G., & Hassiotis, A. (2013). Validity of criteria for dementia in older people with intellectual disability. *The American Journal of Geriatric Psychiatry, 21*, 279–288.

Strydom, A., Chan, T., King, M., Hassiotis, A., & Livingston, G. (2013). Incidence of dementia in older adults with intellectual disabilities. *Research in Developmental Disabilities, 34*, 1881–1885.

Strydom, A., & Hassiotis, A. (2003). Diagnostic instruments for dementia in older people with intellectual disability in clinical practice. *Aging & Mental Health, 7*, 431–437.

Strydom, A., Hassiotis, A., King, M., & Livingston, G. (2009). The relationship of dementia prevalence in older adults with intellectual disability (ID) to age and severity of ID. *Psychological Medicine, 39*, 13–21.

Strydom, A., Shooshtari, S., Lee, L., Raykar, V., Torr, J., Tsiouris, J., … Sinnema, M. (2010). Dementia in older adults with intellectual disabilities—Epidemiology, presentation, and diagnosis. *Journal of Policy and Practice in Intellectual Disabilities, 7*, 96–110.

Strydom, A., Livingston, G., King, M., & Hassiotis, A. (2007). Prevalence of dementia in intellectual disability using different diagnostic criteria. *British Journal of Psychiatry, 191*(2), 150–157.

Teipel, S. J., & Hampel, H. (2006). Neuroanatomy of Down syndrome in vivo: A model of preclinical Alzheimer's disease. *Behavior Genetics, 36*, 405–415.

Theofilas, P., Ehrenberg, A. J., Nguy, A., Thackrey, J. M., Dunlop, S., Mejia, M. B., … Suemoto, C. K. (2018). Probing the correlation of neuronal loss, neurofibrillary tangles, and cell death markers across the Alzheimer's disease Braak stages: A quantitative study in humans. *Neurobiology of Aging, 61*(1–12).

Tuffrey-Wijne, I., & Watchman, K. (2014). Sharing the diagnosis of dementia: Breaking bad news to people with intellectual disabilities. *Journal of Applied Research in Intellectual Disabilities, 27*, 357.

Tully, J., Schirliu, D., & Moran, M. (2012). Application of DC-LD to an intellectual disability population. *Advances in Mental Health and Intellectual Disabilities, 6*, 259–264.

Watchman, K. (2014). Towards understanding individual experiences of people ageing with an intellectual disability and dementia. In *Intellectual Disability and Dementia: Research into Practice* (p. 126). London: Jessica Kingsley.

Watts, S., Cheston, R., Moniz-Cook, E., Burley, C., & Guss, R. (2013). Post-diagnostic support for people living with dementia. *Clinical Psychology in the Early Stage Dementia Care Pathway*, 1–13.

Wilkinson, H., & Janicki, M. (2002). The Edinburgh principles with accompanying guidelines and recommendations. *Journal of Intellectual Disability Research, 46*, 279–284.

Wilkinson, H., Kerr, D., & Cunningham, C. (2005). Equipping staff to support people with an intellectual disability and dementia in care home settings. *Dementia, 4*, 387–400.

Wimo, A., Guerchet, M., Ali, G.-C., Wu, Y.-T., Prina, A. M., Winblad, B., … Prince, M. (2017). The worldwide costs of dementia 2015 and comparisons with 2010. *Alzheimer's & Dementia: The Journal of the Alzheimer's Association, 13*, 1–7.

World Health Organization. (2004). *ICD-10: International statistical classification of deseases and related health problems*. Geneva: WHO.

World Health Organization. (2012). *Dementia: A public health priority*. Geneva: World Health Organization.

Zeilinger, E. L., Stiehl, K. A., & Weber, G. (2013). A systematic review on assessment instruments for dementia in persons with intellectual disabilities. *Research in Developmental Disabilities, 34*, 3962–3977.

Zigman, W. B., Schupf, N., Devenny, D. A., Miezejeski, C., Ryan, R., Urv, T. K., … Silverman, W. (2004). Incidence and prevalence of dementia in elderly adults with mental retardation without Down syndrome. *American Journal on Mental Retardation, 109*, 126–141.

Part XI

Health and Social Behaviors

52

Issues of Sexuality and Relationships

Nathan J. Wilson, Patsie Frawley,
Dilana Schaafsma, Amie O'Shea,
Callista K. Kahonde, Vanessa Thompson,
Judith McKenzie, and David Charnock

Background

Sexuality in the lives of people with intellectual disability is almost always conflated with sexual abuse, sexual behaviours, sexual knowledge and questions about capacity to "be" sexual. Rarely is sexuality discussed in a more holistic way that acknowledges pleasure, desire, identity and "self-authored" sexual expression. Writers like Michael Gill (2015) suggested this is due to sexual ableism which he defined as "the system of imbuing sexuality with determinations of qualification to be sexual based on criteria of ability, intellect, morality, physicality, appearance …" (p. 3). Through this lens, sexuality in the lives of people with intellectual disability is mediated by ideas about capacity and competence, assumptions of desirability and overshadowed by a discourse of risk and vulnerability. Foley (2017) reported that underpinning this discourse is a "paternalistic regime" whereby the sexual lives of people with intellectual disability are strongly surveilled, often by parents or other caregivers. He described this regime as being played out where people with intellectual disability, despite their chronological adulthood, "either must ask permission and/or are prevented by their parents from taking control over their social/sexual lives" (p. 6).

Lost in this discourse are opportunities to consider sexuality as a humanly intrinsic part of life and a right for people with intellectual disability as it is for their peers who do not have the

---

N. J. Wilson (✉)
School of Nursing and Midwifery, Western Sydney University, Penrith, NSW, Australia
e-mail: N.Wilson@westernsydney.edu.au

P. Frawley
Deakin University, Geelong Waterfront Campus, Locked Bag 20001, Geelong, VIC, Australia
e-mail: patsie.frawley@deakin.edu.au

D. Schaafsma
Fontys University of Applied Sciences, Eindhoven, AH, The Netherlands
e-mail: d.schaafsma@fontys.nl

A. O'Shea
Deakin University, Geelong Waterfront Campus, Locked Bag 20001, Geelong, VIC, Australia
e-mail: amie.oshea@deakin.edu.au

C. K. Kahonde
Stellenbosch University, Cape Town, South Africa
e-mail: ckkahonde@sun.ac.za

V. Thompson
NSW Sexology Services, Newcastle, NSW, Australia
e-mail: vanessathompson@nswsexologyservices.com.au

J. McKenzie
Disability Studies Division, Department of Health and Rehabilitation Sciences, University of Cape Town, Cape Town, South Africa
e-mail: Judith.mckenzie@uct.ac.za

D. Charnock
Faculty of Medicine and Health Sciences, Queens Medical Centre, The University of Nottingham, Nottingham, UK
e-mail: David.charnock@nottingham.ac.uk

J. L. Matson (ed.), *Handbook of Intellectual Disabilities*, Autism and Child Psychopathology Series, https://doi.org/10.1007/978-3-030-20843-1_52

restrictive label of intellectual disability. Michael Perlin, a noted scholar in disability and human rights law, noted that "One of the most controversial social policy issues that remains dramatically under-discussed in scholarly literature is the sexual autonomy of persons with psychosocial and intellectual disabilities" (Perlin & Lynch, 2015, p. 10). The absence of a codified right to "be sexual" in society's most progressive and unifying international law on the rights of people with intellectual disability, the United Nations Convention on the Rights of Persons with Disabilities (CRPD) speaks loudly about the way sexuality is positioned in relation to other rights.

Evidence from reviews of the CRPD committee's deliberations about sexuality indicate that a sense of "moral panic" from conservative religious and cultural nations has ensured sexual rights are not included in the Convention (Ruiz, 2017; Schaaf, 2012). Although a number of the articles in the Convention give a nod to sexuality, no single article exists asserting this right, therefore there is no requirement for signatory nations to focus their attention on how to progress this right. While Article 23 asserts the right to "form a family", there is no recognition of how you might do this by being sexual. Likewise, although Article 25 refers to sexual health there is no clear reference to the link between "being sexual" and sexual health. With the CRPD lacking this clear reference to sexuality rights signatory nations can easily overlook sexuality and sexuality rights in their domestic laws and policies. It is important to recognise this absence and its implications as Article 4 of the Convention requires that signatory nations "engage themselves to develop and carry out policies, laws and administrative measures for securing the rights recognized in the Convention and abolish laws, regulations, customs and practices that constitute discrimination" (UN General Assembly, 2007). Rather than the Convention providing a template for the progression of sexual rights for signatory countries, there is greyness about the place of sex in the lives of people with disabilities.

Sexual health is referred to in the CRPD, where Article 25 notes State Parties shall "Provide persons with disabilities with the same range, quality and standard of free or affordable health care and programmes as provided to other persons, including in the area of sexual and reproductive health and population-based public health programmes" (UN General Assembly, 2007). The inclusion of sexual health is important as it acknowledges, albeit secondarily, that people with disabilities are sexual and that their sexual health along with physical and mental health is a significant health area. Very little research is available about the sexual health status of people with intellectual disability with reports generally referring to "disability" more broadly. The research that is available suggests that people with disabilities are experiencing rates of STIs and HIV/AIDS in particular at higher rates than the general population (WHO/UNFPA, 2009).

A report by WHO/UNFPA in 2009 cited the link between poverty and disability as a significant factor in suboptimal sexual health outcomes for people with disabilities (WHO/UNFPA, 2009). This report suggested the need for an intersectional approach to understanding and responding to sexual health in the lives of people with intellectual disability. It makes the following five recommendations for addressing sexual health:

1. Establish partnerships with people with disabilities to address sexual health,
2. Raise awareness and increase accessibility in mainstream health and sexual health services,
3. Ensure all sexual and reproductive health programmes reach and serve persons with disabilities,
4. Address disability in national sexual and reproductive health policy, laws, and budgets, and
5. Promote research on sexual and reproductive health of persons with disabilities at local, national and international levels (p. 1).

This framework provides guidance to signatory countries to the CRPD in how to work towards recognising the right to optimal and equal sexual health for people with disabilities, but it requires that national and international disability advocacy continues to pick this issue up and progress it.

For example, in Australia, the peak women's disability advocacy organisation Women with Disabilities Australia (WWDA) have taken the lead in this area. In their latest work they reported on research they undertook with women with disabilities about access to health services where sexual health was a key focus. In addition to issues of general inaccessibility reported in this research, the report makes an important point about the need for an acknowledgement of the sexuality of people with intellectual disability to ensure access to sexual health services. In this research they also found that gender was a key site of discrimination in relation to accessing sexual health services. This is captured strongly by one woman who noted "My doctor refused to believe me about my sex life" (Women with Disabilities Australia (WWDA), 2018 p. 7).

Many people with intellectual disability live at home with family caregivers, who themselves face a range of issues in supporting access to sexual health supports and services for their family member who has intellectual disability. It is important to understand these issues and to be aware of the underlying issues families deal with. As Foley (2017) noted, the way families see and respond to the sexuality of their family member with intellectual disability are informed by their paternal/maternal frame of protection.

Family Caregiver Roles—Risk Versus Protection

The role of families in supporting the sexuality of their family members with intellectual disability has only received minimal attention in research, practice and theorisation. What has been researched shows that the issue of sexuality is fraught with challenges for families at conceptual, moral and practical levels and that these challenges are similar across cultural contexts (O'Neill, Lima, Thomson Bowe, & Newall, 2015; Rogers, 2010). For example, family caregivers, especially parents, sometimes struggle to accept and understand that their adult children with intellectual disability have sexual needs as their focus tends to be more about the adult's limitations and the moral and practical responsibilities of ensuring the safety of their adult child.

The continued dependency of people with intellectual disability on family caregivers raises conflicts for the family caregivers of how much protection and how much support to give to the family member with intellectual disability in the area of sexuality (Dupras & Dionne, 2014). For example, Foley (2013) described the mothers in his study as "reluctant jailors" in order to illustrate the tension of trying to create a balance between their children's need for sexual empowerment and the maternal desire to offer protection from harm. Although Foley (2013) only sought the views of ten mothers, the quandary faced by these mothers is a common experience (Pownall, Jahoda, Hastings, & Kerr, 2011; Rushbrooke, Murray, & Townsend, 2014).

The responsibilities and emotional attachment that comes with such a caring relationship often pushes family caregivers towards a more conservative outlook as compared to paid caregivers, professionals and other people who do not have that day-to-day responsibility (Aunos & Feldman, 2002; Brown & Pirtle, 2008; Rushbrooke et al., 2014). For example, an Irish study by Evans, McGuire, Healy, and Carley (2009) compared the attitudes of paid and family (i.e. unpaid) caregivers to the sexuality of people with intellectual disability and found that the attitudes of paid caregivers aligned more closely with the rights-based approach. In a recent South African study, although family caregivers had some conceptual understanding of sexuality as a human rights issue, it appeared much more limited and conservative (Kahonde, 2016). These attitudes, in turn, impact upon the type of sexuality education that the family member with intellectual disability may, or may not, receive.

When comparing the family caregivers' perceived role as sexuality educators for their children with and without intellectual disability, there is evidence that the way the family caregivers perceive the cognitive capabilities of their child with intellectual disability impacts on the nature and extent of any sexuality education they provide (Pownall et al., 2011). For example, Pownall, Jahoda, and Hastings (2012) in Scotland

and Gürol, Polat, and Oran (2014) in Turkey reported similar findings that although mothers recognised the need to provide sexuality education for their children with intellectual disability, they did not provide that sexuality education. One major issue is the limited access to education support about sexuality for family caregivers of people with intellectual disability, in particular more explicit sexual activities and expression (Gardiner & Braddon, 2009; Rojas, Haya, & Lazaro-Visa, 2016). By limiting access to education and support for aspects such as intimacy, sexual intercourse, sex for pleasure and procreation, does the person with intellectual disability a disservice which may be attributed to the disablist myth of asexuality. That is, the conception by family caregivers of their sons/daughters with intellectual disability as not capable of, or not interested in, the more intimate aspects of sexuality (Dupras & Dionne, 2014; Pownall et al., 2011). Further to this, family caregivers have also been reported as trying not to "wake sleeping dogs" where the fear is held that providing any sexuality education may inadvertently make their children aware of "new" sexual knowledge that may lead to added problems for the caregiver (Kahonde, 2016).

Family caregivers also identified a lack of skills and knowledge as one of the reasons that prohibit them from providing people with intellectual disability with the necessary supports and education for sexuality (Aunos & Feldman, 2002; Isler, Beytut, Tas, & Conk, 2009; O'Neill et al., 2015). This lack of knowledge and information is even common in high-income countries that, arguably, one would assume to have more progressive disability policies and more accessible services (O'Neill et al., 2015; Wilson & Frawley, 2016). Those family caregivers who do provide sexuality education, seem to do it selectively and with a primary focus on protecting their family member with intellectual disability rather than supporting them to develop friendships and to underpin the formation of healthy relationships. Hence, early intervention in supporting the family caregivers and their specific concerns about sexuality and relationship formation is vital.

Supporting People with Intellectual Disability to Form Meaningful Relationships

Having meaningful relationships and finding someone to share life experiences with are important aspects for our perceived quality of life. Additionally, being able to develop meaningful sexual relationships are an important aspect to our sexual health (World Health Organization, 2010). Unfortunately, many people with intellectual disability do not have the opportunities or experience to develop these relationships and get little help from others to form and maintain relationships. Some studies show that people with intellectual disability experience loneliness more so than those without a disability (Fulford & Cobigo, 2016; Gilmore & Cuskelly, 2014). The social networks of people with intellectual disability are also known to be small and consist mostly of family members or professional caregivers (Van Asselt-Goverts, Embregts, & Hendriks, 2013). Where people with intellectual disability do have friends, some report they do not see them very often (McVilly et al. 2006). In research with people with intellectual disability on relationships, to the question, "Why are relationships important?" many people answered that they want somebody to share their life with and don't want to be alone (Schaafsma, Kok, Stoffelen, & Curfs, 2017). Arguably this is the same response you would expect from anyone who has been asked this question; however, for people with intellectual disability the pursuit of such relationships can be difficult, and loneliness can ensue.

Experiences of loneliness can have detrimental effects on one's quality of life. There are two types of loneliness, social and emotional loneliness. Social loneliness arises when a person feels that their social network is inadequate. People have the need to belong to different groups of people, with each group having their own role and function. Friends, for example, fulfil a different role than colleagues. Emotional loneliness is felt when a person lacks intimate relationships, in other words, when a person does not have anyone they can discuss important issues with or when a person has a lack of physical intimacy with other people (De Jong Gierveld, 1998). Having meaningful relationships also helps to improve one's self-esteem and self-confidence with low self-esteem linked to a range of psychopathology, such as depression, anxiety and eating disorders (Bos, Huijding, Muris, Vogel, & Biesheuvel, 2010; Fulford & Cobigo, 2016). Other benefits of having meaningful relationships that were mentioned by people with intellectual disability are having access to practical and emotional support (Fulford & Cobigo, 2016) and having a "normal identity", which for some is associated with a sense of pride (Bates, Terry, & Popple, 2016).

Therefore, in order to counter loneliness, it is important to not only increase the number of people in one's social network and make sure there is diversity, but also improve the quality of these relationships. Although the characteristics of having intellectual disability may mean difficulties with social skills and social communication, they also face added barriers to making contact with friends on their own and getting to know another person through conversation and social contact. Barriers to developing an intimate relationship can include the negative attitudes of caregivers towards these relationships, leading to restrictive rules (e.g. not allowed to have somebody sleep over) and a lack of privacy; especially for those living in supervised residential settings. Further, a lack of money can limit social contact and has associated barriers including access to transport to spend time developing these relationships (Fulford & Cobigo, 2016).

In the quest of finding a partner, studies have shown that *similarity* is important, meaning that relationship success is related to having things in common. For example, enjoying the same activities or having a similar level of intellectual disability (Fulford & Cobigo, 2016). However, this is not always straightforward. For example, some people with intellectual disability have reported not being attracted to people who have a "visible" disability, such as people who use a wheelchair (Bates et al., 2016). Also, dating agencies for people with intellectual disability have reported some challenges with matching people, partly due to the unrealistic expectations of one partner in the relationship regarding who they see themselves having an intimate relationship with. Some people with intellectual disability have also reported not wanting to date another person with intellectual disability or indeed a person who has a greater degree of intellectual disability than themselves (Fulford & Cobigo, 2016; Schaafsma, n.d.). Other issues for people looking for heterosexual relationships includes that more males than females are looking for relationships, and for people whose lives are closely supported by caregivers and family there are issues for getting the right kind of support for dating and developing intimate relationships.

Having meaningful relationships including intimate relationships is good for our well-being, but people with intellectual disability experience many barriers to this. In order for them to succeed, support by family or professional caregivers is vital. They need help in learning how to make contact with other people, practical support such as being driven to a date or meeting, and support when they do manage to find somebody they enjoy spending time with. For many people with intellectual disability, family caregivers can have the primary role in providing support for developing a range of relationships including intimate relationships. Gender and sexual orientation of the person with intellectual disability can play a major part in the issues thy face getting the support they need.

Issues Facing Women and Girls with Intellectual Disability

Research on the lived experiences of women with intellectual disability has repeatedly concluded that gender and disability cannot be separated, emphasising the importance of gender in understanding people's lives and experiences (McCarthy, 2014; Traustadóttir, 2006). For women and girls with intellectual disability, this includes their access to and experience of health, particularly gynaecological health, and their gendered experiences of sexuality and sexual autonomy. In addition, they face the same gendered disparities in health and gendered messages about sexuality as other women, while somehow also being seen as inherently "unlike" other women.

Research in the area of gynaecological health has shown that women with intellectual disability have lower rates of participation in preventative screening such as cervical screening and breast screening, in numbers often markedly below women without intellectual disability (Cobigo et al., 2013). This research documents the often "extremely limited knowledge" of women with intellectual disability, high anxiety about women's health screening including mammography and a general lack of understanding about women's health (Truesdale-Kennedy, Taggart, & McIlfatrick, 2011).

Menstruation is the subject of many myths for women with intellectual disability, including the idea that they will inevitably have problems with menstrual care, including hygiene, increased discomfort during menstruation and premenstrual stress (Gomez, Carlson, & Van Dooren, 2012). Research with young women with intellectual disabilities confirms that menstruation and management of it is a strong focus of sexuality education for them (Frawley & Wilson, 2016) and clearly the management of menstruation has underpinned the management of reproduction. The way that reproductive rights have been managed for women with disability, including sterilisation–often performed involuntarily–is well documented in the research and oral history literature (e.g. Tilley, Walmsley, Earle, & Atkinson, 2012). Initially it was ideas from eugenics that enabled the control and management of women's reproductive rights. The same arguments are now presented as being in the "best interests" of the woman and/or the carers who support her; that involuntary sterilisation continues today demonstrates how the lives and rights of women with intellectual disability remain surrounded by a "roaring silence" (Tilley et al., 2012).

In research with women with intellectual disability they have highlighted: the importance of intimate romantic relationships (e.g. Rushbrooke et al., 2014); experiencing barriers to their sexual autonomy through implementation of service policies by staff and surveillance by family members (e.g. McCarthy, 2014; O'Shea, 2016); issues relating to knowledge of and choice about contraception (McCarthy, 2009, 2010), and a lack of health information and treatment for menopause (McCarthy, 2002). Where sexual autonomy is achieved, it is something that women fear losing as they get older. As Rojas et al. (2016) noted, the attitudes of others about women and girls' sexuality are significant because of the corollary between restrictive views and how they can hinder women and girls achieving their rights. Further, the messages presented to women with intellectual disability by disability service staff are informed by traditional gendered stereotypes and while there may be a focus on factual information there is no attention given to pleasure, desire or intimacy (Bernert, 2011).

There is evidence that gender can present a positive and productive frame for negotiating desired identities and relationships for women with intellectual disabilities (O'Shea, 2016). However most of the research paints an unhappy picture, where women report predominantly "negative perceptions, negative experiences, thwarted ambitions and abuse that dominate the narratives of the women [who spoke about their lives]" (McCarthy, 2014, p. 124). Women with intellectual disability have also echoed traditional gender beliefs, particularly concerning homophobia, limited and exclusively heterosexual sexual experiences (which were viewed with fear both of the act and of negative consequences), and overwhelmingly negative stories of disconnection and dissatisfaction with sexuality (Fitzgerald & Withers, 2013).

Motherhood is one of the strongest gendered roles available to women, in most cultures it defines contemporary ideals of womanhood and femininity. Many women with intellectual disability have reported their desire to become mothers. A review of the history of sterilisation of women with intellectual disabilities found that this is a hope most often thwarted (Tilley et al., 2012). Women with intellectual disability who do become mothers are significantly marginalised with compulsory removal of their children common. Even if children are not immediately removed, the difficulty in accessing effective maternal and child health and support services and the often high levels of surveillance mothers with an intellectual disability experience have resulted in further isolation and disempowerment for them. The effects of removal of their children are felt deeply and have resulted in significant, longstanding grief and turmoil for many women with intellectual disability.

Issues Facing Men and Boys with Intellectual Disability

Men and boys with intellectual disability face unique gendered issues concerning their sexuality. This is not only due to the greater prevalence of intellectual disability in males when compared to females at a ratio of approximately 3:2, but the reality that the majority of caregivers—paid and unpaid—who provide support and care to people with intellectual disability, are female. This specifically gendered dynamic, although a usually tacit and common occurrence across the globe, has received scant attention in the research literature by practitioners and applied researchers. In addition, unlike the area of women's health and intellectual disability, there are very few, if any, well-known international researchers who are leading a male intellectual disability research agenda looking at health and wellbeing. This limited gendered focus means that there is not a wide body of work about men and boys with intellectual disability that can be drawn upon in the pursuit of evidence-based interventions (Merrick, Morad, & Carmeli, 2014).

The most common area of research focus reported in the literature is about problematic male sexual behaviour (Wilson, Parmenter, Stancliffe, Shuttleworth, & Parker, 2010). Specifically, this problematized research covers socially inappropriate sexual behaviour (Wheeler & Jenkins, 2004), public masturbation (Hingsburger, 1994), prescription of anti-libidinal medication (Sajith, Morgan, & Clarke, 2008), and unwanted touching of female paid caregivers (Thompson, Clare, & Brown, 1997). There are also a few smaller areas of focus including the increased prevalence of hypogonadism (McElduff & Beange, 2003), testicular cancer and undescended testes (Sasco et al., 2008), and one paper that reports a much greater incidence of STIs in a sample of Dutch males with intellectual disability (van Schrojenstein Lantman-de valk, 2005). Given the ageing of the population of people with intellectual disability, the lack of any research about prostate screening represents a major gap that, conceivably, will become more prominent in the years ahead.

In an attempt to chart a more meaningful and positive approach to the sexuality of men and boys with intellectual disability—rather than further reifying the dominant problematized discourse—an Australian study reported that the expression of their sexuality is a combination of the individual's developmental stage and the gendered influence of caregivers (Wilson, Parmenter, Stancliffe, & Shuttleworth, 2011). That is, sexuality and sexual expression is a healthy biological imperative that is often and reluctantly regulated by caregivers who are forced to act as interactional gatekeepers in a service delivery context where policy guidance is severely limited. Missing are guidelines and interventions that provide staff with a framework of supports guiding them on what to do in order to better support men with intellectual disability. For example, in this Australian study female caregivers reported not knowing what to do when changing the incontinence pad for a man with intellectual disability or showering a male with an erection. Rather than a common sense guideline in how to respond to such normative sexual expression, the issue becomes a problem for the male with intellectual disability, thus pathologising their sexuality even further. A different example involved a UK study where adult males with intellectual disability who accessed pornography, were stopped from doing so as it upset a female paid caregiver (Yacoub & Hall, 2009).

Research focusing on masculinity in the lives of boys and men with intellectual disability has identified that external control is often applied in relation to the embodiment and expression of gendered identity (Charncok & Standen, 2013; Wilson, Parmenter, Stancliffe, & Shuttleworth, 2013). Together this small body of research stresses the difficulties experienced across the spectrum of intellectual disability in specialist residential and educational settings. Furthermore, the research highlights the similarities and differences for boys and men at different points on the spectrum, emphasising the need for a focused approach to the critical study of men and masculinity for this group.

Wilson et al. (2013) in their ethnographic study, concerned with the lives of both boys and men living in three group home settings staffed predominately by female carers, describe difficulties with the construct of a masculine identity when the individual has a significant intellectual disability. Data from interviews with carers, participant observation and the collection of relevant artefacts created the concept of *Conditionally Masculine*, which suggests both normalised notions of changing masculinity and change that is conditional on the severity of disability and on the position of workers to the performance of masculinity. The researchers identified the concept to help recognise that significant levels of intellectual disability can be associated with acquired disadvantage in relation to a developing gendered identity. In essence, disadvantage was an accepted feature of being a boy or man if an intellectual disability was present. However, Wilson et al. (2013) argued that *Conditionally Masculine* was identified as neither an entirely negative or positive concept, but provides hope that its further investigation will be helpful in establishing a valued construct of masculinity for boys and men with intellectual disability.

In contrast, the study by Charncok and Standen (2013) explored the development of culturally normative ideas of masculinity for teenage boys in a special educational setting. In this study, the voice of the boys themselves gives shape to their lives and offers an insight into their hopes for the future. This revealed a complex tapestry of both restriction and emancipation, as the boys begin to face the prospect of life in a gendered world and a developing understanding of the effect of their impairment on this. The analysis of the data builds a theoretical model for *the ways we do intellectual disability boy to men*, underpinned by the boy's desire to be like other boys. The model asks those working with boys with intellectual disability to adopt a more critical approach to the unquestioning acceptance of normalised constructs of masculinity. In doing so workers are asked to be mindful of: the changes experienced when making the transition from being a teenager to becoming an adult man; the ideals held as exemplars by boys with intellectual disability with regard to masculinity; boys experience of masculinity in the world; and vicarious positions, observing the lives of boys and men without intellectual disability. In parallel to this the findings warn against ignorance of the effect of impairment in the guise of unachievable hope, as this may magnify vulnerability in this population.

LGBTIQ Issues Experienced by People with Intellectual Disability

People with intellectual disability who are lesbian, gay, bisexual, trans∗, intersex, queer, or questioning (LGBTIQ) have the same desire for intimate sexual and romantic relationships as their heterosexual peers (McClelland et al., 2012; Stoffelen, Kok, Hospers, & Curfs, 2013). Nevertheless, LGBTIQ people with intellectual disability face added regulation of their sexuality from others. For example, existing conservative attitudes about sexuality held by paid caregivers and families of people with intellectual disability, are heightened further when it comes to non-normative sexuality or gender expression (McCann, Lee, & Brown, 2016; Stoffelen et al., 2013; Taylor Gomez, 2012; Wilson et al., 2016). The research literature identifies a number of things that LGBTIQ people with intellectual disability have said is important to them including being listened to and having the opportunity to talk with others about sexuality and relationships in a safe and personal way that respects their wishes and their sexuality (Bane, Deely, Donohoe, & Wolfe, 2012; Fitzgerald & Withers, 2013; Kijak, 2013; Stoffelen et al., 2013; Wheeler, 2007). They have also articulated the need for ongoing support to maintain relationships, to understand themselves in their relationships and to overcome the inevitable relationship difficulties that everyone faces (Brown & McCann, 2018). One source of appropriate support may come from accessing mainstream LGBTIQ support services such as advocacy and counselling (Azzopardi-Lane & Callus, 2015; Friedman, Arnold, Owen, & Sandman, 2014).

LGBTIQ people with intellectual disability have also reported barriers in relation to their sexuality, including a lack of privacy and their rights being ignored in their own homes by caregivers, access to transport and the ability to travel independently (Bernert, 2011; Brown & McCann, 2018; Wheeler, 2007). Safety and privacy at home is a particularly important right for everyone, but for many people with intellectual disability, home is also a place where heteronormative attitudinal barriers can be the most significant (Stoffelen et al., 2013). For example, in a Canadian study, LGBTIQ participants with intellectual disability noted how other people—particularly paid caregivers and parents, but also social workers, counsellors, foster parents, and siblings "had control over their personal time and space to explore their sexuality, whether with themselves or others" (McClelland et al., 2012, p. 814). Participants also reported that because sex at home was forbidden, they sought sexual experiences and opportunities outside the home and in a range of public places. Despite their view that home was one of the most comfortable and safe locations for sexual practices (and where, for example, condoms were the more easily and reliably accessible), participants continued to engage in potentially unsafe sexual practices, which then involved more risk-taking, less physical safety and privacy, reduced opportunity for healthy decision making and unprotected sex (McClelland et al., 2012, pp. 816–817). Consequently, caregiver-driven sexual rules intended to increase safety (i.e. disallowing sex at home, with the assumption that extends to sex in all places) can backfire and end up contributing to increased risk particularly for marginalised groups (see also Cambridge, 1996; Johnson, Hillier, Harrison, & Frawley, 2001).

Sexual Abuse and Sterilisation

Rarely is sexuality and intellectual disability discussed without some mention of sexual abuse. It is widely understood that sexual abuse is about power and control intersecting with gender inequality, and in the lives of people with intellectual disability additionally intersecting with myths about asexuality and a damaged identity or personhood. Researchers since the 1990s have urged us to see the sexual abuse of people with intellectual disability more systemically or ecologically, rather than focusing on the behaviours or characteristics of the individual. Sobsey (1994), Brown and Turk (1994) and Fitzsimons (2009) have highlighted that higher risk of sexual abuse for people with intellectual disability is intrinsically linked to the attitudes perpetrators of abuse hold about intellectual disability that diminishes their humanness and thereby devalues them as people and as people with sexual agency. Prevention of abuse starts with establishing equality of humanness and personhood and changing social and institutional structures that oppress, isolate and "other" people. For people with intellectual disability this means dismantling ideas about "other, special, extraordinary and incapable" and replacing them with ideas about value, equality, worth and agency; sexual abuse cannot be prevented by restricting sexual rights including reproductive rights or privacy or restricting sexual expression.

For many women with intellectual disability, their lived experiences reflect that high levels of abuse and negative experiences in relation to their reproductive rights and parenting are too common. It could be argued that there is a need for a more holistic discussion about sexuality for women with intellectual disabilities, one that takes account of gender, sexual identity, sexual agency and rights. However, the seriousness of reproductive decisions being taken out of their hands and the high rates of sexual abuse warrants a continued focus, albeit perhaps with an intersectional lens that can be used to see these two issues within a more holistic frame, in particular looking at the intersection of gender and disability.

It is difficult to quantify the incidence and prevalence of sexual abuse and of reproductive rights violations for women with intellectual disability. Research suggests that these experiences are under-reported by women with intellectual disability and that "official" data are hard to locate. This is due in part to the often "hidden" nature of abuse and sterilisation and relates to the idea of an "extraordinary sexuality" that exists for people with intellectual disability overviewed earlier in this chapter. Canadian research by Desjardins (2012) argued that there is in fact a "new extraordinary sexuality" or an "adapted sexuality". This extraordinary and adapted sexuality sees sexual expression without the "risk of reproduction" and that takes place under the surveillance of others as "safe" for people with intellectual disability. There are significant risks in aligning these ideas with safety when research has shown that a history of sterilisation—to avoid reproduction—has not stopped abuse and that within living environments where people's lives are watched carefully, abuse has still occurred. Although involuntary sterilisation was meant to be a practice from the past, it is evident that many family caregivers globally still support the sterilisation of people with ID, especially girls and women. This is engendered by both fear of the girls and women with ID falling pregnant and avoiding the challenges of managing menstrual periods of the women with ID (Kahonde, 2016; Tilley et al., 2012; World Health Organization, 2011). Also alarming are narratives of males with intellectual disability being sterilised, particularly those who are being chemically sterilised by anti-libidinal medication that can have a devastating long-term impact on the health and wellbeing of the male.

Cultural Considerations

Research about the intersection of sexuality, intellectual disability and culture is scant and any discussion of this intersection needs to take into account the dominance of research from the global North. Although there is a growing body of literature from low and middle income countries (Adnams, 2010; Aldersey, 2012), the preponderance of research still emanates from high income settings (McKenzie, McConkey, & Adnams, 2013). This globally unrepresentative research raises at least three issues:

1. The impact of poverty on achieving desirable outcomes for people with intellectual disability is not well understood.
2. Different cultural understandings of disability, family, community and what it means to have a good life are underexplored.
3. The rich contribution of different cultures to our overall understanding of the phenomenon is not drawn upon.

Each of these aspects has a particular impact upon our understanding of sexuality of people with intellectual disability. As regards to poverty, Emerson (2007) has noted the dual and cumulative effect of a lack of access to health services for people with intellectual disability limits the economic participation of family members. In these circumstances the pressing needs for food, shelter, and freedom from abuse become the focus with sexuality an after-thought (Kahonde, McKenzie, & Wilson, 2018). Although evident globally, responding to the sexuality of people with intellectual disability in a manner that overemphasises their vulnerability, becomes even more intensified when the family lives in poverty and within a community where the risk of abuse is heightened (Kahonde, 2016). Unfortunately, it is often true that poverty and increased risk within the community restricts independent activity, such as going to the shop or meeting with friends, and the fears of risk of abuse take precedence.

In studies with families of people with intellectual disability in several African contexts it is apparent that family caregivers perceive disability in multiple ways (Chataika & McKenzie, 2013). There is a traditional perspective that sees intellectual disability as a consequence of a transgression of societal norms (such as a woman being unfaithful to her husband). Such a perception leads to stigmatization and isolation of both the mother and the disabled child. This often intensifies their bond and makes the mothers fierce protectors and advocates for their children's rights, often in the absence of fathers who bow to family and social pressure and abandon both mother and child. Caregivers in these tightly knit relationships frequently draw on spiritual sources to gain the necessary strength in this often-lonely task. Mothers describe their child as a gift from God, who has entrusted this child to their care as part of their spiritual journey and who will give them the strength to fulfil this destiny (Ingstad, 1995). This has the effect that people with intellectual disability are cloaked in an aura of innocence and being close to God in ways that prohibit their engagement in sexual activity. The gendered nature of care poses particular problems for male children whose sexuality is perceived as threatening and not the domain of mothers who expect fathers to educate their sons in the meaning of masculinity and male sexuality (Kahonde, 2016). This is most evident when mothers face the dilemma of how their male child with intellectual disability will participate (or not) in male rites of passage.

Perhaps the most overlooked contextual consideration is that of cultural practices that provide potential for greater participation and social inclusion of people with intellectual disability. The African context is a collectivist form of conducting social interaction. That is, the individual is not alone in making their decisions and needs to consult and consider the impact of their decisions on the family and community. This is most clearly expressed in the notion of Ubuntu which recognises that we are only human beings in our relation to others (Mabovula, 2011). Interdependence is at the heart of this philosophy and remains gendered with roles clearly demarcated for different members of society. This has the, as yet unrealized, potential of distributing care and support across the community. However, this is hampered by the stigma that is attached to disability. In terms of sexuality there are signs of how this might be seen differently. Protective mothers in a study by Kahonde (2016) spoke about how they would want their son or daughter to get married or to have children so that the care that they provide could be continued by a family member of a different generation. These notions of extending care to another generation is also reflected in research about the forced marriage of people with intellectual disability in some Asian societies (e.g. Clawson & Fyson, 2017).

In some patriarchal societies, males with intellectual disability are given more support in the area of sexuality than females (Gürol et al., 2014; Morales, Lopez, & Mullet, 2011). Research evidence to support this from studies in Turkey and Mexico reported that the mothers were more supportive of sexuality education for boys than for girls (Gürol et al., 2014) or acceptance of sexual relationships by the family was higher for males with ID than for females (Morales et al., 2011). On the other hand, cultural and religious beliefs around issues like masturbation and contraception may be hindrances blocking people with intellectual disability from freely expressing and enjoying their sexuality (Kahonde, 2016; Lafferty, McConkey, & Simpson, 2012). The fear of the person with intellectual disability being ostracised by society may result in them being disallowed from expressing their sexual identity.

Sexual Knowledge

Adequate and accurate sexual knowledge enables people to identify and ask for what is sexually pleasurable for them. It also allows them to be better able to set healthy sexual boundaries which are essential for safer sex (Abraham & Sheeran, 1994). Conversely, low sexual knowledge has been found to be linked to higher rates of sexually transmissible infections, higher numbers of unplanned pregnancy and more negative feelings about sex (Healy, McGuire, Evans, & Carley, 2009; Lafferty et al., 2012). Many people with intellectual disability have fundamental gaps in their sexual knowledge (Jahoda & Pownall, 2014). It is proposed that sexual knowledge assessment tools should be used with people with intellectual disability to identify gaps in sexual knowledge and that these identified gaps should be addressed through individually tailored sexual health educational programs (Bell & Cameron, 2003; Galea, Butler, Iacono, & Leighton, 2004).

Several studies have utilised pre- and post-sexual knowledge assessments to demonstrate an increase in the sexual knowledge of people with intellectual disability following their participation in a sexual health education program (Murphy, Powell, Guzman, & Hays, 2007).

Many people with intellectual disability have difficulties with communication and comprehension that affect their ability to understand and answer questions, including questions about sexual knowledge. For example, when compared to standard assessments designed for the general population, the responsiveness of people with intellectual disability increased more than three-fold when assessment questions with simplified wording designed specifically for people with intellectual disability were used (Stancliffe, Wilson, Bigby, Balandin, & Craig, 2014). Therefore, to aid in the appropriate assessment of the sexual health knowledge, it is critical that assessment tools have been developed specifically for people with intellectual disability. This approach is needed so that gaps in knowledge can be identified accurately, rather than the person being unable to explain what they know simply because they could not understand the question (Thompson, Stancliffe, Wilson, & Broom, 2016a; Thompson, Stancliffe, Wilson, & Broom, 2016b).

There have been several sexual knowledge assessment tools which have been developed specifically for people with intellectual disability including the Sexual Knowledge, Experience and Needs Scale for People with Intellectual Disability (SexKen-ID) (McCabe, 1994), the Human Relations and Sexuality Knowledge and Awareness Assessment (Family Planning Victoria, 1997), the Assessment of Sexual Knowledge (ASK) (Butler, Leighton, & Galea, 2003), the Socio-Sexual Knowledge and Attitudes Assessment Tool-Revised (SSKAAT-R) (Griffiths & Lunsky, 2003) and the General Sexual Knowledge Questionnaire (GSKQ) (Talbot & Langdon, 2006). All of these sexual knowledge assessment tools are similar in content and structure.

In their current form, sexual knowledge assessment tools contribute to the pathologising of sexual health of people with intellectual disability (Thompson et al., 2016b). Positive aspects of sexual health such as pleasure and sexual aides are not covered by the tools. Yet, content on sexual pathology has been systematically included in the tools. Indeed, the GSKQ (Talbot & Langdon, 2006) was specifically developed to test for counter deviance of men who display sexually offending behaviours and the SSKAAT-R (Griffiths & Lunsky, 2003) was used by its developers to assess the sexual knowledge of men with intellectual disability who sexually offend (Lunsky, Frijters, Griffiths, Watson, & Williston, 2007). They concluded that there was a "need to assess sexual knowledge, sexual attitudes, and prior sex education when an individual commits a sexual offence" (Lunsky et al., 2007, p. 80). Yet, the SSKKAAT-R has not been validated to assess risk of recidivism or to plan for the management of people with intellectual disability who sexually offend or display problematic sexual behaviours.

Several studies have focused on the development of sexual knowledge assessment tools (e.g. Galea et al., 2004), but only one study has focused on their use in a clinical setting. A recent study by Thompson et al. (2016b) found that although clinicians stated that such tools are needed to support the work they do in relation to sexual health of people with intellectual disability, that these tools also have several limitations. These included: missing important sexual health topics (such as libido and sexual pleasure), not enough questions to adequately determine level of knowledge in relation to each of the topic areas, complex wording of items, not enough pictures to support the questions, not enough guidance on how to administer the tools, and the tools are outdated (the latest tool is over a decade old). It has also been identified that sexual knowledge assessment tools are not able to be used for people with more severe levels of intellectual disability as they were too difficult for them to comprehend (Bell & Cameron, 2003). The study by Thompson et al. (2016a) reported that if the person with intellectual disability does not have their sexual knowledge assessed then they generally do not receive any sexual health provision at all. They also reported that sexual knowledge is typically only assessed when there has been an incident of problematic sexualised behaviour, typically perceived to be displayed by males with intellectual disability. This reactive approach perpetuates and further reifies a pathological sexual health discourse for people with intellectual disability.

Sex Education

Traditional sex education has been mainly focused on knowledge transfer. Popular topics include reproduction, puberty, contraception and sexually transmitted infections, that is, a focus on bodily changes, how babies are made and what happens if you do not practice safe sex. Nowadays we understand that in order to live a sexually healthy life, sex education should focus on a much broader array of areas to include topics such as sexual diversity, dating and how to have pleasurable sex. Developing a sex education program for people with intellectual disability poses an extra challenge in relation to their problems with learning and generalizing knowledge and skills to real-life situations (Bruder & Kroese, 2005; Schaafsma et al., 2014). Therefore, special attention needs to be given to how to increase knowledge and improve relevant skills in people with intellectual disability and provide them with opportunities to practice these skills in real-life settings. Tailoring sex education to a person's individual needs is thus necessary, but requires some skills from the person who delivers the sex education. In addition, sexuality is still a taboo topic and often professionals do not know how to talk about sexuality in a common-sense manner (Abbott & Burns, 2007), rather it is often in response to a problem (Schaafsma et al., 2014)

So why is it so difficult to talk about sexuality? One explanation could be that talking about sexuality feels too personal and embarrassing. People do not separate private sexuality (e.g. personal sexual experiences and attitudes) and professional sexuality (e.g. general information and support). A second explanation might be that sex education is associated with coitus and condom use. People often do not realize that topics such as relationships and dating are also important. These are usually easier topics to talk about. A third explanation could be that people (unconsciously) fear that talking about sexuality might lead to problems such as negative experiences (including sexual abuse) and (unwanted) pregnancies. Finally, a fourth explanation might be that both family and professional caregivers do not really know what they should and should not talk about. In other words, where their responsibility lies and when they should involve an expert?

People involved in the care of people with intellectual disability should be aware of three types of sex education: the spontaneous conversation, the goal-oriented conversation and the specialist conversation. The first type, the spontaneous conversation about sexuality is instigated by everyday situations. Something might happen on television or something might be mentioned during dinner. One should utilize these kinds of opportunities to educate and normalize the conversation about sexuality-related topics. It can also be a good way to test the knowledge of a person with intellectual disability or inquire about their experiences. It is important that the person providing this type of education realises they do not have to be all knowing when it comes to the topic of sexuality. Being about the level of expertise on the topic should not be a reason not to talk about sexuality.

The second type is the goal-oriented conversation. We know that people with intellectual disability have lower levels of sexual knowledge than peers without intellectual disability, experience sexual abuse at higher rates than people without intellectual disability and experience barriers forming relationships (Fulford & Cobigo, 2016; Murphy & O'Callaghan, 2004). With this in mind, we know that more goal-oriented sex education is needed. This means that, next to spontaneous conversation, we should also provide more systematic education. Two questions should underpin this type of education: what does a person with intellectual disability want to learn and what do they need to learn? What a person needs to learn is linked to what a person needs to know and be able to do in order to experience sexuality in a healthy and pleasurable way.

Examples of areas that need to be included are forming relationships, enjoyable and safe sex and information about respectful relationships and abuse prevention. The third type is specialist sex education. When a person with ID has experienced sexual abuse or expresses concerning sexual behaviour, one should always involve a specialist. These kinds of issues are beyond regular goal-oriented sex education.

There exist a number of sex education materials available for people with intellectual disability, however, their quality differs. Most do not have a theory and/or evidence base such as a description of how the materials are meant to improve the sexual health of people with intellectual disability and nor is evidence provided on why the materials are effective in improving sexual health (Schaafsma, Kok, Stoffelen, & Curfs, 2016; Schaafsma, Stoffelen, Kok, & Curfs, 2013). Nevertheless, an improvement can be seen in this area as more and more programs are starting to provide a detailed description of the goals of the materials, the methods included in the materials and the evidence base (see for example (Frawley et al., 2017, Frawley, Barrett, & Dyson, 2012). Topics that are covered in some more recent and comprehensive program are: rights, respectful relationships, sexual diversity, parenthood, pleasurable and safe sex and abuse prevention. Identifying the right kind of approach and/or resource to support the goals people have for their education is important. For example, some goals could be reached simply by explaining and practicing relevant skills while require a more formal approach.

Future Needs

Sexuality remains a fraught issue for people with intellectual disability. To summarise, people with intellectual disability have had, and continue to have, their sexuality and sexual health needs problematized rather than being viewed as a holistic need about love, pleasure, desire and identity. Both young men and women with intellectual disability view their sexuality within a fear-based paradigm: women are fearful of the risk of unwanted pregnancy and abuse, and men are fearful of being sexually inappropriate (Frawley & Wilson, 2016). Although much of the policy and practice settings have changed over the decades to more of a rights-based framework, the sexuality and sexual health of people with intellectual disability remains regulated and surveilled by others. This is a human rights issue. People with intellectual disability want friends and to develop meaningful relationships—whether intimate or not—and want access to the right to develop their own health sexuality at a pace of their choosing. The reality is that by virtue of having a cognitive impairment, they have been perceived as lacking capacity to develop skills and knowledge or to "have" sexual autonomy or agency. Most caregivers are family caregivers and in the absence of knowing what to do or where to access the appropriate supports and services, they can react conservatively in what they perceive as the "best interests" of their adult child with intellectual disability. Although not exhaustive and notwithstanding the absolute imperative that mainstream services have a major role to play, below is a summary of urgent needs in this area:

1. People with intellectual disability need more opportunities to form and maintain friendships and relationships. Other than descriptive studies, there is little research reporting on approaches to support relationship building and to counter loneliness.
2. Sex education programs that are based on bodies, pregnancy and disease are outdated. New evidence-based programs from a more salutogenic starting point are needed that promote sexual health and wellbeing, respectful relationships and that support ascertainment of sexual rights. These approaches need to focus on access to information and learning that meet people's needs as articulated by them or through appropriate assessments. In the new millennium, sex education also needs to take account of social media.
3. Sexual knowledge assessment tools should be based on the same salutogenic principles of the sex education programs. In addition, more

accessible formats that not only incorporate sexual diversity but also acknowledges and is responsive to different degrees of intellectual disability.

4. Family caregivers have asked to learn from professionals and to have better access to peer support, practical and affordable sexuality support and education services.
5. There are few notable advocates and researchers with a focus on and expertise in the intersection of male health and masculinity. Most importantly, the pathological discourse of male sexuality needs to be challenged with the development of male-specific approaches that give access to and reinforce a distinctly positive and healthy masculinity and male sexuality.
6. LGBTIQ people with intellectual disability are marginalised and a concerted effort to explore inclusive approaches, particularly from the mainstream LGBTIQ community, that can reduce this marginalisation should be prioritised.
7. Policy development must include greater collaboration with people with intellectual disability and their families. Mainstream service providers should be supported to develop inclusive approaches in partnership with disability supports, services and advocacy. This should always take into account the reality of different degrees of intellectual disability and need for support.
8. Continue to grow the research and evidence base in this complex area, including a focus on the lived experience, to identify ways that can better inform policy, education and practice.

An Example of a Promising Approach—Peer Support

Emerging Australian research shows that a peer education approach in partnership with mainstream services is both productive and positive (Frawley et al., 2012). The work undertaken as part of the *Living Safer Sexual Lives* and later *Sexual Lives and Respectful Relationships* (SL&RR) suite of programs have been shown to underpin best practice in the support of people with intellectual disability having access to a rights-based approach that prioritises the expertise and lived experience of people with intellectual disability (Frawley & O'Shea, 2012). The model that underpins this program has four key components; peer education by and for people with intellectual disability, learning support from trusted allies, community partnership with community based sexual health, sexual abuse and other related sexual and relationship services, and systemic change through the development of an evidence base (Frawley & Bigby, 2015). The program at the centre of this model uses the life stories of people with intellectual disability gathered in research (Johnson et al., 2001; O'Shea & Frawley, in press) as the basis for a respectful relationships program. The program was developed and is continually revised by project teams that comprise people with intellectual disability and academics. People with intellectual disability are the primary facilitators of the program as peer educators supported in program facilitation by professionals from community and sexual health. These partnerships form the basis of local networks that promote and run the program locally for adults with intellectual disability and serve as a resource to and conduit for, broader sexuality and relationship rights activities locally like the month of action against violence.

At the time of writing this chapter, there were over 60 trained peer educators and 60 trained program partners in seven local networks across Australia. This represents a large network of mainstream services working in partnership with people with intellectual disability in the provision of a sexuality and respectful relationships program which is both unique in terms of sexuality and sexual health approaches, and as a model of cross sector partnership unmatched in any other area of work with people with intellectual disability. These local networks run on average four programs a year with an average of eight participants per program. The SL&RR national network implements this program with over two hundred adults with intellectual disability each year across Australia. In 2018 the program has added stories and piloted a program for LGBTIQ adults with intellectual disability and is replicating the model with people with an Acquired Brain Injury.

Using peer education in this way with people with cognitive impairments in sexuality education and support is also unique and offers an important opportunity to build on the “nothing about us without us” dictum of the self-advocacy movement to prioritise and acknowledge the liberating power of working from the strength of shared lived experiences in the area of sexuality. Research on this approach has found that people with intellectual disability who have had the experience of being peer educators in this work feel more confident about their own relationships, their capacity to inform and manage their own sexuality and that through this role they can be role models for their peers (Frawley & Bigby, 2015). Further research is being undertaken on this model to better understand the structural and relational factors that underpin the strength of the local networks and partnerships, the outcomes for community organisations that are involved as partners and the learning and other outcomes for the participants with intellectual disability. Further work is also being undertaken with the peer educators to better understand what enables them to be the powerful sexuality educators they are and what this means for the future of sexuality education for and by people with intellectual disability.

Access to Mainstream Services and Supports

People with intellectual disability are often an overlooked cohort in mainstream sexuality research. One example in Australian research is the longitudinal study of sexual health, behaviour and experiences of secondary school age young people which in its fifth iteration in 2013 still did not recruit students with any type of disabilities (Mitchell, Patrick, Heywood, Blackman, & Pitts, 2014). These missed opportunities mean that the experiences of people with intellectual disability are not being identified, counted and understood alongside their non-disabled peers and are not present in the policy and practice outcomes from research. Mainstream education, health, counselling and support services relating to sexuality are not being shaped by knowledge about the disability experience. Further, although there is an increasingly strong focus on engaging mainstream services in understanding and responding to people with intellectual disability and the growth of inclusive and personalised approaches to funding and support worldwide, sexuality related needs of people with intellectual disability are either overlooked or considered to be the remit of “specialists”. While family planning organisations pay significant attention to and in some cases provide specialist services in sexual health for people with intellectual disability these are not broadly accessible. People with intellectual disability report being left out of sexuality education in mainstream schools (Frawley & Wilson, 2016), women with disabilities report that health professionals refuse to believe that they are sexually active and therefore ignore this area of their health (WWDA, 2018) and services like domestic, family and sexual assault services still struggle to provide fully accessible support to women with disabilities (Dyson, Frawley, & Robinson, 2017). Earlier sections of this chapter have suggested that what underpins this exclusion may well be sexual ableism whereby people with intellectual disabilities are not acknowledged as being imbued with the same sexual characteristics as others that afford them the right to be sexual (Gill, 2015).

Little work has been done that focuses on access to and inclusion in mainstream sexuality related services for people with intellectual disability. Williams, Scott & McKechanie (2014) reported that young people with intellectual disability used a range of sources for their information about sexual health including friends, family, their own research and TV, yet they would only go to their doctor if there was something “medical”. Research in Australia by Frawley and Wilson (2016) supported this view, finding that young people had devised many ways to become informed about sexual health often independent of health services, including talking to friends, using the internet and less frequently asking parents. These studies found that young people wanted and needed access to information about sexual health yet their past education experiences and current information finding approaches did not meet their needs and only a small number used mainstream sexual health services.

To address access to mainstream sexuality related services for people with intellectual disability there needs to be a multi-faceted approach that goes beyond questions of physical access. Research undertaken on access to mainstream domestic violence and sexual assault services for women with disabilities calls for an approach to access that includes physical access, information access, attitudinal access and procedural access Lea (2015). Others refer to this as programmatic access (Frohmader et al., 2015) where this means full access equal to what was available for all users of these services. Unpacking what is meant by access in this more holistic way has been found to be useful in addressing barriers to these mainstream services for women with intellectual disability and should be translatable to other services dealing with other sensitive and complex areas of life including sexuality and sexual health.

The multi-faceted approach recommended for full access to mainstream sexuality and other related services must start with a holistic understanding of disability taking an intersectional approach where this means understanding "the relationship between multiple dimensions and modalities of social relations and subject positions" (McCall, 2005 p. 1771). Dyson et al. (2017) outline that this approach must look at the complex lived experiences of disabilities where gender, race, class, culture, sexual and gender identity are considered where they intersect with other experiences including domestic and sexual violence. Here we suggest this is also applicable when addressing sexual health. Building on this approach services then engage in a range of ways of developing a broadly accessible service while also paying attention to specialist information and access needs. In the research on domestic and family violence services Dyson et al. (2017) found the following approaches were promising; making service adaptations underpinned by training of staff in understanding the complex lived experience of disability; cross sector collaboration where disability and specialist services work together from referral through to future planning; and developing and responding to a broad understanding of access where this includes thinking from the perspective of the person with intellectual disability and considering the approachability, appropriateness, affordability and availability of services. This more nuanced understanding of access informed by the work of Levesque, Harris, and Russell (2013) in health services makes a distinction between access where this means being able to "enter or obtain" a service and accessibility, where this means "it is useable to me".

Research cited earlier in this chapter makes it clear that the sexual health status of people with intellectual disability is not improving. While they are employing a range of strategies to become informed about their sexuality they are not accessing services that could provide them with the same access to beneficial information and support their non-disabled peers use. Learning from research about access to health and domestic violence services organisations like Family Planning Associations, women's health services and sexual health services could engage in a multifaceted approach to better understand disability and to better understand access to ensure people with intellectual disability do find these services "useable to them".

References

Abbott, D., & Burns, J. (2007). What's love got to do with it? Experiences of lesbian, gay, and bisexual people with intellectual disabilities in the United Kingdom and views of the staff who support them. *Sexuality Research & Social Policy: A Journal of the NSRC, 4*, 27–39. https://doi.org/10.1525/srsp.2007.4.1.27

Abraham, C. S., & Sheeran, P. (1994). Modeling and modifying young heterosexuals' HIV-preventive behaviour; a review of theories, findings and educational implications. *Patient Education and Counseling, 23*, 173–186.

Adnams, C. M. (2010). Perspectives of intellectual disability in South Africa: Epidemiology, policy, services for children and adults. *Current Opinion in Psychiatry, 23*(5), 436–440. https://doi.org/10.1097/YCO.0b013e32833cfc2d

Aldersey, H. M. (2012). Family perceptions of intellectual disability: Understanding and support in Dar es Salaam. *African Journal of Disability, 1*(1), 32. https://doi.org/10.4102/ajod.v1i1.32

Aunos, M., & Feldman, M. A. (2002). Attitudes towards sexuality, sterilization and parenting rights of persons with intellectual disabilities. *Journal of Applied Research in Intellectual Disabilities, 15*(4), 285–296. https://doi.org/10.1046/j.1468-3148.2002.00135.x

Azzopardi-Lane, C., & Callus, A. M. (2015). Constructing sexual identities: People with intellectual disability talking about sexuality. *British Journal of Learning Disabilities, 43*(1), 32–37.

Bane, G., Deely, M., Donohoe, B., & Wolfe, M. (2012). Relationships of people with learning disabilities in Ireland. *British Journal of Learning Disabilities, 40*(2), 109–122.

Bates, C., Terry, L., & Popple, K. (2016). The importance of romantic love to people with learning disabilities. *British Journal of Learning Disabilities, 45*, 64–72. https://doi.org/10.1111/bld.12177

Bell, D. M., & Cameron, L. (2003). The assessment of the sexual knowledge of a person with severe learning disability and a severe communication disorder. *British Journal of Learning Disabilities, 31*, 123–129.

Bernert, D. (2011). Sexuality and disability in the lives of women with intellectual disabilities. *Sexuality and Disability, 29*(2), 129–141.

Bos, A. E. R., Huijding, J., Muris, P., Vogel, L. R. R., & Biesheuvel, J. (2010). Global, contingent and implicit self-esteem and psychopathological symptoms in adolescents. *Personality and Individual Differences, 48*, 311–316. https://doi.org/10.1016/j.paid.2009.10.025

Brown, H., & Turk, V. (1994). Sexual abuse in adulthood: Ongoing risks for people with learning disabilities. *Child Abuse Review, 3*(1), 26–35.

Brown, M., & McCann, E. (2018). Sexuality issues and the voices of adults with intellectual disabilities: A systematic review of the literature. *Research in Developmental Disabilities, 74*, 124–138.

Brown, R. D., & Pirtle, T. (2008). Beliefs of professional and family caregivers about the sexuality of individuals with intellectual disabilities: Examining beliefs using a Q-methodology approach. *Sex Education, 8*(1), 59–75. https://doi.org/10.1080/14681810701811829

Bruder, C., & Kroese, B. S. (2005). The efficacy of interventions designed top recent and protect people with intellectual disabilities from sexual abuse: A review of the literature. *Journal of Adult protection, 7*, 13–27. https://doi.org/10.1108/14668203200500009

Butler, J., Leighton, D., & Galea, J. (2003). *The assessment of sexual knowledge*. Melbourne: Centre for Developmental Disability Health Victoria.

Cambridge, P. (1996). Men with learning disabilities who have sex with men in public places: Mapping the needs of services and users in south east London. *Journal of Intellectual Disability Research, 40*(3), 241–251.

Charncok, D., & Standen, P. J. (2013). Second hand masculinity: Do boys with intellectual disabilities use computer games as part of gender practice? *International Journal of Game-Based Learning, 3*(3), 43–53. https://doi.org/10.4018/ijgbl.2013070104

Chataika, T., & McKenzie, J. (2013). Considerations of an African childhood disability studies. In K. Runswick-Cole & T. Curran (Eds.), *Disabled children's childhood studies: Critical approaches in a global context* (pp. 152–163). London: Palgrave MacMillon.

Clawson, R., & Fyson, R. (2017). Forced marriage of people with learning disabilities: A human rights issue. *Disability and Society, 32*, 810–830. https://doi.org/10.1080/09687599.2017.1320271

Cobigo, V., Ouellette-Kuntz, H., Balogh, R., Leung, F., Lin, E., & Lunsky, Y. (2013). Are cervical and breast cancer screening programmes equitable? The case of women with intellectual and developmental disabilities. *Journal of Intellectual Disability Research, 57*(5), 478–488. https://doi.org/10.1111/jir.12035

De Jong Gierveld, J. (1998). A review of loneliness: Concept and definitions, determinants and consequences. *Reviews in Clinical Gerontology, 8*, 73–80. https://doi.org/10.1017/S0959259898008090

Desjardins, M. (2012). The sexualized body of the child, parents and the politics of 'voluntary' sterilization of people labelled intellectually disabled. In R. McRuer & A. Mollow (Eds.), *Sex and disability* (pp. 69–85). Durham, NC: Duke University Press.

Dupras, A., & Dionne, H. (2014). The concern of parents regarding the sexuality of their child with a mild intellectual disability. *Sexologies, 23*(4), e79–e83. https://doi.org/10.1016/j.sexol.2013.09.002

Dyson, S., Frawley, P., & Robinson, S. (2017). *"Whatever it takes": Access for women with disabilities to domestic and family violence services*. Sydney: Australia's National Research Organisation for Women's Safety. Retrieved from https://www.anrows.org.au/node/1413

Emerson, E. (2007). Poverty and people with intellectual disabilities. *Developmental Disabilities Research Reviews, 13*(2), 107–113. https://doi.org/10.1002/mrdd.20144

Evans, D. S., McGuire, B. E., Healy, E., & Carley, S. N. (2009). Sexuality and personal relationships for people with an intellectual disability. Part II: Staff and family carer perspectives. *Journal of Intellectual Disability Research, 53*(11), 913–921. https://doi.org/10.1111/j.1365-2788.2009.01202.x

Family Planning Victoria. (1997). *The human relations and sexuality knowledge and awareness assessment*. Melbourne: FPV.

Fitzgerald, C., & Withers, P. (2013). 'I don't know what a proper woman means': What women with intellectual disabilities think about sex, sexuality and themselves. *British Journal of Learning Disabilities, 41*(1), 5–12. https://doi.org/10.1111/j.1468-3156.2011.00715.x

Fitzsimons, N. M. (2009). *Combating violence and abuse of people with disabilities*. Baltimore, MD: Paul H. Brookes.

Foley, S. (2013). Reluctant ‘jailors’ speak out: Parents of adults with Down syndrome living in the parental home on how they negotiate the tension between empowering and protecting their intellectually disabled sons and daughters. *British Journal of Learning Disabilities, 41*(4), 304–311. https://doi.org/10.1111/j.1468-3156.2012.00758.x

Foley, S. (2017). *Intellectual disability and the right to a sexual life: A continuation of the autonomy/paternalism debate*. Routledge.

Frawley, P., Barrett, C. & Dyson, S. (2012). Real people, core business. Living safer sexual lives: Respectful relationships. *Report on the development and implementation of a peer-led violence and abuse prevention program for people with an intellectual disability*. Melbourne, Australia: Australian Research Centre in Sex, Health and Society, La Trobe University.

Frawley, P., & Bigby, C. (2015). “Molly is just like me”: Peer education and life stories in sexuality programmes in Australia. In R. Chapman, S. Ledger, L. Townson, & D. Docherty (Eds.), *Sexuality and relationships in the lives of people with intellectual disabilities: Standing in my shoes* (pp. 119–132). London: Jessica Kingsley.

Frawley, P., & O’Shea, A. (2012). Relationship education by people with an intellectual disability: An Australian peer educator model. Conference presentation. 14th World Congress International Association for the Scientific Study of Intellectual disabilities, Halifax, Nova Scotia, Canada, Journal of Intellectual Disability Research., *56*(7–8), 726.

Frawley, P., O’Shea, A., Ardley, E., Cini, V., Davie, R., Stokoe, L., & Wellington, M. (2017). *Sexual lives & respectful relationships training manual*. Geelong, Australia: Deakin University.

Frawley, P., & Wilson, N. J. (2016). Young people with intellectual disability talking about sexuality education and information. *Sexuality and Disability, 34*(4), 469–484.

Friedman, C., Arnold, C. K., Owen, A. L., & Sandman, L. (2014). “Remember our voices are our tools”: Sexual self-advocacy as defined by people with intellectual and developmental disabilities. *Sexuality and Disability, 32*(4), 515–532.

Frohmader, C., Dowse, L. & Didi, A. (2015). Preventing violence against women and girls with disabilities: Integrating a human rights perspective. Sydney, Australia: Women with Disabilities Australia. ISBN: 978-0-9585268-4-5

Fulford, C., & Cobigo, V. (2016). Friendships and intimate relationships among people with intellectual disabilities: A thematic synthesis. *Journal of Applied Research in Intellectual Disabilities*. https://doi.org/10.1111/jar.12312. [advance online publication].

Galea, J., Butler, J., Iacono, T., & Leighton, D. (2004). The assessment of sexual knowledge in people with intellectual disability. *Journal of Intellectual & Developmental Disability, 29*(4), 350–365.

Gardiner, T., & Braddon, E. (2009). ‘A Right to Know’: Facilitating a relationship and sexuality programme for adults with intellectual disabilities in Donegal. *British Journal of Learning Disabilities, 37*(4), 327–339. https://doi.org/10.1111/j.1468-3156.2009.00591.x

Gill, M. (2015). *Already doing it: Intellectual disability and sexual agency*. University of Minnesota Press.

Gilmore, L. & Cuskelly, M. (2014). Vulnerability to Loneliness in People with Intellectual Disability: An Explanatory Model. Journal of Policy and Practice in Intellectual Disabilities, 11(3), 192-199. https://doi.org/10.1111/jppi.12089

Gomez, M. T., Carlson, G. M., & Van Dooren, K. (2012). Practical approaches to supporting young women with intellectual disabilities and high support needs with their menstruation. *Health Care for Women International, 33*(8), 678–694. https://doi.org/10.1080/07399332.2012.684812

Griffiths, D., & Lunsky, Y. (2003). *The socio-sexual knowledge and attitude assessment tool instruction manual*. Wood Dale, IL: Stoelting.

Gürol, A., Polat, S., & Oran, T. (2014). Views of mothers having children with intellectual disability regarding sexual education: A qualitative study. *Sexuality and Disability, 32*(2), 123–133. https://doi.org/10.1007/s11195-014-9338-8

Healy, E., McGuire, B. E., Evans, D. S., & Carley, S. N. (2009). Sexuality and personal relationships for people with an intellectual disability. Part I: Service-user perspectives. *Journal of Intellectual Disability Research, 53*, 905–912.

Hingsburger, D. (1994). Masturbation: A consultation for those who support individuals with developmental disabilities. *Canadian Journal of Human Sexuality, 3*(3), 278–282.

Ingstad, B. (1995). Mpho Ya Modimo--a gift from God. In B. Ingstad & S. R. Whyte (Eds.), *Disability and culture* (pp. 246–263). Berkley & Los Angeles, CA: University of California Press.

Isler, A., Beytut, D., Tas, F., & Conk, Z. (2009). A study on sexuality with the parents of adolescents with intellectual disability. *Sexuality and Disability, 27*(4), 229–237. https://doi.org/10.1007/s11195-009-9130-3

Jahoda, A., & Pownall, J. (2014). Sexual understanding, sources of information and social networks: The reports of young people with intellectual disabilities and their nondisabled peers. *Journal of Intellectual Disability Research, 58*(5), 430–441. https://doi.org/10.1111/jir.12040

Johnson, K., Hillier, L., Harrison, L., & Frawley, P. (2001). *People with intellectual disabilities living safer sexual lives*. Victoria, Australia: La Trobe University, Australian Research Centre in Sex, Health, and Society.

Kahonde, C. (2016). *A grounded theory study of family caregivers’ responses to the sexuality of young adults with intellectual disabilities*. South Africa: PhD thesis, University of Cape Town.

Kahonde, C. K., McKenzie, J., & Wilson, N. J. (2018). Discourse of needs versus discourse of rights: Family caregivers responding to the sexuality of young South African adults with intellectual disability. *Culture,* *Health and Sexuality*. https://doi.org/10.1080/13691058.2018.1465202

Kijak, R. (2013). The sexuality of adults with intellectual disability in Poland. *Sexuality and Disability, 31*(2), 109–123.

Lafferty, A., McConkey, R., & Simpson, A. (2012). Reducing the barriers to relationships and sexuality education for persons with intellectual disabilities. *Journal of Intellectual Disability, 16*(1), 29–43. https://doi.org/10.1177/1744629512438034

Lea, M. (Ed.). (2015).Women With Disability and Domestic and Family Violence: A Guide For Policy and Practice. Sydney, NSW: People with Disabilities Australia and Domestic Violence NSW.

Levesque, J., Harris, M. F., & Russell, G. (2013). Patient-centred access to health care: Conceptualising access at the interface of health systems and populations. *International Journal for Equity in Health, 12*(1), 171–182. https://doi.org/10.1186/1475-9276-12-18

Lunsky, Y., Frijters, J., Griffiths, D. M., Watson, S. L., & Williston, S. (2007). Sexual knowledge and attitudes of men with intellectual disability who sexually offend. *Journal of Intellectual & Developmental Disability, 32*, 74–81.

Mabovula, N. N. (2011). The erosion of African communal values: A reappraisal of the African Ubuntu philosophy. *Inkanyiso: Journal of Humanities and Social Sciences, 3*(1), 38–47. https://doi.org/10.4314/ijhss.v3i1.69506

McCabe, M. P. (1994). *Sexual knowledge, experience and needs scale for people with intellectual disability (SexKen-ID)*. Melbourne: Psychology Research Centre, Deakin University.

McCall, L. (2005). The complexity of intersectionality. *Signs: Journal of Women in Culture and Society, 30*(3), 1771–1800.

McCann, E., Lee, R., & Brown, M. (2016). The experiences and support needs of people with intellectual disabilities who identify as LGBT: A review of the literature. *Research in Developmental Disabilities, 57*, 39–53.

McCarthy, M. (2002). Going through the menopause: Perceptions and experiences of women with intellectual disability. *Journal of Intellectual and Developmental Disability, 27*(4), 281–295.

McCarthy, M. (2009) 'I have the jab so I can't be blamed for getting pregnant': Contraception and women with learning disabilities. In *Women's Studies International Forum* (Vol. 32(3), pp. 198–208). Oxford, UK: Pergamon Press.

McCarthy, M. (2010). Exercising choice and control–women with learning disabilities and contraception. *British Journal of Learning Disabilities, 38*(4), 293–302.

McCarthy, M. (2014). Women with intellectual disability: Their sexual lives in the 21st century. *Journal of Intellectual and Developmental Disability, 39*(2), 124–131. https://doi.org/10.3109/13668250.2014.894963

McClelland, A., Flicker, S., Nepveux, D., Nixon, S., Vo, T., Wilson, C., … Proudfoot, D. (2012). Seeking safer sexual spaces: Queer and trans young people labelled with intellectual disabilities and the paradoxical risks of restriction. *Journal of Homosexuality, 59*(6), 808–819.

McElduff, A., & Beange, H. (2003). Men's health and well-being: Testosterone deficiency. *Journal of Intellectual and Developmental Disabilities, 28*(2), 211–213.

McKenzie, J., McConkey, R., & Adnams, C. (2013). Intellectual disability in Africa: Implications for research and service development. *Disability and Rehabilitation, 35*(20), 1750–1755. https://doi.org/10.3109/09638288.2012.751461

McVilly, K.R., Stancliffe, R.J., Parmenter, T.R. & Burton-Smith, R.M. (2006). "I get by with a little help from by friends": Adults with intellectual disability discuss loneliness. *Journal of Applied Research in Intellectual Disabilities, 19*(2), 191–203. https://doi.org/10.1111/j.1468-3148.2005.00261.x

Merrick, J., Morad, M., & Carmeli, E. (2014). Intellectual and developmental disabilities: Male health. *Frontiers in Public Health, 2*, 208. https://doi.org/10.3389/fpubh.2014.00208

Mitchell, A., Patrick, K., Heywood, W., Blackman, P. & Pitts, M. (2014). 5th National Survey of Australian Secondary Students and Sexual Health 2013, (ARCSHS Monograph Series No. 97). Australian Research Centre in Sex, Health and Society, La Trobe University, Melbourne, Australia.

Morales, G. E., Lopez, E. O., & Mullet, E. (2011). Acceptability of sexual relationships among people with learning disabilities: Family and professional caregivers' views in Mexico. *Sexuality and Disability, 29*(2), 165–174. https://doi.org/10.1007/s11195-011-9201-0

Murphy, G., Powell, S., Guzman, A., & Hays, S. (2007). Cognitive-behavioural treatment for men with intellectual disabilities and sexually abusive behaviour: A pilot study. *Journal of Intellectual Disability Research, 51*, 902–912.

Murphy, G. H., & O'Callaghan, A. (2004). Capacity of adults with intellectual disabilities to consent to sexual relationships. *Psychological Medicine, 34*, 1347–1357. https://doi.org/10.1017/S0033291704001941

O'Shea, A. (2016). *"Just a rare girl": Gender and disability in life stories produced with young women with disabilities*. PhD Thesis, La Trobe University.

O'Neill, J., Lima, S., Thomson Bowe, K., & Newall, F. (2015). The experiences and needs of mothers supporting young adolescents with intellectual disabilities through puberty and emerging sexuality. *Research and Practice in Intellectual and Developmental Disabilities, 3*(1), 37–47. https://doi.org/10.1080/23297018.2015.1077342

Perlin, M. L., & Lynch, A. J. (2015). How teaching about therapeutic jurisprudence can be a tool of social justice, and lead law students to personally and socially rewarding careers: Sexuality and disability as a case example. *Nevada Law Journal, 16*, 209.

Pownall, J. D., Jahoda, A., & Hastings, R. (2012). Sexuality and sex education of adolescents with intellectual disability: Mothers' attitudes, experiences, and support needs. *Intellectual and Developmental Disabilities, 50*(2), 140–154. https://doi.org/10.1352/1934-9556-50.2.140

Pownall, J. D., Jahoda, A., Hastings, R., & Kerr, L. (2011). Sexual understanding and development of young people with intellectual disabilities: Mothers' perspectives of within-family context. *American Journal on Intellectual and Developmental Disabilities, 116*(3), 205–219. https://doi.org/10.1352/1944-7558-116.3.205

Rogers, C. (2010). But it's not all about the sex: Mothering, normalisation and young learning disabled people. *Disability & Society, 25*(1), 63–74. https://doi.org/10.1080/09687590903363336

Rojas, S., Haya, I., & Lazaro-Visa, S. (2016). 'My great hope in life is to have a house, a family and a daughter': Relationships and sexuality in intellectually disabled people. *British Journal of Learning Disabilities, 44*(1), 56–62. https://doi.org/10.1111/bld.12110

Ruiz, F. J. (2017). The Committee on the rights of persons with disabilities and its take on sexuality. *Reproductive Health Matters, 25*(50), 92–103. https://doi.org/10.1080/09688080.2017.1332449

Rushbrooke, E., Murray, C. D., & Townsend, S. (2014). What difficulties are experienced by caregivers in relation to the sexuality of people with intellectual disabilities? A qualitative meta-synthesis. *Research in Developmental Disabilities, 35*(4), 871–886. https://doi.org/10.1016/j.ridd.2014.01.012

Sajith, S. G., Morgan, C., & Clarke, D. (2008). Pharmacological management of inappropriate sexual behaviours: A review of its evidence, rationale, and scope in relation to men with intellectual disabilities. *Journal of Intellectual Disability Research, 52*(12), 1078–1090.

Sasco, A. J., Ah-Song, R., Nishi, M., Culine, S., Rethore, M. O., & Satge, D. (2008). Testicular cancer and intellectual disability. *International Journal of Disability and Human Development, 7*(4), 397–403.

Schaaf, M. (2012). Negotiating sexuality in the convention on the rights of persons with disabilities. *Sur International Journal on Human Rights, 8*(14), 113.

Schaafsma, D. (n.d.). *Dating agencies for people with an intellectual disability* (unpublished manuscript)

Schaafsma, D., Kok, G., Stoffelen, J. M. T., & Curfs, L. M. G. (2016). Seksuele voorlichtingsmaterialen voor mensen met een verstandelijke beperking: Beschikbaarheid en kwaliteit. *Nederlands Tijdschrift voor de Zorg aan mensen met verstandelijke beperking, 1*, 31–45.

Schaafsma, D., Kok, G., Stoffelen, J. M. T., & Curfs, L. M. G. (2017). People with intellectual disabilities talk about sexuality: Implications for the development of sex education. *Sexuality and Disability, 35*, 21–38. https://doi.org/10.1007/s11195-016-9466-4

Schaafsma, D., Kok, G., Stoffelen, J. M. T., van Doorn, P., & Curfs, L. M. G. (2014). Identifying the important determinants of teaching sex education to people with intellectual disabilities: A cross-sectional survey among paid care staff. *Journal of Intellectual and Developmental Disability, 39*, 157–166. https://doi.org/10.3109/13668250.2014.899566

Schaafsma, D., Stoffelen, J. M. T., Kok, G., & Curfs, L. M. G. (2013). Exploring the development of existing sex education programs for people with intellectual disabilities: An intervention mapping approach. *Journal of Applied Research in Intellectual Disabilities, 26*, 157–166. https://doi.org/10.1111/jar.12017

Sobsey, R. (1994). *Violence and abuse in the lives of people with disabilities: The end of silent acceptance?* Baltimore, MD: Paul H Brookes.

Stancliffe, R. J., Wilson, N., Bigby, C., Balandin, S., & Craig, D. (2014). Responsiveness to self-report questions about loneliness: A comparison of mainstream and intellectual disability-specific instruments. *Journal of Intellectual Disability Research, 58*(5), 399–405.

Stoffelen, J., Kok, G., Hospers, H., & Curfs, L. (2013). Homosexuality among people with a mild intellectual disability: An explorative study on the lived experiences of homosexual people in the Netherlands with a mild intellectual disability. *Journal of Intellectual Disability Research, 57*(3), 257–267.

Talbot, T. J., & Langdon. (2006). A revised sexual knowledge assessment tool for people with intellectual disabilities: Is sexual knowledge related to sexual offending behaviour? *Journal of Intellectual Disability Research, 50*(7), 523–531. https://doi.org/10.1111/j.1365-2788.2006.00801.x

Taylor Gomez, M. (2012). The S Words: Sexuality, sensuality, sexual expression and people with intellectual disability. *Sexuality and Disability, 30*(2), 237–245.

Thompson, D., Clare, I., & Brown, H. (1997). Not such an ordinary relationship: The role of women support staff in relation to men with learning disabilities who have difficult sexual behaviour. *Disability and Society, 12*(4), 573–592.

Thompson, V. R., Stancliffe, R. J., Wilson, N. J., & Broom, A. (2016a). Clinicians' use of sexual knowledge assessment tools for people with intellectual disability. *Journal of Intellectual & Developmental Disability, 41*(3), 243–254. https://doi.org/10.3109/13668250.2016.1164303

Thompson, V. R., Stancliffe, R. J., Wilson, N. J., & Broom, A. (2016b). The content, usefulness and usability of sexual knowledge assessment tools for people with intellectual disability. *Sexuality and Disability, 34*(4), 495–512.

Tilley, E., Walmsley, J., Earle, S., & Atkinson, D. (2012). 'The silence is roaring': Sterilization, reproductive rights and women with intellectual disabilities. *Disability & Society, 27*(3), 413–426. https://doi.org/10.1080/09687599.2012.654991

Traustadóttir, R. (2006). Disability and gender: Introduction to the special issue. *Scandinavian*

Journal of Disability Research, 8(2), 81–84. https://doi.org/10.1080/15017410600831341

Truesdale-Kennedy, M., Taggart, L., & McIlfatrick, S. (2011). Breast cancer knowledge among women with intellectual disabilities and their experiences of receiving breast mammography. *Journal of Advanced Nursing, 67*(6), 1294–1304. https://doi.org/10.1111/j.1365-2648.2010.05595.x

UN General Assembly (2007) Convention on the Rights of Persons with Disabilities: Resolution/adopted by the General Assembly, A/RES/61/106. Retrieved 4 April 2018, from http://www.refworld.org/docid/45f973632.html

Van Asselt-Goverts, A. E., Embregts, P. J. C. M., & Hendriks, A. H. C. (2013). Structural and functional characteristics of the social networks of people with mild intellectual disabilities. *Research in Developmental Disabilities, 34*, 1280–1288. https://doi.org/10.1016/j.ridd.2013.01.012

van Schrojenstein Lantman-de Valk, H. M. J. (2005). Health in people with intellectual disabilities: Current knowledge and gaps in knowledge. *Journal of Applied Research in Intellectual Disabilities, 18*, 325–333.

Wheeler, P. (2007). "I count myself as normal, well, not normal, but normal enough": Men with learning disabilities tell their stories about sexuality and sexual identity. *Tizard Learning Disability Review, 12*(1), 16–27.

Wheeler, P., & Jenkins, R. (2004). The management of challenging sexual behaviour. *Learning Disability Practice, 7*(5), 28–37.

WHO/UNFPA. (2009). *Promoting sexual and reproductive health for persons with disabilities. WHO/UNFPA guidance note*. Geneva: WHO.

Williams, F., Scott, G. & McKechanie, A. (2014). Sexual health services and support: The views of younger adults with intellectual disability. *Journal of Intellectual and Developmental Disability, 39*, 147–156. https://doi.org/10.3109/13668250.2014.899326

Wilson, N. J., & Frawley, P. (2016). Transition staff discuss sex education and support for young men and women with intellectual and developmental disability. *Journal of Intellectual and Developmental Disability, 41*(3), 209–221. https://doi.org/10.3109/13668250.2016.1162771

Wilson, N. J., Macdonald, J., Hayman, B., Bright, A. M., Frawley, P., & Gallego, G. (2016). A narrative review of the literature about people with intellectual disability who identify as lesbian, gay, bisexual, transgender, intersex or questioning. *Journal of Intellectual Disabilities, 22*(2), 171–196. https://doi.org/10.1177/1744629516682681

Wilson, N. J., Parmenter, T. R., Stancliffe, R. J., & Shuttleworth, R. P. (2011). Conditionally sexual: Men and teenage boys with moderate to profound intellectual disability. *Sexuality and Disability, 29*(3), 275–289.

Wilson, N. J., Parmenter, T. R., Stancliffe, R. J., & Shuttleworth, R. P. (2013). From diminished men to conditionally masculine: Sexuality and Australian men and boys with intellectual disability. *Culture Health and Sexuality, 15*(6), 738–751. https://doi.org/10.1080/13691058.2013.780262

Wilson, N. J., Parmenter, T. R., Stancliffe, R. J., Shuttleworth, R. P., & Parker, D. (2010). A masculine perspective of gendered topics in the research literature on males and females with intellectual disability. *Journal of Intellectual and Developmental Disability, 35*(1), 1–8. https://doi.org/10.3109/13668250903496351

Women with Disabilities Australia (WWDA). (2018). *Brief Submission to the Special Rapporteur on the Rights of Persons with Disabilities on the Right of Persons with Disabilities to the Highest Attainable Standard of Health*. 30 March 2018. WWDA: Hobart, Tasmania.

World Health Organization. (2010). *Standards for Sexuality Education in Europe: A framework for policy makers, educational and health authorities and specialists*. Keulen: WHO Regional Office for Europe and BZgA.

World Health Organization. (2011). *World report on disability.* Retrieved 1 September 2014, from http://whqlibdoc.who.int/publications/2011/9789240685215_eng.pdf.

Yacoub, E., & Hall, I. (2009). The sexual lives of men with mild learning disability: A qualitative study. *British Journal of Learning Disabilities, 37*(1), 5–11.

Pain

53

Lara M. Genik, Kaytlin Constantin,
Frank J. Symons, and C. Meghan McMurtry

This chapter begins with a short review of pain relevant terms and concepts, as well as a summary of life span/developmental perspectives on pain including prevalence, assessment, and management. This information will provide important context for our subsequent review of what is known about pain assessment and management in people with DD. The complexities of pain assessment and management in populations with DD will be further illustrated through a special section on advancing the field through research and two cases studies. Throughout, we have made reference to "pain in developmental disability ('pain in DD')". There is enormous heterogeneity associated with the DD label which does encompass people with and without intellectual disability (ID); however, for the purposes of readability, we have used the term "DD" interchangeably with ID throughout the chapter.

Introduction to Pain

Pain is a common experience that occurs throughout the life span, and can be defined as an "unpleasant sensory and emotional experience associated with actual or potential tissue damage, or described in terms of such damage" (Merskey & Bogduk, 1994, p. 210). The International Association for the Study of Pain (IASP) further indicates that pain is subjective, and that difficulty communicating verbally does not prevent an individual from experiencing or requiring relief from pain (International Association for the Study of Pain (IASP), 2012, Pain terms, para. 2; Merskey & Bogduk, 1994). The complexity of pain is well captured by the biopsychosocial model, which considers an individual's internal pain experience and external pain expression to be multiply determined by biological (e.g., genetics, hormones), psychological (e.g., emotions), and social factors (e.g., caregiver behaviors; Craig, 2009; Craig, Versloot, Goubert, Vervoort, & Crombez, 2010; Hadjistavropoulos et al., 2011); in turn, pain also influences these factors. For example, if an athlete were injured during a tournament, a number of factors such as adrenaline levels, how the player feels, and the number

L. M. Genik (✉) · K. Constantin
Department of Psychology, University of Guelph, Guelph, ON, Canada
e-mail: lgenik@uoguelph.ca

F. J. Symons
Department of Educational Psychology, University of Minnesota, Minneapolis, MN, USA

C. M. McMurtry
Department of Psychology, University of Guelph, Guelph, ON, Canada

Pediatric Chronic Pain Program, McMaster Children's Hospital, Hamilton, ON, Canada

Department of Paediatrics, Western University, London, ON, Canada

J. L. Matson (ed.), *Handbook of Intellectual Disabilities*, Autism and Child Psychopathology Series, https://doi.org/10.1007/978-3-030-20843-1_53

of people watching the game could impact his or her subsequent pain experience and expression.

An important distinction can be made between two types of pain: acute and chronic. Acute pain is limited to a short period of time and is generally viewed as adaptive, serving to alert an individual of potential harm and motivate him or her to take appropriate action (American Academy of Pediatrics, 2001). Common sources of acute pain include injury, illness, brief tissue-damaging medical procedures (e.g., finger pricks, lumbar punctures, venipuncture) and non-tissue-damaging medical procedures (e.g., suctioning; American Academy of Pediatrics, 2001; Stevens & Zempsky, 2013). In a typical week, approximately 30–40% of children experience some type of acute pain (McGrath, 1990). Epidemiological data from eight Canadian hospitals revealed that just over 78% of children admitted had undergone at least one painful procedure, and common procedures varied in pain severity from mild (e.g., suctioning) to moderate/severe (e.g., venipuncture; Stevens et al., 2011). In adults, it is estimated that approximately 56% experience some level of pain within a three-month period (Nahin, 2015). Poorly managed acute pain can lead to a number of negative impacts, including an increased risk of developing chronic pain conditions (Sinatra, 2010).

Chronic pain is typically defined as pain lasting longer than three months' duration (Merskey & Bogduk, 1994). It can involve recurrent pain, which includes episodic acute pain (e.g., headaches, stomachaches) or persistent pain, which is more enduring (e.g., backaches; Stevens & Zempsky, 2013). The occurrence of chronic pain in both children and adults increases with age, and once reaching adolescence, females report more frequent and intense experiences (Johannes, Le, Zhou, Johnston, & Dworkin, 2010; King et al., 2011). Pain in children and adolescents has been deemed a public health concern (Haraldstad, Sørum, Eide, Natvig, & Helseth, 2011; Perquin et al., 2000); the prevalence of headaches ranges from 8% to 83%, abdominal pain from 4% to 40%, and back pain from 14% to 24% (King et al., 2011). Approximately one in five adults experience chronic pain (Schopflocher, Taenzer, & Jovey, 2011), with prevalence estimates ranging from 8.7% to 64.4% depending on the population, type of pain and survey method (Johannes et al., 2010; Leadley, Armstrong, Lee, Allen, & Kleijnen, 2012; Nahin, 2015; Steingrímsdóttir, Landmark, Macfarlane, & Nielsen, 2017).

Taken together, existing research on pain across the life span demonstrates the high prevalence of acute and chronic pain in children, youth, and adults. The experience of pain, particularly chronic pain, can interfere significantly with daily functioning and is associated with health and economic consequences. Children and adolescents may miss school, withdraw from social activities, and are at risk of developing internalizing symptoms (Palermo, 2000). Further, children commonly report that their pain has significant impact on their sleep quality and ability to engage in activities and hobbies (Haraldstad et al., 2011; Valrie, Bromberg, Palermo, & Schanberg, 2013). Similarly, adults with chronic pain tend to experience lower health-related quality of life, interference with daily activities, job loss or reduced job responsibilities, higher rates of internalizing symptoms, and strained relationships (Doth, Hansson, Jensen, & Taylor, 2010; Sessle, 2012). There are also financial burdens associated with chronic pain, including the cost for patients, caregivers, the health care system and the economy more generally. Individuals with chronic pain tend to utilize almost twice as many health care resources as the general population, and the management of pain requires access to a wide range of services, including physical therapy, inpatient, pharmaceutical, and primary care (Sessle, 2012). In Canada, the personal financial costs for patients with pain is approximately $1500 per month; the combination of direct and indirect costs to the Canadian economy has been estimated to be greater than $40 billion per year (Lynch, 2011; Sessle, 2012). In the USA, this cost is estimated at more than $500 billion a year (Sessle, 2012).

Pain in People with Developmental Disabilities (DD)

Pain in people with DD is frequently underrecognized and underreported and has received minimal attention in the research literature

(McGuire, Daly, & Smyth, 2010). A number of false beliefs and assumptions may contribute to this dearth of research, such as historical views associated with dehumanization and depersonalization of people with DD (Sobsey, 2006). For example, in the past, people tended to view those with DD as either (a) "less than human," and therefore less deserving of appropriate treatment, or (b) suffering in extreme ways which were ultimately unmanageable (Sobsey, 2006). In either case, these polarized beliefs often led to inadequate care, neglect or abuse of those with DD (Sobsey, 2006). There are many examples of the impact of these beliefs, such as the use of pain to teach people with DD in the 1960s and 1970s (Sobsey, 2006). At that time, it was not uncommon for caregivers and therapists to use painful stimuli such as hitting and shocks for punishment or conditioning stimuli (Sobsey, 2006).

One of the most significant and long-standing beliefs related to pain and people with DD regards their ability to experience or feel pain (Sobsey, 2006; Symons, Shinde, & Gilles, 2008). Specifically, it was believed that people with DD were (a) insensitive or incapable of experiencing pain, or (b) in a constant state of severe and unmanageable pain, inevitably leading to a culture in which pain was disregarded and untreated (Sobsey, 2006). Appropriate pain assessment and management for people with DD continues to be problematic, and there is still a tendency for pain to be missed, discounted, or ignored. For example, following spinal fusion surgery, children with DD were less often assessed for pain and given fewer pain-relieving opioids compared to those without DD (Malviya et al., 2001). Similarly, parents report that their children with DD are treated differently in medical settings, with professionals often seeming to overlook the pain or "pass it off" as someone else's responsibility (Carter, McArthur, & Cunliffe, 2002). Still further, work by McGuire et al. (2010) suggests continued under-recognition of pain in people with DD. This may be particularly true for those with more severe disabilities and limitations (McGuire et al., 2010). Despite accumulation of research to the contrary, false beliefs and related misunderstandings such as these can deeply impact and limit the quality of care provided to people in need.

Prevalence

In reality, true insensitivity to pain is very rare and not commonly associated with DD (Nagasako, Oaklander, & Dworkin, 2003; Sobsey, 2006). Much of the DD literature instead points to increased sensitivity to pain compared to people without DD (e.g., Defrin, Pick, Peretz, & Carmeli, 2004; Nader, Oberlander, Chambers, & Craig, 2004; Symons, Harper, McGrath, Breau, & Bodfish, 2009). It is therefore understood that people with DD do feel pain (Oberlander, O'Donnell, & Montgomery, 1999); however, limitations and challenges associated with their diagnoses may impact or alter their experience and/or expression of pain (e.g., Dubois, Capdevila, Bringuier, & Pry, 2010; Hennequin, Morin, & Feine, 2000; Nader et al., 2004; Valkenburg, Tibboel, & Dijk, 2015). For example, experimental work with both children and adults diagnosed with Down syndrome has demonstrated differences when compared to typically developing samples such as slower reaction times to painful stimuli and decreased ability to accurately demonstrate pain location (Hennequin et al., 2000; Valkenburg et al., 2015).

While the true prevalence of pain in people with DD is unclear, research suggests that pain is not only common for people with DD but may actually occur more frequently than in populations without DD, particularly in children (Breau, Camfield, McGrath, & Finley, 2003; Stallard, Williams, Lenton, & Velleman, 2001). For example, in a study of children with disabilities, 75% of the sample was reported to have experienced pain over a two-week period, 84% of whom had experienced pain on at least 5 days (Stallard et al., 2001). Similarly, in another sample more than one third of children with severe DD experienced pain lasting more than 1 h on a weekly basis (Breau, Camfield, et al., 2003). In both of these studies, the frequency of pain experience increased alongside the severity of the child's disability (Breau, Camfield, et al., 2003; Stallard et al., 2001). Despite small sample sizes, these rates are much higher than those reported in prevalence studies associated with pain in typically developing children (e.g., Perquin et al., 2000). Prevalence studies focusing on adults with DD have also suggested that pain occurs frequently (e.g., Boerlage et al., 2013; McGuire et al., 2010; Walsh, Morrison, & McGuire, 2011). For example, in one study caregivers reported that at least 15.4% of the sample of adults with DD experienced chronic pain (Walsh et al., 2011). Notably, the rates of pain prevalence for adults with DD are somewhat lower than in other studies of adults without DD (e.g., Boerlage et al., 2013). Given the history of misbeliefs and misunderstandings associated with pain in people with DD, these may be under-representations of true prevalence rates, particularly for adults with limited ability to communicate effectively (Boerlage et al., 2013; McGuire et al., 2010).

Pain sources are diverse and include everyday pains (e.g., headaches), unintentional injury (e.g., bumps, fall), and medical procedures (e.g., venipuncture). Most frequently for people with DD, however, pain seems to originate from comorbid medical conditions and related medical procedures (Breau, Camfield, et al., 2003); thus, certain types of pain may be more prevalent for people with specific disabilities. For example, people with Down syndrome may be more susceptible to pain associated with congenital health defects and oral health-related diseases (Bottos & Chambers, 2006), while musculoskeletal and gastrointestinal pain may be more common among people with cerebral palsy, severe DD, and autism spectrum disorder (Walsh et al., 2011).

Impact on Functioning

In many ways, the broad impact of pain on people with DD is similar to that of people without DD described above. For example, in youth with autism spectrum disorder and DD pain is associated with parasomnias, sleep duration, and sleep disordered breathing (Breau & Camfield, 2011; Tudor, Walsh, Mulder, & Lerner, 2015). Breau, Camfield, McGrath, and Finley's (2007) work highlights the deleterious impact that pain has on adaptive functioning skills of youth with DD across four domains: communication (e.g., understanding language), socialization (e.g., interacting with others), daily living skills (e.g., feeding), and motor skills (e.g., moving). Similarly, pain in adults with DD has been associated with limitations in exercising, maintaining relationships, completing self-care activities and/or emotional distress such as depression, anxiety, and self-injury (Walsh et al., 2011).

Importantly, while the functional domains impacted by pain may be similar, the degree of impairment seems to be greater for people with DD compared to those without. For children and youth, this phenomenon is particularly the case as the severity of DD increases (Breau et al., 2007). This is likely due to a number of factors including the cognitive limitations experienced by people with DD, and preexisting limitations in adaptive functioning (Breau et al., 2003). Further, a number of behaviors less commonly seen in people without DD tend to occur in the presence of pain for people with DD. For both children and adults, "problem behavior" such as aggression may increase in both frequency and intensity in tandem with the level of pain experienced and/or pain-related indicators (Carr & Owen-DeSchryver, 2007; Courtemanche, Schroeder, Sheldon, Sherman, & Fowler, 2012). Further, it has been suggested that some of these behaviors such as self-injury may serve a communicative and/or regulatory purpose (Breau, Camfield, et al., 2003).

In recognizing the impact of pain on this population, the pediatric literature (the adult pain in DD literature is extremely limited) most clearly demonstrates that not only will pain lead to a decrease in functioning in the short term, but also in the long term it leads to decreased practice of skills leading to difficulty acquiring skills (e.g., Breau et al., 2007). As shown with children diagnosed with cerebral palsy, corresponding difficulties associated with poor adaptive functioning skills may in turn greatly impact the level of independence and support needed in daily living, participation in desired activities, and overall quality of life (e.g., Houlihan, O'Donnell, Conaway, & Stevenson, 2004). In sum, pain has a number of implications and addressing it to the best of one's ability could improve quality of life and functional ability of people with DD (Breau et al., 2007).

Challenges in Pain Assessment and Management

For people with DD, pain assessment and management are complex and challenging processes (Carter et al., 2002; Chen-Lim et al., 2012). Indeed, many of the challenges unique to this population stem from varied levels of cognitive and physical abilities as well as limited adaptive functioning skills (Breau, McGrath, & Zabalia, 2006; Doody & Bailey, 2017). A number of these challenges are discussed below.

First, self-report has historically been regarded as the "gold standard" in pain assessment, particularly given the subjectivity of the pain experience (Schiavenato & Craig, 2010; Twycross, Voepel-Lewis, Vincent, Franck, & von Baeyer, 2015). A number of limitations to this approach have been indicated in recent years, however, such as oversimplification of the pain experience (Twycross et al., 2015). Further, people with limited ability to effectively communicate through verbal or nonverbal means may be unable to utilize tools developed for self-reporting one's pain (Chen-Lim et al., 2012; Doody & Bailey, 2017; Dubois et al., 2010; Schiavenato & Craig, 2010; Temple et al., 2012). For example, pain assessment scales often rely on a number of cognitive abilities, such as understanding graded responses/seriation, in order to be used as intended (Ely, Chen-Lim, Carpenter, Wallhauser, & Friedlaender, 2016). If people with DD have not developed this understanding or are unable to follow directions to respond in the intended manner, self-report will be inaccurate (Schiavenato & Craig, 2010). As such, adaptations to the type of measures and questions asked when obtaining self-report from people with DD may be necessary to ensure the accuracy of these reports.

Second, although behavioral observations are often considered an important component of a multi-dimensional pain assessment approach, these too can prove difficult for people with DD. Indeed, pain in people with DD can be expressed through verbal and nonverbal pain behaviors, which may be different than those used by people without DD to communicate pain (Carter et al., 2002; Doody & Bailey, 2017; Dubois et al., 2010; Temple et al., 2012). Results from a systematic review have identified 14 types of behavioral pain indicators utilized by people with DD (de Knegt et al., 2013). The four most common types of pain-related expression include motor activity such as pacing, facial activity such as grimacing, social behavior such as one's interaction with others, and vocal expressions such as the production of distressing sounds (de Knegt et al., 2013). Beyond pain intensity, a number of factors may influence the types of behavior used to communicate pain such as verbal ability, mental age, and sex (de Knegt et al., 2013). Additionally, the expression of pain in people with DD can at times be atypical/idiosyncratic, unclear, and inconsistent (de Knegt et al., 2013). For example, a person with DD may become aggressive when they are tired, hungry, overwhelmed by noises/crowds, and when in pain. Further, a person may sometimes become aggressive but other times withdraw when experiencing pain. Behavioral cues may not always be reliable, and it is important to compare observations against what is typical for the person on pain-free days (Carter et al., 2002; Temple et al., 2012).

Third, a number of unique context-dependent barriers to assessing and managing pain may exist. For example, in school settings, maladaptive or problem behavior may be misinterpreted as defiance or work avoidance and may therefore represent missed opportunities for pain assessment (Quinn & Serna, 2017). Similarly, in school contexts it may at times be difficult to understand whose role it is to assess and manage pain, which may result in confusion and a lack of action (Quinn & Serna, 2017; Symons et al., 2008). Finally, pain is a subjective experience and given the difficulties with self-report described above, people with DD may rely on others to make judgments about their pain. It can be difficult for others to assess the pain of another person and this judgement is accompanied, and likely informed, by biases and prior beliefs. Thus, for certain carers such as those in health care, pain-related decision making processes may be highly subjective (Temple et al., 2012).

The Unique Role of Caregivers

Due to the limitations and challenges described above, caregivers of people with DD are often relied upon to ascertain whether pain is present and which interventions are needed (Findlay, Williams, Baum, & Scior, 2015). In this way, the pain assessment and management process is bidirectional (Craig, 2009), and consideration of the caregiver's contribution to these processes is important. For example, primary and secondary caregivers may hold pain and disability-related beliefs and biases (e.g., about pain sensation in people with DD) which have the potential to impact or predict care decisions (Beacroft & Dodd, 2011; Breau, MacLaren, McGrath, Camfield, & Finley, 2003; Findlay et al., 2015; Genik, McMurtry, & Breau, 2017; Shinde & Symons, 2007). Awareness of these is important, as research suggests that in its complexity, caregivers may fall for stereotypes, overlook, or discount pain (Carter et al., 2002; Hunt, Mastroyannopoulou, Goldman, & Seers, 2003). In pediatric DD literature, three knowledge domains have been identified as important for accurate pain assessment and management: "knowing the child," "familiarity with children with the same or similar conditions," and "knowing the science" (Hunt et al., 2003). This breadth of knowledge is likely to be uncommon in any one person (Hunt et al., 2003) and further highlights the need for collaborative and improved communication efforts between those caring for people with DD (Beacroft & Dodd, 2011).

Although often overlooked, parents and primary caregivers are important resources who can assist in the pain assessment and management process (Baldridge & Andrasik, 2010; Carter et al., 2002; Carter, Simons, Bray, & Arnott, 2016). This support from parents and primary caregivers typically continues into adulthood with respect to provision of pain ratings and access to medical and health services (Clarke, Thompson, Buchan, & Combes, 2008; McGuire et al., 2010; Walsh et al., 2011; Weissman-Fogel, Roth, Natan-Raav, & Lotan, 2015). Parents know their children well and have developed skills and competence specific to their child with respect to pain expression and capacity for self-report (Carter et al., 2002). Knowledge of an individual's baseline and history is critical to placing his/her behavior into context (Hunt et al., 2003). Parents of both children and adults with DD describe the development of their knowledge as an active process which involves guesswork, trial and error, and intuition (Carter et al., 2002; Clarke et al., 2008; Hunt et al., 2003). People with DD also report seeking assistance from their caregivers not only for pain assessment but also as much needed support (e.g., verbal children with autism spectrum disorder; Ely, Chen-Lim, Carpenter, Wallhauser, & Friedlaender, 2016). It is therefore critical to gather information from parents and other highly involved caregivers to assist in this regard (Carter et al., 2002; Clarke et al., 2008).

Importantly, primary caregivers are not always available to assist with pain assessment and management, and may seek the support of or rely on others in making pain-related decisions. Unfortunately, parents have expressed concerns that health professionals and other caregivers may treat their children differently due to their special needs, or lack knowledge to adequately assess, treat, and refer their loved ones to appropriate services (Carter et al., 2002). This can lead to parents feeling isolated in their journey and concerned that their children will need to accept and live with pain rather than having these needs met (Carter et al., 2002). Parents emphasize the need for and importance of skilled and knowledgeable professionals who can help them with their children's pain (Carter et al., 2002; Carter et al., 2016; Hunt et al., 2003).

In order for other caregivers to reach a more nuanced level of personal knowledge about those they support, time as well as a commitment to building relationships with and hearing the stories of people with DD and their families are required (Hunt et al., 2003). Unsurprisingly, pain assessment and management are viewed by secondary caregivers such as school nurses and residential support workers as difficult and complex; this is particularly true when they are lacking critical information or resources such as pain assessment tools (Beacroft & Dodd, 2011; Quinn, 2016). Although some research suggests that these caregivers may be able to identify important factors to consider when assessing pain and useful management strategies, their application may be suboptimal. Indeed, an audit of 33 residential services for adults with DD in Surrey, UK found that none of nine researcher-developed pain assessment and management standards were met (Beacroft & Dodd, 2011). These included items such as the recognition that people with DD do not have a higher pain threshold than others without DD, having access to appropriate pain assessment tools and having pain management policies in place (Beacroft & Dodd, 2011). Similarly, work regarding a variety of caregivers supporting children and adults with DD have highlighted deficits of pain-related knowledge, skills, confidence, and resources, and the need for targeted pain-related education (Beacroft & Dodd, 2011; Carter et al., 2016; Genik, McMurtry, Breau, Lewis, & Freedman-Kalchman, 2017; Malviya, Voepel-Lewis, Merkel, & Tait, 2005; Nilsson, Johnson, & Adolfsson, 2016; Quinn, 2016; Quinn & Serna, 2017).

Thus far, provision of pain assessment and management training programs in settings including pediatric respite, school, and adult residential settings have demonstrated positive impacts immediately following training and/or at follow-up. These include increases in pain-related knowledge, changes in perceptions and attitudes, and increased intent to implement/use new strategies in practice (Genik, McMurtry, Breau, Lewis, & Freedman-Kalchman, 2017; Mackey & Dodd, 2010; Quinn & Smolinski, 2017). The broad dissemination of these types of programs and resources is necessary in order to be of their intended use (Findlay et al., 2015).

Pain Assessment

As with people who do not have a DD diagnosis, there are a number of approaches used to assess pain and arrive at management-related decisions, such as self-report and observation. The use of multiple methods is recommended (Solodiuk et al., 2010). An understanding of pain assessment tools and resources is necessary but not sufficient, as "knowing the child" is also critical (Carter et al., 2002). Consultation with primary caregivers regarding the child's pain history and usual pain behavior patterns is always recommended and can improve assessment. While some of this information can be gleaned from informal conversations with caregivers, more formal communication using structured tools between caregivers may be helpful in this regard. For example, one might use the Pediatric Pain Profile (described below; Hunt et al., 2004) across settings to assist with pain assessment. Further, although not specific to pain, resources such as the health passport communication tools developed by the Health Care Access Research and Developmental Disabilities team have been successfully implemented in medical settings (Heifetz & Lunsky, 2018).

Self-Report

Despite the challenges described earlier, literature suggests that people with mild to moderate levels of impairment can generally provide some form of pain self-report; however, the approach used and depth of information provided may vary (Benini et al., 2004; Fanurik et al., 1999). For example, one study found that although 80% of children with Down syndrome were able to indicate when they were in pain, most were unable to describe how severe their pain was or where the pain was located (Valkenburg et al., 2015). In contrast, findings from a study with high functioning youth with autism spectrum disorder suggested they were able to provide self-reports to hypothetical scenarios no different than those reports of "typically developing" peers (Bandstra, Johnson, Filliter, & Chambers, 2012). In a pilot study of adults with Down syndrome, the majority (73%) could complete portions of a self-report measure of pain for people with intellectual disabilities, although only 18% could complete the entire measure without assistance from a trained caregiver (de Knegt, Lobbezoo, Schuengel, Evenhuis, & Scherder, 2016).

Research exploring the ability of people with DD to provide self-report highlights the importance and need for individualized or adapted approaches tailored to a person's developmental level (Benini et al., 2004; Dubois et al., 2010). For example, when asking for self-report, it is important to adapt the language used such as saying "hurt" or "cry" rather than "pain" and using words, pictures, or numbers as needed (Ely et al., 2016). Consideration of the child's preferences, such as type of self-report activity, and other abilities may also be important (Crosta, Ward, Walker, & Peters, 2014; Ely et al., 2016). For example, if someone is unable to respond verbally they may provide self-report by pointing or nodding. In other cases where physical limitations may prohibit the ability to point, people with DD may respond using assistive technology or by looking in the direction of their desired response.

Although psychometrics have generally not been explored in a population of people with DD, a number of self-report tools originally designed for "typically developing" children and/or adults have been successfully utilized in pediatric DD research (see Table 53.1). The use of these tools has largely remained unexplored in adult DD literature. In addition to those listed in Table 1, adaptations of formal self-report tools (e.g., modified Eland Color Scale and Cube Test; Benini et al., 2004) as well as the use of informal assessment approaches (e.g., iPad applications allowing children to draw their pain or indicate its location; Ely et al., 2016) have also been successfully applied in pediatric DD populations. In some cases, provision of a "training session" instructing children how to use the self-report measures prior to a medical procedure has also proven beneficial (Benini et al., 2004).

Recently, a self-report measure of pain specific to adults with DD was developed, called the Self-Reporting Tool on Pain in People with Intellectual Disabilities (STOP-ID!; de Knegt et al., 2016). The STOP-ID is an online tool that aims to capture various qualities of the pain experience, including: intensity, affect, quality, and location of the pain using graphic images and pictograms. The measure can be completed on either a laptop or a tablet device. This computer-based approach has the advantage of providing an efficient way to collect information, thereby reducing the burden on caregivers associated with monitoring and reporting of pain conditions. Although further research is needed, preliminary findings suggest this measure has the potential to enable people with DD to self-report pain and provide a standardized tool to cover multiple aspects of the pain experience (de Knegt et al., 2016).

The wide range of measures discussed here as well as results from preliminary work demonstrate that there is no one self-report measure that can be utilized for people with DD (Ely et al., 2016). Rather, an assortment of tools may be needed to meaningfully assess pain in this population (Ely et al., 2016). Further, it is important to keep in mind that these studies have largely focused on people with mild to moderate DD; thus, the efficacy of these tools in those who are more severely impacted by DD or have other comorbidities is unclear (Bandstra et al., 2012). For example, preliminary research indicates that adults with severe DD have difficulties using self-report tools and comprehending the questions posed (e.g., STOP-ID; de Knegt et al., 2016).

Behavioral Observations

As with self-report, observational tools and approaches used with people who are "typically developing" may lead to inaccurate judgements and underestimations of pain in people with DD given differential expression of pain (Dubois et al., 2010). A number of specialized behavioral observation tools have been developed and involve assessment of agreed upon behaviors commonly exhibited by people with DD when experiencing pain, such as facial expressions and vocalizations (Crosta et al., 2014). The goal of these measures is to improve the ease and accuracy with which observers make judgements about pain for people with DD. These measures have been implemented in various degrees in clinical settings, and differ in aspects such as format, time for completion and degree of primary caregiver input required (Crosta et al., 2014).

Table 53.1 Summary of self-report assessment measures used in research with children with DD

Self-report measure	Initial age range intended for use	Brief description of task	Link to original measure	Relevant DD-specific citation(s)
Measures of pain intensity				
Faces Pain Scale—Revised (Hicks, von Baeyer, Spafford, van Korlaar, & Goodenough, 2001)	4 to 19	Six drawings of faces representing pain ranging from 0 (*No Pain*) to 10 (*Very Much Pain*). The person providing self-report is asked to indicate which face best represents how he/she is feeling.	www.iasp-pain.org/FPSR	Bandstra et al. (2012) ∗Study using hypothetical scenarios; Faces Pain Scale—Revised converted from a 10-point scale to a 5-point scale to correspond with another measure used in the study
Hester Pieces of Hurt Tool (Hester, Foster, & Kristensen, 1990)	3 to 18	If the person providing self-report indicates that he/she is in pain, four red poker chips are provided to them. The person is told that each piece represents one piece of hurt, and are asked to choose "how many pieces of hurt" he/she has.	Attachment B7: http://www.homesteadschools.com/Nursing/courses/ManagementCancerPain/AttachmentB.html	Ely et al. (2016)
Numeric Rating Scale (von Baeyer, Chambers, Forsyth, Eisen, & Parker, 2013)	8 and above	Most typically, a scale ranging from 0 (*No Pain At All*) to 10 (*Most Pain Possible*). The person providing self-report is asked to indicate on paper or verbally the level of pain he/she is experiencing.	n/a	Bandstra et al., 2012 ∗Study using hypothetical scenarios; used a 0–5 scale for increased simplicity and to correspond with the number of response options from another measure used in the study
Visual Analogue Scale (Scott & Huskisson, 1976)	8 and above	On a 10 cm line (0 = "*No Pain*"; 10 = "*Severe Pain*," the person providing self report is asked to point to or mark a point on the line which represents the amount of pain he/she is experiencing.	n/a	Benini et al. (2004) Ely et al. (2016)
Wong Baker FACES Scale (Wong & Baker, 1988)	3 and above	Six cartoon faces ranging from "*No Hurt*" to "*Worst Hurt*". The person providing self-report is asked to select the face that is most representative of how he/she feels.	http://wongbakerfaces.org/types-of-access/	Ely et al. (2016)
Measures of pain intensity and location				
Eland Colour Scale (Eland & Anderson, 1977)	4 to 10	After assigning colours to four pain categories (No Pain, Mild Pain, Moderate Pain, Severe Pain), the person is asked to colour where he/she hurts using the corresponding colours based on pain severity.	Attachment B6: http://www.homesteadschools.com/Nursing/courses/ManagementCancerPain/AttachmentB.html	Benini et al. (2004)

Table 53.2 Summary of behavioral assessment measures for pain in children and adults with DD freely available online

Measure	Validation	Intended/ Validated age range	Acute pain	Chronic pain	Link to measure
Chronic pain scale for nonverbal adults with intellectual disabilities	∗Pilot study only Burkitt et al. (2009)	18 and above		X	http://pediatric-pain.ca/wp-content/uploads/2013/04/CPSNAID.pdf
Individualized numeric rating scale	Solodiuk et al. (2010)	6 to 18	X		http://www.marthaaqcurley.com/inrs.html
Non-communicating adult pain checklist	Lotan, Ljunggren, et al. (2009); Lotan, Moe-Nilssen, et al. (2009); Meir et al. (2012); Weissmann-Fogel, Roth, Natan-Raav, & Lotan, 2015	15 to 75	X		https://www.painbc.ca/sites/default/files/Non-Communicating%20Adult%20Pain%20Checklist.pdf
Non-communicating children's pain checklist—postoperative version	Breau, Finley, et al. (2002) French: Zabalia et al., 2011	3 to 18	X		http://pediatric-pain.ca/wp-content/uploads/2013/04/NCCPCPV_200901.pdf ∗Also available on pediatric-pain.ca in German and French
Non-communicating children's pain checklist—revised	Breau, McGrath, et al. (2002) German: Kleinknecht (2007) Swedish: Johanssen, Carlberg, & Jylli, 2009	3 to 18	X	X	http://pediatric-pain.ca/wp-content/uploads/2013/04/NCCPCR_200901.pdf ∗Also available on pediatric-pain.ca in German and Swedish
Pediatric pain profile	Hunt et al., 2004	1 to 18	X	X	http://www.ppprofile.org.uk/ppptooldownload.php?s=209 ∗Also available in Portuguese
Revised face, leg, activity, cry, and consolability scale	Malviya et al., 2006	4 to 21	X		http://ilemsc.org/PainMgt/rFLACC.pdf

They can be used on their own or in combination with other assessment modalities.

For the purpose of this chapter, only behavioral measures that are freely available online, have known psychometric properties in English, and are frequently discussed in the literature will be described in detail. A summary of these measures for both children and adults with DD can also be found in Table 53.2. Findings from individual pediatric studies or research groups have been used to make suggestions about which scale may hold the most clinical utility, but unfortunately, recommendations are not consistent (e.g., Chen-Lim et al., 2012; Crosta et al., 2014; Ely et al., 2016; Quinn, Seibold, & Hayman, 2015; Taddio et al., 2015). The variability may reflect the need for varied assessment approaches to meet the unique needs of each setting, observer and person with DD. Importantly, while some of these measures indicate guidelines and interpretations/cut off scores (e.g., r-FLACC; 0–3 represents mild pain; Malviya, Voepel-Lewis, Burke, Merkel, & Tait, 2006), these have only been used in research, and their accuracy may be questionable (Breau, McGrath, Camfield, & Finley, 2002). As such, there is potential for underestimation of pain, and these scores should not be the only method for determining the presence or absence of pain and subsequent treatment (Breau, McGrath, et al., 2002).

With respect to children and youth with DD, two versions of the Non-Communicating Children's Pain Checklist (NCCPC) have been developed: the Revised Version (30 items) and the Postoperative Version (27 items), both of which are designed for use with people with DD ranging from 3 to 18 years of age (Breau, Finley, McGrath, & Camfield, 2002; Breau, McGrath, et al., 2002). These measures ask caregivers to indicate the frequency with which a number of pain-related behaviors are observed over a period of time ranging from 0 (“*Not At All*”) to 3 (“*Very Often*”); the revised version uses a 2 h time frame whereas the postoperative version uses 10 min (Breau, Finley, et al., 2002; Breau, McGrath, et al., 2002). Subscale and total scores can then be derived, and may be compared to cut-off scores used in research as follows: revised version: > 7 = pain; postoperative version: 6–10 = mild pain and = 11 = moderate to severe pain (Breau, Finley, et al., 2002; Breau, McGrath, et al., 2002;). Both of these measures have demonstrated excellent validity and reliability which appear to be maintained when translated into other languages (Breau, Finley, et al., 2002; Breau, McGrath, et al., 2002; Johansson, Carlberg, & Jylli, 2010; Kleinknecht, 2007).

Based on the Face, Leg, Activity, Cry, and Consolability Scale (FLACC) initially designed for young children who have not yet developed verbal abilities, the revised version (r-FLACC) measures pain intensity by rating each of the five titular domains from 0 to 2 (Crosta et al., 2014; Malviya et al., 2006). This revised measure is individualized, allowing parents to add in specific behaviors unique to their child in each section. A total score is calculated by adding the scores from each subcategory; a total score from 0–3 represents mild pain, 4-6 moderate pain, and 7-10 severe pain (Malviya et al., 2006); again, as noted previously, these cut offs have been primarily used for research purposes. Both the English and Danish version of the r-FLACC have demonstrated excellent interrater reliability, construct validity and criterion validity, and can be used for people ranging in age from 4 to 21 (Malviya et al., 2006; Pedersen et al., 2015). This, combined with its advantages (e.g., quick to complete, easy to use) makes the r-FLACC a strong candidate for integration into practice (Quinn et al., 2015).

Adapted from more traditional numeric rating scales, the Individualized Numeric Rating Scale (INRS) ranges from 0 (“*No Pain*”) to 10 (“*Worst Possible Pain*”) and allows parents and other caregivers to provide behavioral descriptions which correspond with each intensity rating (Solodiuk et al., 2010). For example, a caregiver may indicate that a child screams when their pain intensity is at 10, but may withdraw socially when their pain intensity is at a 3. Previous work by Solodiuk and colleagues (2003, Solodiuk et al., 2010) has suggested that the r-FLACC can be used to assist parents in identifying pain behaviors to highlight on the INRS. Research with a sample of nonverbal children with DD ranging in age from 6 to 18 suggested good inter-rater reliability, construct validity, and convergent validity (Solodiuk et al., 2010).

The Pediatric Pain Profile (PPP) developed by Hunt et al. (2004) is an observational measure designed for use with children who have severe disabilities ranging from 1 to 18 years of age. This measure includes 20 items and was designed to simultaneously serve as a record/pain communication tool (Hunt et al., 2004). The PPP is an in-depth document including sections for caregivers to share information about the child’s pain history, baseline, and current pain behavior. It also allows parents and caregivers to complete ongoing pain assessments and track information related to pain assessments, outcomes, and interactions with other professionals (Hunt et al., 2004). With respect to ongoing pain assessments, caregivers are asked to rate the frequency of a series of pain-related behaviors (e.g., disturbed sleep, hard to console, appears withdrawn/depressed) during a specific time period from 0 (“*Never*”) to 3 (“*A Great Deal*”). Ratings can be summed and result in a total score ranging from 0 to 60, with higher scores representing high pain intensity. General interpretation patterns based on previous research are provided; however, it is acknowledged that each child’s range of pain behaviors will be unique. Evidence of construct, concurrent and face validity as well as interrater reliability of the PPP have been demonstrated (Hunt et al., 2004). An initial implementation study of the PPP in an in-patient setting suggested that although aspects of this measure were valued, there are a number of implementation barriers such as completion of pre-admission assessment, length of time it takes to complete, and the requirement for ongoing documentation (Hunt & Franck, 2011).

Observational methods are not as commonly employed with adults. However, a number of assessment tools have been developed from the NCCPC-R (Breau & Burkitt, 2009). The NCCPC-R was piloted in individuals over 18 years of age with ID by having two staff members from an adult residential treatment centre watch residents and rate their behaviors using the NCCPC-R. Items related to "jumping around," "screaming," and "floppy" were removed as they were not sensitive to pain in the adult sample. This resulted in the development of the Chronic Pain Scale for Nonverbal Adults with Intellectual Disabilities (CPS-NAID), a 24-item scale that requires caregivers to indicate the frequency of observed pain-related behaviors, ranging from 0 ("*Not At All*") to 3 ("*Very Often*"). This scale has demonstrated preliminary psychometric properties (Breau & Burkitt, 2009; Burkitt, Breau, Salsman, Sarsfield Turner, & Mullan, 2009). Given that the CPS-NAID includes a subset of items from the NCCPC-R and the NCCPC-PV, scores on the NCCPC as an adolescent can be compared to their scores as a young adult on the CPS-NAID, and thus allowing for monitoring of long-term conditions (Breau & Burkitt, 2009). Nevertheless, this scale has only been evaluated in one study and further research is needed before using this scale in clinical situations (Breau & Burkitt, 2009). The Non-Communicating Adult Pain Checklist (NCAPC) is an 18-item scale that was developed from the NCCPC-R to be used for adults with DD (Lotan et al., 2009). Similar to the NCCPC-R, caregivers rate the frequency of observed pain behaviors on a four point scale (Lotan, Ljunggren, et al., 2009). The behaviors are divided into six subcategories: vocal expression, emotional reaction, facial expression, body language, protective reactions, and physiological signs (Lotan, Ljunggren, et al., 2009). Scores can range from 0 (no pain behavior observed) to 54 (maximal duration of all pain behaviors observed). Research supports the validity and reliability of the NCAPC in the context of acute procedural pain and chronic pain, and this measure demonstrates high sensitivity to pain in adults at all levels of DD (Lotan, Ljunggren, et al., 2009; Lotan, Moe-Nilssen, Ljunggren, & Strand, 2009; Meir, Strand, & Alice, 2012; Weissman-Fogel et al., 2015).

Other measures developed for use with children and/or adults are not as widely used, may not be freely available in English, and psychometric properties may require further exploration for intended target audiences. Examples of these include the Checklist Pain Behavior Scale (original: Terstegen, Koot, de Boer, & Tibboel, 2003; reduced: Duivenvoorden, Tibboel, Koot, van Dijk, & Peters, 2006), Pain Behavior Checklist (van der Putten & Vlaskamp, 2011), Pain Evaluation Scale for Clients with Cerebral Palsy (Collignon & Giusiano, 2001), Pediatric Indicator for Communication Impaired Children (Stallard et al., 2002), and the Disability Distress Assessment Tool (DisDAT; Regnard et al., 2007).

Combining Subject and Objective Assessment Approaches

Given the above described inherent difficulties in measuring pain among individuals with motor, communicative, and/or intellectual impairment associated with DD, yet another approach is to pair observational ratings with an exam-based procedure with or without additional reports from others such as caregivers. The Pain and Discomfort Scale (PADS) is a pain assessment tool which has been used to characterize nonverbal behavioral pain reactivity/expression of people with DD during a standardized physical exam known as the Pain Examination Procedure (PEP; Bodfish, Harper, Deacon, & Symons, 2001). PADS is composed of 18 items across five domains such as vocal and facial expressions; all items were derived directly from the NCCPC (Breau, Finley, et al., 2002; Breau, McGrath, et al., 2002). Preliminary work with the PADS has demonstrated excellent reliability for clinical screening, content validity, and sensitivity to painful procedures (Bodfish et al., 2001; Phan, Edwards, & Robinson, 2005). A benefit of pairing the PADS with a standard examination procedure such as the PEP is the ability to ensure that all body areas are examined and observed for possible pain signs. The pain examination procedure (PEP) consists of 5 assessment steps (head, arms, hands, torso, legs/feet), with a 3-part observation at each step (pain sign present/absent, intermittent, continuous). During the PEP, the PADS take approximately 5 min to complete by a trained nurse, direct care staff, or other provider, including parents.

Special Illustration of Advancing the Field: Research Application of Combined Assessment Approach for People Who Are Nonverbal with Severe DD

In addition to pain assessment, combined approaches such as the PADS/PEP may also be used to explore and illustrate the associated challenges and complexities of understanding pain in people with DD who have complex health problems and communication needs. For example, Rett syndrome (RTT) is a rare X-linked developmental disability causing severe cognitive, motor and language/communication problems, occurring almost exclusively in girls (Neul et al., 2010). From a clinical care perspective, there are frequent health issues that when combined with communication impairments lead to an increased risk that pain may be overlooked or discounted. This is also complicated by the inclusion of pain insensitivity as part of the diagnostic criteria for the syndrome.

The initial support for pain insensitivity was derived from several case study reports in which the apparent absence of pain sensitivity/reactivity associated with nociceptive events (e.g., falls, exposure to fire/flame) was documented (Hagberg, 2002). In a large survey study, the majority of parents reported their daughters had apparent reduced pain sensitivity, although some did report increased sensitivity (Down et al., 2010). Importantly, however, in a subsequent survey where parents were asked more specific questions about their daughter's pain based on items from a validated nonverbal pain scale (NCCPC-R described above; Breau, McGrath, et al., 2002), the majority reported that their daughters experienced a great deal of chronic pain (Symons, Byiers, Tervo, & Beisang, 2013). Indeed, almost a quarter of the sample reported their daughters experiencing pain for greater than 1 week in the prior month. It was also found that the frequency of pain episodes significantly correlated with the number of pain sources and the number of chronic health conditions.

Thus, it seems as though parents will report on pain insensitivity as well as problems with chronic pain about the same population. Part of the issue may be in how pain is assessed (as we suggested above—both in how it is "asked" about and in the "sampling frame"—in what sort of time window) as well as "what kind of pain is it" (nociceptive acute, chronic, etc.). To this point, in a further investigation examining the relation between parent proxy report of pain symptoms and directly observed pain behaviors using the PADS/PEP combination as described above, Barney, Feyma, Beisang, and Symons (2015) documented that the majority of parents (89%) reported their daughter as having experienced pain in the previous week. Moreover, based on direct observation, pain expression appeared intact and frequent when coded observationally during the PEP consistent with behavioral pain expression in other nonverbal populations with severe DD.

Overall, the detailed subjective reports of pain experience provided by parents and objective coding of pain expression during a physical-based standardized exam in RTT suggests the issue surrounding pain in RTT may be a detection issue rather than a sensitivity issue. This specific finding may be part of a more general issue associated with complex cases associated with severe or profound DD. Severe motor and communicative impairments limit the ability to self-report subjective pain experiences in conventional ways. Because of this, there is the possibility that pain may be overlooked and undertreated; indeed, despite parental concerns about health, almost half (43%) of parents reported they were unsure how to reliably determine when their daughter was in pain (Symons et al., 2013). One can clearly understand through this example the complexities and bidirectional nature of the challenges in assessing and understanding pain in people with DD.

Pain Management

Unsurprisingly, research related to pain management in people with DD has also lagged behind that regarding those who are considered "typically developing." The following section will therefore highlight pain management more broadly, while also citing work which has investigated these strategies with people with DD. As with pain assessment, consideration of a person's abilities, preferences, and needs in addition to the completed pain assessment will help guide pain treatment; adaptations to certain approaches may be warranted. Strategies for pain management are often organized into categories known as the "three P's": pharmacological, psychological, and physical. Arguably, a fourth "P" also exists. Process interventions can be thought of as alterations of the environment surrounding the painful incident or painful procedure itself that could exacerbate one's pain experience (Riddell et al., 2015). For example, if a person is bothered by loud noises or crowds, their pain may be better managed in a quiet environment with limited carers present.

Importantly, preliminary work in medical settings has suggested a number of barriers which may interfere with the implementation of adequate management strategies including difficulty with pain assessment and related tools, reliable documentation, and inadequate pain-related knowledge specific to people with DD (Baldridge & Andrasik, 2010; Malviya et al., 2005). Despite the lack of research regarding the efficacy of pain management strategies for people with DD, these are still applied by caregivers in an attempt to relieve pain of those they support. For example, carers of children and adults with DD report having implemented a wide range of strategies including pharmacological (e.g., acetaminophen), psychological (e.g., distraction), and physical (e.g., massage; Carter, Arnott, Simons, & Bray, 2017; McGuire et al., 2010; Walsh et al., 2011).

Pharmacological

Analgesic pharmacological strategies are widely used and recommended for children and adults alike (e.g., Berde & Sethna, 2002; Walsh et al., 2011). Children and youth with high-functioning autism spectrum disorder have indicated that medication can help them to feel better when in pain (Ely et al., 2016). Unfortunately, however, little is known about the use and implications of pharmacological strategies for people with DD. Challenges with pain assessment as well as ethical considerations (e.g., consent) may be partially to blame for this dearth of research (e.g., Valkenburg et al., 2015). Valkenburg, van Dijk, de Klein, van den Anker, and Tibboel (2010); Valkenburg et al. (2015) have shared a "plea" for more research to be conducted in this area, and have spoken about the need to shift foci from mainly pain assessment to pain management. To date, only a handful of pharmacology-related studies have been conducted, and they have focused largely on surgical procedures (e.g., Koh, Fanurik, Harrison, Schmitz, & Norvell, 2004; Malviya et al., 2001). Importantly, when people with DD report high levels of pain and are considered to be in pain by observers, they still may not receive pharmacological treatment or they may receive fewer analgesics than the general population (Boerlage et al., 2013; Hodgkinson et al., 2001).

Medication management of medical comorbidities in people with DD can complicate pharmacological management of pain (Taddio & Oberlander, 2006). Careful consideration of potential side effects is crucial when prescribing medication for pain in people with DD (Taddio & Oberlander, 2006). For example, when prescribing medications of a certain analgesic drug class (e.g., opioids, nonsteroidal anti-inflammatories, acetaminophen), it is important to think about the person's underlying condition, relevant contraindications and potential drug interactions prior to prescribing (Taddio & Oberlander, 2006). Ensuring that communication is open and ongoing across caregivers will be helpful in ensuring that people with DD do not experience unnecessary side effects and instead the desired effects of decreased pain (Taddio & Oberlander, 2006). Resources in relation to pharmacological management of pain are limited for people with DD; readers are encouraged to consult Taddio and Oberlander (2006) for further information regarding pharmacological interventions for pain in children with DD.

Psychological

Psychological management strategies have been broadly studied and found effective for both acute and chronic pain (e.g., Birnie et al., 2015; Eccleston et al., 2014; McManus, Treacy, & McGuire, 2014; Palermo, Eccleston, Lewandowski, Williams, & Morley, 2010; Uman et al., 2013; Williams, Eccleston, & Morley, 2012). Examples of these strategies are provided in Table 53.3. Critically, however, the majority of these strategies have not been formally examined in DD literature, and none of these strategies have substantive research support for their use with this population.

Based on various individual factors (e.g., developmental level, preferences), adaptations to these management strategies may be made. For example, some people may require the assistance of caregivers to utilize distraction, while others may be able to engage in this independently. Similarly, some people may require breathing props such as bubbles to help them engage in and successfully apply deep breathing. Finally, more concrete approaches may be important to help people with DD master these skills such as using a stress ball to teach how to squeeze and release in progressive muscle relaxation.

These strategies are often used across settings. In some cases, people less severely impacted by DD may be able to discuss and implement these approaches. For example, youth in a study by Ely et al. (2016) spoke about the helpful use of distraction such as engaging in preferred activities. Notably, psychological strategies have also been successfully used in combination with others such as topical anesthetics and positive reinforcement (Slifer et al., 2011).

A widely used and commonly researched class of psychological interventions for both acute and chronic pain fall under the umbrella of cognitive behavioral therapy (CBT), which aim to foster an individual's self-management (Logan et al., 2014; McManus et al., 2014). The goal is to enhance an individual's adaptive coping in order to resume their daily life, despite the pain (Turk, 2003). Preliminary research has incorporated modified

Table 53.3 Brief descriptions and examples of commonly used psychological strategies for pain management

Strategy	Description	Examples
Distraction	Activities aimed at shifting an individual's attention away from pain through presentation of other stimuli (Cohen, Cousins, & Martin, 2014).	Interactive toys, video games, virtual reality; auditory distraction (music); audiovisual distraction (television, online videos), Slifer et al. (2011) Koller and Goldman (2012).
Imagery	Involves having the individual imagine a safe and comfortable situation.	Guided imagery: encouraged to let their minds wander, imagine, and focus on a scene/environment that is pleasurable, peaceful, or relaxing. Can be facilitated using audio prompts. Logan, Coakley, and Barber Garcia (2014)
Progressive muscle relaxation	Systematically relaxing body parts, at times following brief periods of tensing the muscle.	See Cautela and Groden (1978) for progressive muscle relaxation for individuals with special needs.
Hypnosis	Dissociation from painful experience and distress via hypnotic induction, suggestions, and imagined fantasy, similar to but more involved than imagery; requires specialized training.	Koller and Goldman (2012)
Deep breathing	Breathing with long breaths, deliberately pacing the breath.	Bubble blowing, blowing exercises.

CBT in the treatment of chronic pain in adults with DD (McManus et al., 2014). An eight-session manualized intervention program called Feeling Better was included, which aimed to increase pain management knowledge and coping skills (e.g., increase wellness-focused coping and reduce illness-focused coping). Following the intervention, participants had higher ratings of pain management knowledge and wellness-focused coping; however, gains were not maintained at one-month follow-up. Additional research examining a prolonged and intensive intervention incorporating caregivers is required (McManus et al., 2014). A group-based CBT treatment for menstrual pain management has been developed from the Feeling Better program for young women with DD, although results from this clinical trial have not yet been released (Kennedy, O'Higgins, Sarma, Willig, & McGuire, 2014).

Physical

Similar to psychological management strategies, a number of physical strategies may be effective in managing both acute and chronic pain (Rakel & Barr, 2003; Taddio et al., 2015). Examples of physical strategies include positioning, stretches, hot/cold compresses, hugs, and exercise. Physical therapy is an integral part of chronic pain treatment (Tupper, Swiggum, O'Rourke, & Sangster, 2013). Particular physical techniques have been recommended to reduce vaccination related pain in all populations (Taddio et al., 2015). Many complementary and alternative medicine approaches such as massage therapy and acupuncture may be helpful in mitigating experiences of pain (Breau, Lotan, & Koh, 2011). In this way, involvement of other professionals may be beneficial when considering interventions such as massage or stretching. However, techniques such as massage, vibration and acupuncture may inflict sensations similar to procedural pain; thus, the ability to understand and tolerate these types of strategies should be considered for individuals with DD (Doody & Bailey, 2017).

Conclusions and Future Directions

Pain in people with DD is prevalent and can greatly impact all areas of adaptive functioning and quality of life. There has been some increasing research interest in particular topics regarding pain assessment for people with DD. Yet as a whole, this is a very understudied research area and pain continues to be under-recognized and mismanaged among this group of people. A number of intra and interpersonal challenges play a role in some of these difficulties. For example, people with DD often struggle to communicate their pain effectively through self-report, and may express pain in idiosyncratic, atypical, and/or inconsistent ways leading to difficulties associated with behavioral observation. Caregivers play an important role in assisting with pain assessment and management, yet may hold inaccurate pain-related beliefs that could negatively impact care. Further, limited pain-specific education is available for caregivers despite their unique role and the risks associated with under-recognized and mismanaged pain. Future work is needed in all areas related to pain in people with developmental disabilities, and it will be important to include not only caregivers of those with DD but also people with DD themselves.

First, while some basic information can be gleaned from prevalence studies previously conducted, there are no larger scale studies examining pain prevalence, for example. Further, little is truly known or understood about pain sensitivity and expression. Developing a better understanding of pain sensitivity, for example, will be helpful in informing pain assessment, management, and related care for people with DD.

Notably, not only is assessment important to better understand a person's difficulties, but it is also critical to inform treatment approaches. With respect to self-report, emerging research continues to suggest that people with DD may be able to provide self-reports of their pain when measures are adapted and explicitly taught. Pain is subjective; providing people with DD a fair opportunity to share their pain experiences is important. To date, there are no validated measures or approaches for gathering self-report in this population. Further work in this area to develop a better understanding of which measures may be useful for people with DD and how they can be adapted will therefore be useful. Although a number of behavioral observation scales have been developed and validated specific to people with DD, these are largely intended for use in medical settings. Focusing on pain assessment beyond that which occurs when primary caregivers are present or in health care settings could be helpful in improving other caregivers' abilities to ensure that these children's needs in this regard are adequately met.

Intervention strategies have not been systematically evaluated. This area has lagged well behind even that of pain assessment for people with DD. Specifically, understanding which strategies are most helpful to people with DD and how to best adapt/apply them in clinical settings will be critical to ensuring pain is managed adequately. This is the case for both pharmacological and nonpharmacological strategies.

Finally, there has been a growing body of literature associated with development of education and resources to improve caregivers' abilities across settings to effectively assess and manage pain in people with DD (e.g., Genik, McMurtry, Breau, Lewis, & Freedman-Kalchman, 2017; Mackey & Dodd, 2010; Quinn & Smolinski, 2017). These efforts have been positively endorsed by study participants (e.g., Genik, McMurtry, Breau, Lewis, & Freedman-Kalchman, 2017). Continued efforts in this domain will be helpful to ensure that all caregivers of people with DD have adequate training and baseline pain assessment and management skills. Given the tendency to under-recognize pain, more information could help lessen the likelihood that pain is missed among people with DD. Additional efforts in ensuring that new knowledge continues to be shared with and translated to a lay audience will also be critical in ensuring that people with DD receive the best possible support and care.

Case Study 1

Ellory is a 13-year-old girl diagnosed with autism spectrum disorder and a mild intellectual disability. She is in good health and has started menstruation last month. Although Ellory is able to communicate her needs relatively well and understands simple instructions, she tends to be slow to process information and becomes overwhelmed if it is provided too quickly. Further, she struggles with abstract concepts and often requires problems to be described in concrete terms or accompanied by aids, such as images or manipulatives. Ellory responds best when her environment and the responses of others are predictable and consistent.

Ellory attends school full time, spending the first part of her day in a mainstream Grade Eight class, and the remainder of the day in a class specifically for children with mild intellectual disabilities. Her favorite subject is art and her least favorite is math. Ellory has an educational assistant, Mrs. Smith, who supports her on an as-needed basis in both classrooms. Ellory has known Mrs. Smith since Grade Three, and they have a strong relationship. Ellory's parents have worked closely with Mrs. Smith to facilitate communication (e.g., advocating for her needs, communicating how she feels) and employ consistent behavioral strategies at home and at school. Each morning and afternoon, Ellory colors a picture of a person to show how she feels. Ellory's parents use a structured document outlining how to recognize when she is in pain and help her feel better. This document outlines pain behaviors that Ellory may exhibit when in pain, self-report approaches, and strategies that tend to help Ellory feel better (see Fig. 53.1, as developed by Genik, Millett, & McMurtry, 2016[1]). Ellory's parents have also developed a 'pain management menu' at home

---

[1] Currently under review with a peer reviewed journal, this resource as developed by Genik this resource as developed by Genik, Millett, and McMurtry (2016) addresses initial feasibility and usability of the resource in respite care settings. Future work will also include implementation of this resource in secondary care settings.

How to Tell if My Child is in Pain

(Please circle all answers that apply for each question)

When in pain...

1) What facial expression does your child show?

Changes in mouth Furrowed brow
[✓ Changes in eyes] Grimace Stone face
Grinding teeth None
Other/Describe: Squinches her eyes sometimes

2) Does your child's sleeping patterns change?

Sleeps more Sleeps regularly Sleeps Less
Sleeps on and off Restless [✓ No]
Other/Describe:

3) Does your child avoid eating?

Yes Sometimes [✓ No]

4) Does your child's breathing change?

Holds breath Gasps [✓ No]
Other/Describe :

5) Does your child tense his/her muscles or become rigid?

Tenses muscles Becomes rigid [✓ No]
Other/Describe:

6) Does your child show you the area of the body where the pain is located?

[✓ Touch/ grasp area] Gesture at area Guard/Protect Area
Favour area Move a certain way No
Other/Describe: Only sometimes

7) Does your child become sweaty or clammy ?

Sweaty / Clammy [✓ No]
Other/Describe:

8) Does your child's comfort seeking behaviour change?

Seeks emotional comfort Seeks physical comfort
Withdraws from comfort [✓ No]
Other/Describe:

9) Does your child's behaviour change?

Irritable Agitated Uncooperative
Distracted [✓ Aggressive]
Fidgety/ cannot sit still No
Other/Describe: Verbally and physically - will say 'shut up', hit or bite

10) Does your child's activity level change?

Loses interest Less active
More active [✓ No]
Other/Describe:

11) Does your child make vocal sounds?

Whimpering Screaming Crying
Grunting Groaning No
Other/Describe: If asked directly, can tell you if she is in pain; non-verbal response options are best

12) Does your child engage in any atypical behaviour ?

Self-injury Smiling
Laughing [✓ No]
Other/Describe :

Ways to Help My Child Feel Better

(Please circle all answers that apply for each question)

When in pain…

1) Does physical comfort help soothe your child ?

No Rubbing area Hugging
[✓ Other/Describe]: Hot/Cold Compress

2) Does your child self– soothe?

No Yes [✓ Sometimes]

3) Are distractions helpful in managing the pain?

No [✓ Yes] Sometimes

If yes, what are your child's favourite distractions?

Favourite toy(s): Art/colouring, puzzles

Favourite comfort(s): Bean bag chair; stuffies and other soft items

Favourite movie/ T.V show(s): No specific show; loves music

Favourite Snack(s): Fish crackers

Other/Describe: Blowing bubbles
*Can have ibuprofen for headaches and stomach aches

4) Has your child had experience with another secondary caregiver who would be available for further consultation?

If yes, what is the caregiver's:

Name: Mrs. Smith at school

Number: (   )   -   -

Parents: Please make sure to fill out the back of the pamphlet →

Fig. 53.1 Snapshot of communication document between Ellory's parents and school staff outlining ways to help Ellory feel better when she is experiencing pain

which allows Ellory to choose from a variety of pain management options (see Fig. 53.2).

This week, Ellory's parents indicated that she may be nearing her time of menstruation and may be prone to stomach cramps. During math class, Mrs. Smith began to notice Ellory becoming more irritable (e.g., skipping over questions that were difficult rather than persevering as she normally would, mumbling angrily to herself). As this is common for Ellory given her difficulty with and dislike of math, Mrs. Smith provided some additional support, and also tried moving Ellory to the resource room where she could work in a more quiet setting. When this additional support did not appear to alleviate Ellory's frustration and upset, Mrs. Smith continued to monitor closely for other shifts in behavior. She noticed that Ellory was holding her stomach with one hand while working, which can be a sign that she may be experiencing pain. Mrs. Smith then used simple language consistent with that used at home, to ask if she had any hurt. Mrs. Smith allowed Ellory to respond nonverbally by pointing to her response (yes/no) on a piece of paper. She then provided Ellory with the picture of the person she colors each morning, and asked Ellory to show where her pain was. Ellory pointed to the person's stomach, and colored the stomach "red" to represent "a lot" of pain.

Based on parent requests, Mrs. Smith allowed Ellory to remain in the quieter resource room setting and provided Ellory with a physician approved amount of ibuprofen to help alleviate her stomach cramps. Mrs. Smith obtained the "pain management menu" that was co-created with Ellory and allowed her to select what she needed to help her feel better. Mrs. Smith provided Ellory with a hot compress for her stomach, and she went to lay down in the sensory break room. Ellory listened to music while laying

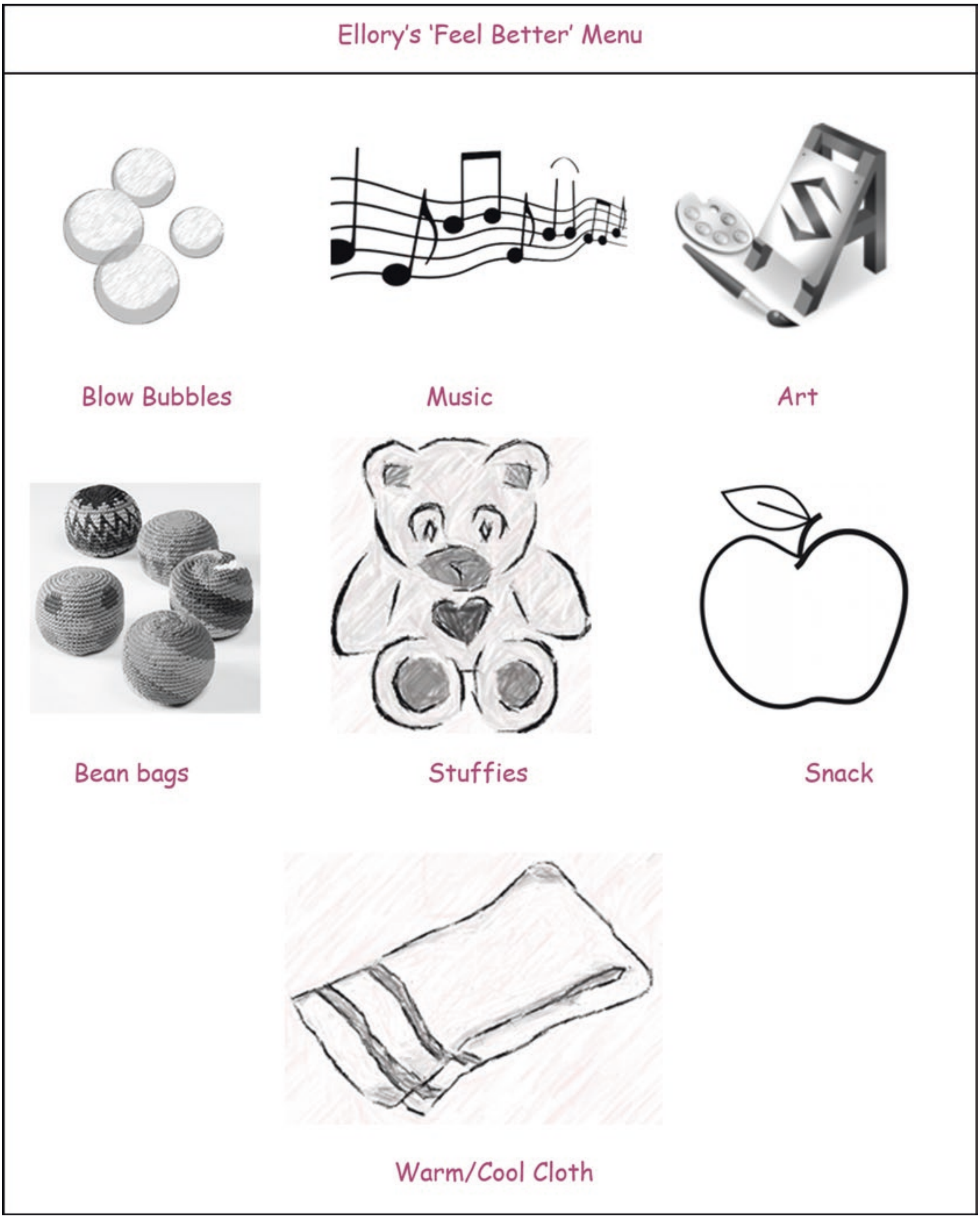


Fig. 53.2 Ellory's "pain management menu"

in the bean bag chair. Mrs. Smith continued to check in with Ellory to determine when the medication began to take effect and when she was feeling better. After about 30 min, Ellory colored another picture of a person with a yellow crayon, indicating that she only had "a little" pain in her stomach. Mrs. Smith helped Ellory return to her classroom to participate in the remaining activities. Mrs. Smith continued to monitor and assess Ellory's pain through observation and self-report for the remainder of the day. When Ellory's parents picked her up from school. Mrs. Smith provided them with a brief report of what occurred, actions that were taken, and the time Ellory took the initial dose of ibuprofen.

Case Study 2

Colton is a 40-year-old man diagnosed with a severe intellectual disability. He is generally in good health, but is lactose intolerant and experiences severe stomach aches after ingesting products containing lactose. Colton is able to understand very basic language but has extremely limited abilities to express himself through verbal means. For example, he generally speaks only with single words and simple sentences. Further, while he is able to respond to questions with "yes" or "no" these responses are at times acquiescent in nature or inconsistent. He uses a tablet to communicate many of his needs, such as when he is feeling hungry, through pictures.

Colton lives in a residential setting with three other people who have varying levels of DD. Supports are provided 24/7 by a core group of full time staff. He looks forward to weekly visits with his parents. Colton is generally a happy-go-lucky and easy going gentleman who enjoys taking walks, swimming, and helping with food preparation before meals. Although not validated for adults with DD, Colton's support workers use adapted documents derived from the Pediatric Pain Profile (Hunt et al., 2004) to maintain updated, relevant and organized information about Colton's pain history and his behavior "on a good day" versus "when in pain" (see Fig. 53.3 for example of Colton's behavior when in pain). They use an individualized document adapted from the Non-Communicating Adult Pain Checklist (NCAPC; Lotan, Ljunggren, et al., 2009) to assess his pain based on his individualized behaviors. As Colton spends at least one full day per week visiting his parents, Colton's support workers and parents maintain open communication about any pain he experiences and how he communicates it to others.

Colton's ability to understand and use verbal language to communicate often decreases when pain is present, making it more challenging to communicate with him. He does have pain-related pictures that he can select on his tablet, but uses these inconsistently. Knowing this, caregivers often ask for self-report but also rely on the use of structured behavioral observation measures to make judgments about the presence or absence of pain. Colton's physician has provided approval for him to take acetaminophen when in pain, and he responds well to the use of distraction. Often, Colton wishes to stay in his room when in pain, so support staff respect this and support him there as needed. As he does not respond positively to touch when in pain, staff generally do not use physical pain management strategies.

On a morning when Colton was to spend the day with his parents, Colton woke up 2 h earlier than usual (5 a.m.). When Sarah, the overnight support worker tried to support him in beginning his morning routine, Colton refused to get off of the living room couch. He refused breakfast as well as the opportunity to prepare breakfast for his housemates. Around 7 a.m., he began to grind his teeth. As Sarah's shift was ending, she consulted with the replacement staff member Celeste and they decided to document their observations by completing an ongoing pain assessment sheet. Although Colton often protects the area in pain, he seemed to be holding his face and continued to open and close his mouth. When Celeste showed Colton a picture he usually associates with headaches, he responded "NO" on his tablet. Nevertheless, Celeste decided that Colton was experiencing some form of pain and to try and help to alleviate it.

Based on documentation of pain management strategies that are most helpful for Colton, he was given a physician approved dose of an over-the-counter pain-relieving medication (acetaminophen) and his tablet to watch his favorite show. Celeste then contacted Colton's parents to provide them with an update and share her observations. Given the unclear and unusual nature of Colton's pain expression (i.e., opening and closing his mouth), his parents offered to take him to a walk-in clinic. Upon their return, they shared with Celeste that Colton had a toothache. They provided Colton with prescription medication provided by the

When in pain Colton&	Not at all	A little	Quite a lot	A great deal
• Withdraws from social situations			X	
• Becomes uncooperative in daily routines				X
• Sleeps less		X		
• Eats less		X		
• Grinds his teeth			X	
• Protects/guards areas in pain			X	

Fig. 53.3 Sample of baseline document used to communicate Colton's pain behaviors adapted from the Pediatric Pain Profile (Hunt et al., 2004)

doctor at the walk-in clinic, and made an appointment with his dentist for the following day. Celeste continued to check on Colton throughout the afternoon and assisted with the use of distraction as needed. Finally, Celeste recorded relevant information in Colton's adapted NCAPC and shared this information with the support worker covering the overnight shift.

References

American Academy of Pediatrics. (2001). The assessment and management of acute pain in infants, children, and adolescents. *Pediatrics, 108*(3), 793–797.

Baldridge, K. H., & Andrasik, F. (2010). Pain assessment in people with intellectual or developmental disabilities. *AJN The American Journal of Nursing, 110*(12), 28–35.

Bandstra, N. F., Johnson, S. A., Filliter, J. H., & Chambers, C. T. (2012). Self-reported and parent-reported pain for common painful events in high-functioning children and adolescents with autism spectrum disorder. *The Clinical Journal of Pain, 28*(8), 715–721.

Barney, C. C., Feyma, T., Beisang, A., & Symons, F. J. (2015). Pain experience and expression in Rett syndrome: Subjective and objective measurement approaches. *Journal of Developmental and Physical Disabilities, 27*(4), 417–429.

Beacroft, M., & Dodd, K. (2011). 'I Feel Pain'–audit of communication skills and understanding of pain and health needs with people with learning disabilities. *British Journal of Learning Disabilities, 39*(2), 139–147.

Benini, F., Trapanotto, M., Gobber, D., Agosto, C., Carli, G., Drigo, P., et al. (2004). Evaluating pain induced by venipuncture in pediatric patients with developmental delay. *The Clinical Journal of Pain, 20*(3), 156–163.

Birnie, K. A., Chambers, C. T., Taddio, A., McMurtry, C. M., Noel, M., Riddell, R. P., & Shah, V. (2015). Psychological interventions for vaccine injections in children and adolescents: Systematic review of randomized and quasi-randomized controlled trials. *The Clinical Journal of Pain, 31*(Suppl 10), S72.

Bodfish, J. W., Harper, V. N., Deacon, J. R. & Symons, F. J. (2001). Identifying and measuring pain in persons with developmental disabilities: A manual for the Pain and Discomfort Scale (PADS).. *Western Carolina Center Research Reports*.

Boerlage, A. A., Valkenburg, A. J., Scherder, E. J., Steenhof, G., Effing, P., Tibboel, D., & van Dijk, M. (2013). Prevalence of pain in institutionalized adults with intellectual disabilities: A cross-sectional approach. *Research in Developmental Disabilities, 34*(8), 2399–2406.

Bottos, S., & Chambers, C. T. (2006). The epidemiology of pain in developmental disabilities. In F. Symons & T. Oberlander (Eds.), *Pain in children and Adults with Developmental Disabilities* (pp. 67–87). Baltimore: Paul H. Brookes.

Breau, L. M., & Burkitt, C. (2009). Assessing pain in children with intellectual disabilities. *Pain Research and Management, 14*(2), 116–120. https://doi.org/10.1155/2009/642352

Breau, L. M., & Camfield, C. S. (2011). Pain disrupts sleep in children and youth with intellectual and developmental disabilities. *Research in Developmental Disabilities, 32*(6), 2829–2840.

Breau, L. M., Camfield, C. S., McGrath, P. J., & Finley, G. A. (2003). The incidence of pain in children with severe cognitive impairments. *Archives of Pediatrics & Adolescent Medicine, 157*(12), 1219–1226.

Breau, L. M., Camfield, C. S., McGrath, P. J., & Finley, G. A. (2007). Pain's impact on adaptive functioning. *Journal of Intellectual Disability Research, 51*(2), 125–134.

Berde, C. B., & Sethna, N. F. (2002). Analgesics for the treatment of pain in children. *New England Journal of Medicine, 347*(14), 1094–1103.

Breau, L. M., Finley, G. A., McGrath, P. J., & Camfield, C. S. (2002). Validation of the non-communicating children's pain checklist–postoperative version. *Anesthesiology: The Journal of the American Society of Anesthesiologists, 96*(3), 528–535.

Breau, L. M., Lotan, M., & Koh, J. L. (2011). Pain in individuals with intellectual and developmental disabilities. In *Neurodevelopmental Disabilities* (pp. 255–276). Dordrecht: Springer.

Breau, L. M., MacLaren, J., McGrath, P. J., Camfield, C. S., & Finley, G. A. (2003). Caregivers' beliefs regarding pain in children with cognitive impairment: Relation between pain sensation and reaction increases with severity of impairment. *The Clinical Journal of Pain, 19*(6), 335–344.

Breau, L. M., McGrath, P. J., Camfield, C. S., & Finley, G. A. (2002). Psychometric properties of the non-communicating children's pain checklist-revised. *Pain, 99*(1–2), 349–357.

Breau, L. M., McGrath, P. J., & Zabalia, M. (2006). Assessing pediatric pain and developmental disabilities. In F. Symons & T. Oberlander (Eds.), *Pain in children and adults with developmental disabilities* (pp. 67–87). Baltimore: Paul H. Brookes.

Burkitt, C., Breau, L. M., Salsman, S., Sarsfield Turner, T., & Mullan, R. (2009). Pilot study of the feasibility of the Non-Communicating Children's Pain Checklist Revised for pain assessment for adults with intellectual disabilities. *Journal of Pain Management, 2*(1), 37–49.

Carr, E. G., & Owen-DeSchryver, J. S. (2007). Physical illness, pain, and problem behavior in minimally verbal people with developmental disabilities. *Journal of Autism and Developmental Disorders, 37*(3), 413–424.

Carter, B., Arnott, J., Simons, J., & Bray, L. (2017). Developing a sense of knowing and acquiring the skills to manage pain in children with profound cognitive impairments: Mothers' perspectives. *Pain Research and Management, 2017*.

Carter, B., McArthur, E., & Cunliffe, M. (2002). Dealing with uncertainty: Parental assessment of pain in their children with profound special needs. *Journal of Advanced Nursing, 38*(5), 449–457.

Carter, B., Simons, J., Bray, L., & Arnott, J. (2016). Navigating uncertainty: Health professionals' knowledge, skill, and confidence in assessing and managing pain in children with profound cognitive impairment. *Pain Research and Management.* https://doi.org/10.1155/2016/8617182

Cautela, J. R., & Groden, J. (1978). *Relaxation: A Comprehensive Manual for Adults, Children, and Children with Special Needs*. Champaign: Research Press Company.

Chen-Lim, M. L., Zarnowsky, C., Green, R., Shaffer, S., Holtzer, B., & Ely, E. (2012). Optimizing the assessment of pain in children who are cognitively impaired through the quality improvement process. *Journal of Pediatric Nursing, 27*(6), 750–759.

Clarke, Z. J., Thompson, A. R., Buchan, L., & Combes, H. (2008). Parents' experiences of pain and discomfort in people with learning disabilities. *British Journal of Learning Disabilities, 36*(2), 84–90.

Cohen, L. L., Cousins, L. A., & Martin, S. R. (2014). Procedural pain distraction. In P. J. McGrath, B. J. Stevens, S. M. Walker, & W. T. Zempsky (Eds.), *Oxford Textbook of Paediatric Pain* (pp. 553–559). Oxford: Oxford University Press.

Collignon, P., & Giusiano, B. (2001). Validation of a pain evaluation scale for patients with severe cerebral palsy. *European Journal of Pain, 5*(4), 433–442.

Courtemanche, A., Schroeder, S., Sheldon, J., Sherman, J., & Fowler, A. (2012). Observing signs of pain in relation to self-injurious behaviour among individuals with intellectual and developmental disabilities. *Journal of Intellectual Disability Research, 56*(5), 501–515.

Craig, K. D. (2009). The social communication model of pain. *Canadian Psychology, 50*, 22–32. https://doi.org/10.1037/a0014772

Craig, K. D., Versloot, J., Goubert, L., Vervoort, T., & Crombez, G. (2010). Perceiving pain in others: Automatic and controlled mechanisms. *The Journal of Pain, 11*(2), 101–108. https://doi.org/10.1016/j.jpain.2009.08.008

Crosta, Q. R., Ward, T. M., Walker, A. J., & Peters, L. M. (2014). A review of pain measures for hospitalized children with cognitive impairment. *Journal for Specialists in Pediatric Nursing, 19*(2), 109–118.

de Knegt, N. C., Lobbezoo, F., Schuengel, C., Evenhuis, H. M., & Scherder, E. J. (2016). Self-reporting tool on pain in people with intellectual disabilities (STOP-ID!): A usability study. *Augmentative and Alternative Communication, 32*(1), 1–11. https://doi.org/10.3109/07434618.2015.1100677

de Knegt, N. C., Pieper, M. J., Lobbezoo, F., Schuengel, C., Evenhuis, H. M., Passchier, J., & Scherder, E. J. (2013). Behavioral pain indicators in people with intellectual disabilities: A systematic review. *The Journal of Pain, 14*(9), 885–896. https://doi.org/10.1016/j.jpain.2013.04.016

Defrin, R., Pick, C. G., Peretz, C., & Carmeli, E. (2004). A quantitative somatosensory testing of pain threshold in individuals with mental retardation. *Pain, 108*(1–2), 58–66.

Doody, O., & Bailey, M. E. (2017). Pain and pain assessment in people with intellectual disability: Issues and challenges in practice. *British Journal of Learning Disabilities, 45*(3), 157–165.

Doth, A. H., Hansson, P. T., Jensen, M. P., & Taylor, R. S. (2010). The burden of neuropathic pain: A systematic review and meta-analysis of health utilities. *Pain, 149*(2), 338–344.

Down, J., Geranton, S. M., Bebbington, A., Jacoby, P., Bahi-Buisson, N., Ravine, D., et al. (2010). Linking MECP2 and pain sensitivity: the example of Rett syndrome. *American Journal of Medical Genetics, 152A*, 1197–1205.

Dubois, A., Capdevila, X., Bringuier, S., & Pry, R. (2010). Pain expression in children with an intellectual disability. *European Journal of Pain, 14*(6), 654–660.

Duivenvoorden, H. J., Tibboel, D., Koot, H. M., van Dijk, M., & Peters, J. W. B. (2006). Pain assessment in profound cognitive impaired children using the Checklist Pain Behavior; Is item reduction valid? *Pain, 126*(1–3), 147–154.

Eccleston, C., Palermo, T. M., Williams, A. C. D. C., Lewandowski Holley, A., Morley, S., Fisher, E., & Law, E. (2014). Psychological therapies for the management of chronic and recurrent pain in children and adolescents. *The Cochrane Library, 5*, 1–78. https://doi.org/10.1002/14651858.CD003968.pub4

Eland, J. M., & Anderson, J. E. (1977). *The experience of pain in children* (pp. 453–473). Pain: A source book for nurses and other health professionals.

Ely, E., Chen-Lim, M. L., Carpenter, K. M., Wallhauser, E., & Friedlaender, E. (2016). Pain assessment of children with autism spectrum disorders. *Journal of Developmental & Behavioral Pediatrics, 37*(1), 53–61.

Fanurik, D., Koh, J. L., Schmitz, M. L., Harrison, R. D., Roberson, P. K., & Killebrew, P. (1999). Pain assessment and treatment in children with cognitive impairment: A survey of nurses' and physicians' beliefs. *The Clinical Journal of Pain, 15*(4), 304–312.

Findlay, L., Williams, A. C. D. C., Baum, S., & Scior, K. (2015). Caregiver experiences of supporting adults with intellectual disabilities in pain. *Journal of Applied Research in Intellectual Disabilities, 28*(2), 111–120.

Genik, L. M., McMurtry, C. M., & Breau, L. M. (2017). Caring for children with intellectual disabilities part 1: Experience with the population, pain-related beliefs, and care decisions. *Research in Developmental Disabilities, 62*, 197–208.

Genik, L. M., McMurtry, C. M., Breau, L. M., Lewis, S. P., & Freedman-Kalchman, T. (2017). Pain in children with developmental disabilities: Development and preliminary effectiveness of a pain training workshop for respite workers. *The Clinical Journal of Pain.* https://doi.org/10.1097/AJP.0000000000000554

Hadjistavropoulos, T., Craig, K. D., Duck, S., Cano, A., Goubert, L., Jackson, P. L., … Fitzgerald, T. D. (2011). A biopsychosocial formulation of pain communication. *Psychological Bulletin, 137*, 910–939. https://doi.org/10.1037/a0023876

Hagberg, B. (2002). Clinical manifestations and stages of Rett syndrome. *Mental Retardation and Developmental Disabilities Research Reviews, 8*, 61–65.

Haraldstad, K., Sørum, R., Eide, H., Natvig, G. K., & Helseth, S. (2011). Pain in children and adolescents: Prevalence, impact on daily life, and parents' perception, a school survey. *Scandinavian Journal of Caring Sciences, 25*(1), 27–36.

Heifetz, M., & Lunsky, Y. (2018). Implementation and evaluation of health passport communication tools in emergency departments. *Research in Developmental Disabilities, 72*, 23–32.

Hennequin, M., Morin, C., & Feine, J. S. (2000). Pain expression and stimulus localisation in individuals with Down's syndrome. *The Lancet, 356*(9245), 1882–1887.

Hester, N., Foster, R., & Kristensen, K. (1990). Measurement of pain in children: Generalizability and validity of the pain ladder and poker chip tool. *Advanced Pain Research and Therapy, 15*, 79–84.

Hicks, C. L., von Baeyer, C. L., Spafford, P. A., van Korlaar, I., & Goodenough, B. (2001). Faces Pain Scale-Revised: Toward a common metric in pediatric pain measurement. *Pain, 93*, 173–183. https://doi.org/10.1016/S0304-3959(01)00314-1

Hodgkinson, I., Jindrich, M. L., Duhaut, P., Vadot, J. P., Metton, G., & Berard, C. (2001). Hip pain in 234 non-ambulatory adolescents and young adults with cerebral palsy: A cross-sectional multicentre study. *Developmental Medicine and Child Neurology, 43*(12), 806–808.

Houlihan, C. M., O'Donnell, M., Conaway, M., & Stevenson, R. D. (2004). Bodily pain and health-related quality of life in children with cerebral palsy. *Developmental Medicine and Child Neurology, 46*(5), 305–310.

Hunt, A., Goldman, A., Seers, K., Crichton, N., Mastroyannopoulou, K., Moffat, V., … Brady, M. (2004). Clinical validation of the paediatric pain profile. *Developmental Medicine and Child Neurology, 46*(1), 9–18.

Hunt, A., Mastroyannopoulou, K., Goldman, A., & Seers, K. (2003). Not knowing—the problem of pain in children with severe neurological impairment. *International Journal of Nursing Studies, 40*(2), 171–183.

Hunt, K. A., & Franck, L. S. (2011). Special needs require special attention: A pilot project implementing the paediatric pain profile for children with profound neurological impairment in an in-patient setting following surgery. *Journal of Child Health Care, 15*(3), 210–220.

International Association for the Study of Pain (IASP). (2012). *IASP Taxonomy*. http://www.iasp-pain.org/Content/NavigationMenu/GeneralResourceLinks/PainDefinitions/default.htm

Johansson, M., Carlberg, E. B., & Jylli, L. (2010). Validity and reliability of a Swedish version of the non-communicating children's pain checklist–postoperative version. *Acta Paediatrica, 99*(6), 929–933.

Johannes, C. B., Le, T. K., Zhou, X., Johnston, J. A., & Dworkin, R. H. (2010). The prevalence of chronic pain in United States adults: Results of an Internet-based survey. *The Journal of Pain, 11*(11), 1230–1239.

Kennedy, S., O'Higgins, S., Sarma, K., Willig, C., & McGuire, B. E. (2014). Evaluation of a group based cognitive behavioural therapy programme for menstrual pain management in young women with intellectual disabilities: Protocol for a mixed methods controlled clinical trial. *BMC Women's Health, 14*(1), 107.

King, S., Chambers, C. T., Huguet, A., MacNevin, R. C., McGrath, P. J., Parker, L., & MacDonald, A. J. (2011). The epidemiology of chronic pain in children and adolescents revisited: A systematic review. *Pain, 152*(12), 2729–2738.

Kleinknecht, M. (2007). Reliability and validity of the German language version of the "NCCPC-R". *Pflege, 20*(2), 93–102.

Koh, J. L., Fanurik, D., Harrison, R. D., Schmitz, M. L., & Norvell, D. (2004). Analgesia following surgery in children with and without cognitive impairment. *Pain, 111*(3), 239–244.

Koller, D., & Goldman, R. D. (2012). Distraction techniques for children undergoing procedures: A critical review of pediatric research. *Journal of Pediatric Nursing: Nursing Care of Children and Families, 27*(6), 652–681.

Leadley, R. M., Armstrong, N., Lee, Y. C., Allen, A., & Kleijnen, J. (2012). Chronic diseases in the European Union: the prevalence and health cost implications of chronic pain. *Journal of Pain & Palliative Care Pharmacotherapy, 26*(4), 310–325.

Logan, D. E., Coakley, R. M., & Barber Garcia, B. N. (2014). Cognitive behavioural interventions. In P. J. McGrath, B. J. Stevens, S. M. Walker, & W. T. Zempsky (Eds.), *Oxford Textbook of Paediatric Pain* (pp. 519–530). Oxford: Oxford University Press.

Lotan, M., Ljunggren, E. A., Johnsen, T. B., Defrin, R., Pick, C. G., & Strand, L. I. (2009). A modified version of the Non-Communicating Children Pain Checklist-Revised, adapted to adults with intellectual and developmental disabilities: Sensitivity to pain and internal consistency. *The Journal of Pain, 10*(4), 398–407. https://doi.org/10.1016/j.jpain.2008.09.006

Lotan, M., Moe-Nilssen, R., Ljunggren, A. E., & Strand, L. I. (2009). Reliability of the non-communicating adult pain checklist (NCAPC), assessed by different groups of health workers. *Research in Developmental Disabilities, 30*(4), 735–745.

Lynch, M. E. (2011). The need for a Canadian pain strategy. *Pain Research and Management, 16*(2), 77–80.

Mackey, E., & Dodd, K. (2010). Evaluation and effectiveness of pain recognition and management training for staff working in learning disability services. *British Journal of Learning Disabilities, 39*(3), 243–251.

Malviya, S., Voepel-Lewis, T., Burke, C., Merkel, S., & Tait, A. R. (2006). The revised FLACC observational pain tool: Improved reliability and validity for pain assessment in children with cognitive impairment. *Pediatric Anesthesia, 16*(3), 258–265.

Malviya, S., Voepel-Lewis, T., Merkel, S., & Tait, A. R. (2005). Difficult pain assessment and lack of clinician knowledge are ongoing barriers to effective pain management in children with cognitive impairment. *Acute Pain, 7*(1), 27–32.

Malviya, S., Voepel-Lewis, T., Tait, A. R., Merkel, S., Lauer, A., Munro, H., & Farley, F. (2001). Pain management in children with and without cognitive impairment following spine fusion surgery. *Pediatric Anesthesia, 11*(4), 453–458.

McGrath, P. A. (1990). *Pain in children: Nature, assessment, and treatment*. New York: Guilford.

McGuire, B. E., Daly, P., & Smyth, F. (2010). Chronic pain in people with an intellectual disability: Under-recognised and under-treated? *Journal of Intellectual Disability Research, 54*(3), 240–245. https://doi.org/10.1111/j.1365-2788.2010.01254.x

McManus, S., Treacy, M., & McGuire, B. E. (2014). Cognitive behavioural therapy for chronic pain in people with an intellectual disability: A case series using components of the Feeling Better programme. *Journal of Intellectual Disability Research, 58*(3), 296–306.

Meir, L., Strand, L. I., & Alice, K. (2012). A model for pain behavior in individuals with intellectual and developmental disabilities. *Research in Developmental Disabilities, 33*(6), 1984–1989.

Merskey, H., & Bogduk, N. (1994). *Classification of chronic pain: Descriptions of chronic pain syndromes and definitions of pain terms*. Seattle, WA: IASP Press.

Nader, R., Oberlander, T. F., Chambers, C. T., & Craig, K. D. (2004). Expression of pain in children with autism. *The Clinical Journal of Pain, 20*(2), 88–97.

Nagasako, E. M., Oaklander, A. L., & Dworkin, R. H. (2003). Congenital insensitivity to pain: An update. *Pain, 101*(3), 213–219. https://doi.org/10.1016/S0304-3959(02)00482-7

Nahin, R. L. (2015). Estimates of pain prevalence and severity in adults: United States, 2012. *The Journal of Pain, 16*(8), 769–780.

Neul, J. L., Kaufmann, W. E., Glaze, D. G., Christodoulou, J., Clarke, A. J., Bahi-Buisson, N., … Percy, A. K. (2010). Rett syndrome: Revised diagnostic criteria and nomenclature. *Annals of Neurology, 68*(6), 944–950.

Nilsson, S., Johnson, E., & Adolfsson, M. (2016). Professionals' perceptions about the need for pain management interventions for children with cerebral palsy in South African school settings. *Pain Management Nursing, 17*(4), 249–261.

Oberlander, T. F., O'Donnell, M. E., & Montgomery, C. J. (1999). Pain in children with significant neurological impairment. *Journal of Developmental and Behavioral Pediatrics: JDBP, 20*(4), 235–243.

Palermo, T. M. (2000). Impact of recurrent and chronic pain on child and family daily functioning: A critical review of the literature. *Journal of Developmental and Behavioral Pediatrics, 21*(1), 58–69.

Palermo, T. M., Eccleston, C., Lewandowski, A. S., Williams, A. C. D. C., & Morley, S. (2010).

Randomized controlled trials of psychological therapies for management of chronic pain in children and adolescents: An updated meta-analytic review. *Pain, 148*(3), 387–397.

Pedersen, L. K., Rahbek, O., Nikolajsen, L., & Møller-Madsen, B. (2015). The revised FLACC score: Reliability and validation for pain assessment in children with cerebral palsy. *Scandinavian Journal of Pain, 9*, 57–61.

Perquin, C. W., Hazebroek-Kampschreur, A. A., Hunfeld, J. A., Bohnen, A. M., van Suijlekom-Smit, L. W., Passchier, J., & van der Wouden, J. C. (2000). Pain in children and adolescents: A common experience. *Pain, 87*(1), 51–58.

Phan, A., Edwards, C. L., & Robinson, E. L. (2005). The assessment of pain and discomfort in individuals with mental retardation. *Research in Developmental Disabilities, 26*, 433–439.

Quinn, B. L. (2016). 'It's Not Easy': School nurse pain assessment practices for students with special needs. *Pain Management Nursing, 17*(2), 119–131.

Quinn, B. L., Seibold, E., & Hayman, L. (2015). Pain assessment in children with special needs: A review of the literature. *Exceptional Children, 82*(1), 44–57.

Quinn, B. L., & Serna, R. W. (2017). Educators' experiences identifying pain among students in special education settings. *The Journal of School Nursing*. https://doi.org/10.1177/1059840517747974

Quinn, B. L., & Smolinski, M. (2017). Improving school nurse pain assessment practices for students with intellectual disability. *The Journal of School Nursing*. https://doi.org/10.1177/1059840517722591

Rakel, B., & Barr, J. O. (2003). Physical modalities in chronic pain management. *Nursing Clinics of North America, 38*, 477–494.

Regnard, C., Reynolds, J., Watson, B., Matthews, D., Gibson, L., & Clarke, C. (2007). Understanding distress in people with severe communication difficulties: Developing and assessing the Disability Distress Assessment Tool (DisDAT). *Journal of Intellectual Disability Research, 51*(4), 277–292.

Riddell, R. P., Taddio, A., McMurtry, C. M., Shah, V., Noel, M., & Chambers, C. T. (2015). Process interventions for vaccine injections: Systematic review of randomized controlled trials and quasi-randomized controlled trials. *The Clinical Journal of Pain, 31*(Suppl 10), S99.

Schiavenato, M., & Craig, K. D. (2010). Pain assessment as a social transaction: Beyond the "gold standard". *The Clinical Journal of Pain, 26*(8), 667–676.

Schopflocher, D., Taenzer, P., & Jovey, R. (2011). The prevalence of chronic pain in Canada. *Pain Research and Management, 16*(6), 445–450.

Scott, J., & Huskisson, E. C. (1976). Graphic representation of pain. *Pain, 2*(2), 175–184.

Sessle, B. J. (2012). The pain crisis: what it is and what can be done. *Pain Research and Treatment, 2012*, 1–6. https://doi.org/10.1155/2012/703947

Shinde, S. K., & Symons, F. (2007). Educator perceptions and ratings of pain in school-age children with mental retardation and developmental disabilities. *Education and Training in Developmental Disabilities*, 224–229.

Sinatra, R. (2010). Causes and consequences of inadequate management of acute pain. *Pain Medicine, 11*(12), 1859–1871.

Slifer, K. J., Hankinson, J. C., Zettler, M. A., Frutchey, R. A., Hendricks, M. C., Ward, C. M., & Reesman, J. (2011). Distraction, exposure therapy, counterconditioning, and topical anesthetic for acute pain management during needle sticks in children with intellectual and developmental disabilities. *Clinical Pediatrics, 50*(8), 688–697.

Sobsey, D. (2006). Pain and disability in an ethical and social context. In F. Srymons & T. Oberlander (Eds.), *Pain in Children and Adults with Developmental Disabilities* (pp. 19–39). Baltimore: Paul H. Brookes.

Solodiuk, J. C., Scott-Sutherland, J., Meyers, M., Myette, B., Shusterman, C., Karian, V. E., … Curley, M. A. (2010). Validation of the Individualized Numeric Rating Scale (INRS): A pain assessment tool for nonverbal children with intellectual disability. *Pain, 150*(2), 231–236.

Solodiuk, J., & Curley, M. A. (2003). Pain assessment in nonverbal children with severe cognitive impairments: The individualized numeric rating scale (INRS). *Journal of Pediatric Nursing, 18*(4), 295–299.

Stallard, P., Williams, L., Lenton, S., & Velleman, R. (2001). Pain in cognitively impaired, non- communicating children. *Archives of Disease in Childhood, 85*(6), 460–462.

Stallard, P., Williams, L., Velleman, R., Lenton, S., McGrath, P. J., & Taylor, G. (2002). The development and evaluation of the pain indicator for communicatively impaired children (PICIC). *Pain, 98*(1–2), 145–149.

Steingrímsdóttir, Ó. A., Landmark, T., Macfarlane, G. J., & Nielsen, C. S. (2017). Defining chronic pain in epidemiological studies: A systematic review and meta-analysis. *Pain, 158*(11), 2092–2107.

Stevens, B. J., Abbott, L. K., Yamada, J., Harrison, D., Stinson, J., Taddio, A., … Campbell, F. (2011). Epidemiology and management of painful procedures in children in Canadian hospitals. *Canadian Medical Association Journal, 183*(7), E403–E410.

Stevens, B. J., & Zempsky, W. T. (2013). Prevalence and distribution of pain in children. In P. J. McGrath, B. J. Stevens, S. M. Walker, & W. T. Zempsky (Eds.), *Oxford Textbook of Paediatric Pain* (pp. 12–19). Oxford: Oxford University Press.

Symons, F. J., Byiers, B., Tervo, R., & Beisang, A. (2013). Parent reported pain in rett syndrome. *The Clinical Journal of Pain, 29*(8), 744–746.

Symons, F. J., Harper, V. N., McGrath, P. J., Breau, L. M., & Bodfish, J. W. (2009). Evidence of increased nonverbal behavioral signs of pain in adults with neurodevelopmental disorders and chronic self-injury. *Research in Developmental Disabilities, 30*(3), 521–528.

Symons, F. J., Shinde, S. K., & Gilles, E. (2008). Perspectives on pain and intellectual disability. *Journal of Intellectual Disability Research, 52*(4), 275–286.

Taddio, A., McMurtry, C. M., Shah, V., Riddell, R. P., Chambers, C. T., Noel, M., … Bleeker, E. V. (2015). Reducing pain during vaccine injections: Clinical practice guideline. *Canadian Medical Association Journal, 187*, 975–982. https://doi.org/10.1503/cmaj.150391

Taddio, A., & Oberlander, T. F. (2006). Pharmacological management of pain in children and youth with significant neurological impairments. In *Pain in Children and Adults with Developmental Disabilities* (pp. 193–211). Baltimore, MD: Paul H. Brookes Publishing Co.

Temple, B., Dubé, C., McMillan, D., Secco, L., Kepron, E., Dittberner, K., & Vipond, G. (2012). Pain in people with developmental disabilities: A scoping review. *Journal on Developmental Disabilities, 18*, 73–86.

Terstegen, C., Koot, H. M., de Boer, J. B., & Tibboel, D. (2003). Measuring pain in children with cognitive impairment: Pain response to surgical procedures. *Pain, 103*(1–2), 187–198.

Tudor, M. E., Walsh, C. E., Mulder, E. C., & Lerner, M. D. (2015). Pain as a predictor of sleep problems in youth with autism spectrum disorders. *Autism, 19*(3), 292–300.

Tupper, S. M., Swiggum, M. S., O'Rourke, D., & Sangster, M. L. (2013). Physical therapy interventions for pain in childhood and adolescence. In P. J. McGrath, B. J. Stevens, S. M. Walker, & W. T. Zempsky (Eds.), *Oxford Textbook of Paediatric Pain* (pp. 581–589). Oxford: Oxford University Press.

Turk, D. C. (2003). Cognitive-behavioral approach to the treatment of chronic pain patients. *Regional Anesthesia and Pain Medicine, 28*(6), 573–579.

Twycross, A., Voepel-Lewis, T., Vincent, C., Franck, L. S., & Von Baeyer, C. L. (2015). A debate on the proposition that self-report is the gold standard in assessment of pediatric pain intensity. *The Clinical Journal of Pain, 31*(8), 707–712.

Uman, L. S., Birnie, K. A., Noel, M., Parker, J. A., Chambers, C. T., McGrath, P. J., & Kisely, S. R. (2013). Psychological interventions for needle-related procedural pain and distress in children and adolescents.. *The Cochrane Library*.

Valkenburg, A. J., Tibboel, D., & Dijk, M. (2015). Pain sensitivity of children with Down syndrome and their siblings: Quantitative sensory testing versus parental reports. *Developmental Medicine & Child Neurology, 57*(11), 1049–1055. https://doi.org/10.1111/dmcn.12823

Valkenburg, A. J., van Dijk, M., de Klein, A., van den Anker, J. N., & Tibboel, D. (2010). Pain management in intellectually disabled children: Assessment, treatment, and translational research. *Developmental Disabilities Research Reviews, 16*(3), 248–257.

Valrie, C. R., Bromberg, M. H., Palermo, T., & Schanberg, L. E. (2013). A systematic review of sleep in pediatric pain populations. *Journal of Developmental and Behavioral Pediatrics, 34*(2), 120–128.

van der Putten, A., & Vlaskamp, C. (2011). Pain assessment in people with profound intellectual and multiple disabilities; A pilot study into the use of the Pain Behaviour Checklist in everyday practice. *Research in Developmental Disabilities, 32*(5), 1677–1684.

von Baeyer, C. L., Chambers, C. T., Forsyth, S. J., Eisen, S., & Parker, J. A. (2013). Developmental data supporting simplification of self-report pain scales for preschool-age children. *The Journal of Pain, 14*(10), 1116–1121.

Walsh, M., Morrison, T. G., & McGuire, B. E. (2011). Chronic pain in adults with an intellectual disability: prevalence, impact, and health service use based on caregiver report. *Pain, 152*(9), 1951–1957. https://doi.org/10.1016/j.pain.2011.02.031

Weissman-Fogel, I., Roth, A., Natan-Raav, K., & Lotan, M. (2015). Pain experience of adults with intellectual disabilities–caregiver reports. *Journal of Intellectual Disability Research, 59*(10), 914–924.

Williams, A. C. D. C., Eccleston, C., & Morley, S. (2012). Psychological therapies for the management of chronic pain (excluding headache) in adults. *Cochrane Database of Systematic Reviews, 11*, 1–108. https://doi.org/10.1002/14651858.CD007407.pub3

Wong, D. L., & Baker, C. M. (1988). Pain in children: Comparison of assessment scales. *Pediatric Nursing, 14*(1), 9–17.Zabalia, M., Breau, L. M., Wood, C., Lévêque, C., Hennequin, M., Villeneuve, E., … Breau, G. (2011). Validation of the French version of the non-communicating children's pain checklist-postoperative version. *Canadian Journal of Anaesthesia= Journal canadien d'anesthesie, 58*(11), 1016–1023.

54

Nutrition

Regina M. Subach and Lori Klein

> **Food can be the greatest form of medicine, or the slowest form of poison.—Ann Wigmore**

Despite the ancient wisdom that the food we eat determines the robustness of our health, we continue to struggle with adequate nutrition at a global level. When humans eat healthful, fresh, whole foods in the amounts needed to maintain a healthy body weight, the potential result is a vibrant, longer life with lower risk of chronic diseases. The number of people classified as overweight or obese remains disastrously high across the population, and individuals with intellectual disabilities (ID or IDD) suffer from overweight and obesity at equal or greater rates in comparison (Ptomey, Goetz, Lee, Donnelly, & Sullivan, 2013). An unhealthy body weight is one of the strongest risk factors for the development of other chronic diseases including diabetes and cardiovascular disease.

There is limited research on the prevalence of overweight/obesity, chronic disease, and nutritional risk for individuals with ID, but the available information paints a gloomy picture for these vulnerable citizens (Ptomey et al., 2013). Current data indicates obesity rates double that of the general population which translates into increased chronic disease risk (Ptomey & Wittenbrook, 2015).

The Academy of Nutrition and Dietetics (AND) is the leading authority on evidence-based nutrition information and publishes “Position Papers” on critical areas of nutrition research with guidelines for special populations. These articles summarize nutrition policy and guide the 100,000 credentialed nutrition professionals who are members of AND. The 2016 Position Paper on Nutrition Services for Individuals with Intellectual and Developmental Disabilities and Special Health Care Needs details a multitude of nutritional risk factors for this underserved group, including altered growth patterns, difficulty with self-feeding, metabolic disturbances, and medication interactions (Ptomey & Wittenbrook, 2015). As noted previously, individuals with ID also have a higher risk of developing comorbid conditions such as obesity and metabolic syndrome that further heighten the need for programs that address nutritional needs (Ptomey et al., 2013). Intellectual disabilities complicate an already challenging problem with additional barriers related to nutrition knowledge, limited resources, and perceptual differences.

This chapter discusses the Dietary Guidelines for Americans, the additional barriers faced by individuals with ID in meeting these recommendations, and practical applications to improve the

---

R. M. Subach (✉) · L. Klein
West Chester University, West Chester, PA, USA
e-mail: rsubach@wcupa.edu; lklein@wcupa.edu

J. L. Matson (ed.), *Handbook of Intellectual Disabilities*, Autism and Child Psychopathology Series, https://doi.org/10.1007/978-3-030-20843-1_54

nutrition status of this population. Specific disabilities and case studies are included to demonstrate nutrition risk and interventions.

Nutrition Basics

Before reviewing the Dietary Guidelines for Americans, it can be helpful to understand basic nutrition terms and the components of the foods we eat and how they support overall health. Nutrition has been defined as the "science of food, the nutrients and other substances therein, their action, interaction and balance in relation to health and disease, and the process by which the organism ingests, absorbs, transports, utilizes and excretes food substances" (Lagua & Claudio, 1995). For a more simplified definition, nutrition is the process of consuming food to meet the dietary needs of the human body.

The food we eat every day comprises macronutrients (protein, carbohydrates, and fat), micronutrients (vitamins and minerals), and dietary fiber. Macronutrients provide energy needed for physical activity, growth, and structure for basic body functions. Micronutrients have important roles aiding in immune function, neurological function, cell growth, and cell repair.

Macronutrients

Carbohydrate broken down into glucose is the main energy nutrient and preferred source of energy for the brain and nervous system. Dietary recommendations for carbohydrates come primarily from plant foods: cereals, grains, fruits, vegetables, as well as dairy products. Whole grains are the preferred choice over processed grains due to their higher fiber and nutrient content. Consuming plant sources is an added bonus because they also contribute as the primary source for dietary fiber.

Dietary fibers are the nondigestible part of plants is found in all foods derived from plants. Whole grains and non-processed plant foods provide good amounts of dietary fiber. Dietary fiber found in legumes, oat, barley, and citrus fruits are classified as soluble fibers. Soluble fibers form gels when dissolved in water and are digested by bacteria in the intestinal tract. They reduce the risk of cardiovascular disease and diabetes by lowering blood cholesterol and stabilizing blood sugars. Dietary fiber found in whole grains and vegetables and some fruits such as apples, raspberries, and strawberries are classified as insoluble fiber. This type of fiber aids in elimination and helps reduce constipation. Fiber recommendations are 25 g for women and 38 g per day for men. The average fiber intake of Americans is ~15 g. Following recommendations of MyPlate and Dietary Guidelines for Americans will aid in meeting daily fiber recommendations.

Proteins are amino acids which make up most tissues in the major part of the human body. Amino acids have both functional and structural roles which aid in the building and repairing muscle, bone, and skin; act as enzymes for metabolic reactions; are hormones, regulate fluid balance; act as antibodies; and aid in glucose production when needed. Dietary recommendations for protein include both animal- and plant-based protein sources. Animal proteins include meat, poultry, fish, eggs, and dairy products. Plant-based proteins are found in beans, legumes, nuts, soy, vegetables, and grains.

Fat is the most concentrated source of energy. It serves as the structure and insulation of cell membranes and also aids in the absorption of fat-soluble vitamins A, D, E, and K. Dietary intake of fat comes from both animal and plant sources. Animal sources of fat from meat, eggs, poultry skin, and dairy contribute the highest amount of saturated fat and cholesterol in the diet. Saturated fat plays a role in increasing blood cholesterol and should be limited to no more than 10% of calorie intake. The most recent DGA has no specific recommendation for cholesterol intake, but adherence to a diet that limits saturated fat to less than 10% will limit cholesterol intake to 100–300 mg across the calories levels (U.S. Dept., 2015). Plant foods, poultry, and fish provide polyunsaturated and monounsaturated fatty acids to the diet. These types of fatty acids aid in heart health through maintenance of healthy blood cholesterol level.

Micronutrients

Vitamins and minerals are essential nutrients needed in small amounts to prevent deficiency and promote health. They are classified as "micro" or "transfer nutrients." They aid in the transformation of energy nutrients into fuel and are critically important for supporting heart function, immune response, cell repair, and numerous other vital functions. Food selectivity, limited intake of fruits and vegetables, and omission of foods or entire food groups puts individuals at risk for micronutrient deficiencies. This reduced diet quality has been seen to lead to deficiencies of vitamin A, vitamin D, vitamin E, vitamin K, folate, potassium, fiber, and calcium among a group of persons with intellectual disabilities (Ptomey et al., 2013). The Academy of Nutrition and Dietetics also notes several vitamins and minerals of concern for this group overall as well as for specific disabilities that occur in conjunction with intellectual disabilities. See Table 54.1.

Nutrient deficiencies may be more likely to go undetected in individuals with ID, because of difficulty communicating fatigue, pain, and illness. If there are not enough food sources of vitamin A in the diet, the end result can be a permanent visual impairment. Vitamin D insufficiency can occur when there is not enough exposure to sunlight and a lack of fortified foods, and this can impair bone density and lead to osteoporosis. Vitamin E deficiency is rarely seen because it is prominent in oils (as well as nuts and seeds) but can occur with metabolic disorders related to fat absorption, and could lead to neurological disturbances, muscle fatigue, and visual impairment. Vitamin K from leafy greens is important for proper blood clotting, and a dietary deficiency can result in bruising and uncontrolled bleeding. Folate is responsible for proper nervous system function. Many subtle signs can signal inadequate intake, which could eventually lead to anemia in adults; children may suffer additional neurological effects including seizures. Calcium works in conjunction with vitamin D for bone health and deficiency symptoms are well documented through osteoporosis. On a larger scale the imbalance of higher sodium intake with lower potassium intake is a contributor to high blood pressure and increased risk of cardiovascular disease.

Vitamin D and vitamin B-12 are two critical micronutrients for this population due to potential food limitations, malabsorption conditions, and issues that limit sun exposure.

Vitamin D

Vitamin D is a fat-soluble vitamin that is also classified as a hormone that can be produced endogenously by the body through exposure to ultraviolet rays from the sun. With the exception of cod liver oil and fatty fishes vitamin D is not found abundantly in foods. Milk and dairy products are often fortified with vitamin D, serving as the most common dietary source of vitamin D; beef liver, egg yolks, and cheese contain smaller amounts. Vitamin D from sun exposure and food is biologically inert and must be activated in the body.

Vitamin D is associated primarily with bone health in conjunction with calcium and phosphorus, both necessary for adequate bone mineralization. In addition to its role in bone health vitamin D aids in cell growth, neuromuscular and immune functions, and reduction of inflammation (NIH, 2010). Research continues on other possible health effects of vitamin D, including reduced risk of chronic disease and depression.

The Recommended Dietary Allowances (RDA) for vitamin D vary based on age and stages of the life cycle ranging from 400 to 600 International units (IU). Average American Intake of vitamin D solely from foods has been estimated to be lower than the RDA. More research must be conducted on vitamin D supplementation (Tripkovic et al., 2012).

Persons at risk for vitamin D deficiencies include breastfed infants, the elderly, persons with dark skin, and individuals with limited sun exposure, inflammatory bowel disease, malabsorption, and/or who have undergone gastric bypass surgery. Individuals with disabilities,

Table 54.1 Key vitamins and minerals of concern for individuals with intellectual disabilities

Vitamin/mineral	Primary function	Food sources	Deficiency signs and symptoms	At-risk groups
Vitamin A	Vision	Green leafy, orange, and yellow vegetables, cantaloupe, and dairy	Vision impairment or loss	Restricted diets that avoid fruits, vegetables, and dairy
Vitamin D ∗produced in the skin with sunlight exposure	Bone Strength	Fatty fish such as salmon, tuna, mackerel and fortified foods milk	Rickets Osteomalacia	Restricted diets that avoid dairy Residence in inpatient facilities, low mobility, reduced times outdoors
Vitamin E	Nerve Function	Vegetable oils, nuts, seeds, green vegetables such as spinach and broccoli	Anemia Neurological impairment Seizures	Individuals with disabilities have lower intake, but clinical deficiency is rare
Vitamin K	Blood Clotting	Leafy green vegetables, broccoli, and canola oils	Bruising Excessive bleeding	Individuals with disabilities have lower intake, but clinical deficiency is rare
Vitamin B-6	Protein metabolism	Fish, beans, beef, poultry, Fortified cereals/foods, potatoes, starchy vegetables	Microcytic anemia Dermatitis with scaling near mouth and swollen tongue Depression	Restricted diets Malabsorption disorders (celiac, Crohn's)
Vitamin B-12	Nerve Function Red Blood Cell Formation DNA synthesis	Fish, meat, poultry, eggs, milk, and other dairy products Fortified foods	Megaloblastic Anemia Neuropathy Depression/ Confusion	Elderly Vegetarians/Vegans Malabsorption disorders (celiac, Crohn's) Resection of stomach/ small intestine
Folate	Neural tube formation Nerve function Blood Cells	Leafy greens, citrus, beans and legumes Fortified foods	Neural tube defects in fetus Weakness/fatigue Headache Behavior disorders Megaloblastic anemia	Medication interference Malabsorption disorders (celiac, Crohn's) Inadequate intake of food sources
Calcium	Bone Strength	Milk, cheese, and yogurt; vegetables like kale, broccoli, cabbage, and canned sardines and salmon with soft bones	Limited stature in children Osteopenia Osteoporosis	Medication interference Inadequate intake of food sources Lactose intolerance Thyroid disorders
Potassium	Electrolyte balance Blood pressure regulation Muscle and nerve function	A variety of leafy greens and root vegetables, fruits like grapes, apricots, and citrus.	Weakness/fatigue Gastrointestinal upset High blood pressure Heart palpitations	Inadequate intake of food sources

especially those residing in group home or disability rehabilitation centers, have higher incidences of vitamin D deficiencies (Wimalawansa, 2016). Diets of individuals who are lactose intolerant have a milk allergy or follow an ovo-vegetarian (a vegetarian diet that excludes dairy products but includes eggs) or vegan diet are often vitamin D deficient (IOM, 2010). Food selectivity and "fad diets" that limit or eliminate vitamin D fortified foods increase the risk of insufficient intake. Vitamin D deficiency results in rickets in children and osteomalacia in adults, and these conditions are characterized by softening of the bones and may lead to an inability to bear weight on the legs.

Individuals with medically complex developmental disabilities (MCDD) may have an even higher risk of vitamin D deficiency due to use of medications such as steroids, phenobarbital, and phenytoin known to interfere with vitamin D metabolism (NIH, 2010; Wimalawansa, 2016). Increased exposure to sunlight, ultraviolet therapy, and/or supplementation may be necessary for at-risk individuals (Wimalawansa, 2016). Supplementation should be done under the supervision of a professional.

Vitamin B-12

Vitamin B-12 is a water-soluble vitamin that is responsible for red blood cell maturation, nerve function, and DNA synthesis (ODS, 2018). This vitamin is prevalent in animal products including beef, fish, poultry, milk, and eggs. Additionally, many plant-based foods such as cereal and tofu are fortified with vitamin B-12 (ODS, 2018). The recommended intake varies throughout the life span ranging from 0.4–0.5 micrograms (mcg) in infancy, 0.9–1.2 mcg during childhood, 1.8 mcg adolescence, to 2.4 mcg in adults (pregnancy-2.6 mcg, lactation-2.8 mcg) (ODS, 2018). For comparison, fortified cereal contains 6.0 mcg per serving of active B-12 that is readily absorbed. A serving of beef contains 1.4 mcg of vitamin B-12 (ODS, 2018).

Although vitamin B-12 deficiency is generally uncommon due to its abundance in animal products, there are some groups known to be at higher risk (ODS, 2018). Regular consumption of fortified foods or oral supplementation is highly recommended for individuals who follow a vegetarian or vegan diet and therefore do not consume animal products, especially for women who are pregnant or breastfeeding to ensure adequacy for the fetus/infant (ODS, 2018). Hydrochloric acid and intrinsic factor present in the stomach are both needed for the breakdown and intestinal absorption of vitamin B-12 and factors that can impair these processes include the aging process with decreased HCL production and increased rates of pernicious anemia, the use of medications for acid reflux, malabsorption disorders like Crohn's disease and celiac disease, and surgical resection of the stomach and/or small intestine (ODS, 2018). Individuals in these high-risk groups should have regular blood samples checked for B-12 adequacy. A medical professional must recommend appropriate supplementation in these cases, because intramuscular injection may be necessary when processing and absorption are impaired or when the deficiency is severe (ODS, 2018).

When vitamin B-12 deficiency does occur, it is critical to address to avoid permanent damage. Three separate physiological symptom categories may be present when there is a lack of B-12—hematological, psychiatric, and neurological—and this spectrum of symptoms can lead to misdiagnosis (ODS, 2018). Megaloblastic anemia presents with large, immature red blood cells that may cause fatigue and weakness. Neuropathy, tingling in hands and feet, and loss of balance are signs and symptoms of neurological impacts that can be permanent and may occur without anemia. Confusion, depression, and memory loss are psychiatric components of vitamin B-12 deficiency that could be mistaken for Alzheimer's disease or dementia. It is also important to note that when folic acid intake is excessive, particularly through supplementation, it can hide the B-12 deficiency and increase the chances of permanent nerve damage (ODS, 2018). Routine screening should be done for high-risk groups, and blood work is warranted for anyone who exhibits potential signs of this deficiency.

The Dietary Guidelines for Americans

The current dietary guidelines for 2015–2020 focus on 5 overarching guidelines designed for disease prevention and health promotion. This section serves as an outline of each guideline followed by barriers and "practical strategies" for improving nutritional health of persons with ID.

The Dietary Guidelines for Americans (DGA), released every 5 years since their introduction in 1980, provide evidence-based food and beverage recommendations for Americans ages 2 and older. The DGA serve as a resource for policy makers, public health agencies, educational institutions, and health professionals in the development and implementation of nutrition policies and programs designed to improve the overall health of all Americans. They are the basis for federal nutrition programs like The Federal School Lunch Program and Child and Adult Care Feeding program. The DGA 2015–2020 are based on the latest scientific evidence that reinforces the innate ancient wisdom connecting nourishing food intake and positive health outcomes.

Guideline One: Follow a Healthy Eating Pattern Across the Life Span

> *All food and beverage choices consumed matter. Choose a healthy eating pattern at an appropriate calorie level to help achieve and maintain a healthy body weight, support nutrient adequacy, and reduce the risk of chronic disease*—(U.S. Dept., 2015).

The typical eating pattern of many Americans does not align with the recommendations of the DGA. The primary emphasis is on "Healthy Eating Patterns as a Whole" rather than attention to specific nutrients like calcium, vitamin D, saturated fats, or "superfoods" such as nuts, berries, or dairy to improve overall heath. An eating pattern, often referred to as a "diet," is defined as a combination of foods and or beverages that constitute an individual's complex intake of food over time (U.S. Dept., 2015). There is a strong emphasis on the synergistic effect of food and beverage on health and wellness.

Healthy eating patterns have been associated with positive health outcomes. There is strong evidence between healthy patterns and reduced risk of cardiovascular disease. Moderate evidence shows an association between healthy eating patterns and reduced risks of type 2 diabetes and certain types of cancer, overweight, and obesity (U.S. Dept., 2015**).** Emerging research is indicating a relationship between eating patterns and neurocognitive disorders and congenital abnormalities (U.S. Dept., 2015).

Healthy eating patterns are adaptable and include a variety of food sources including proteins from plant and/or animal sources, whole fruits, whole grains, dairy, vegetables, and oils. Half of all grains consumed should be "whole grain" such as brown rice, oats, and popcorn; rather than processed grains in white rice, white bread, and sugary baked goods. Vegetable intake should include subgroups of dark green, red and orange, beans and peas, starchy and other vegetables. Dairy should be from low-fat sources. Fat should come in the form of healthy oils such as olive or canola instead of solid fats. Healthy eating patterns limit added sugars, saturated fats, trans fats, and processed foods. Recommended eating patterns highlighted in the guidelines include the Healthy US Style meal pattern, the Mediterranean diet, the Dash Diet, and vegetarian and vegan diets. Links to additional information on these eating patterns can be found on the My Plate website.

MyPlate

The recommendations in the Dietary Guidelines for Americans are communicated to the public through the MyPlate message. MyPlate translates the macronutrients (carbohydrates, fat, and protein) into a more easily understood depiction of food groups and how foods fit into a healthy eating pattern. The image of MyPlate (Fig. 54.1)

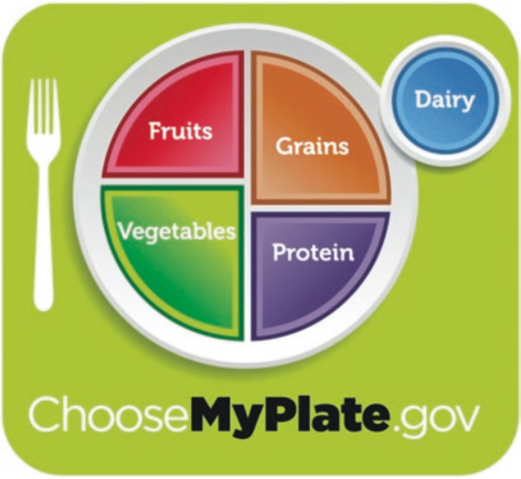


Fig. 54.1 MyPlate

was introduced to transform the previous Pyramid depiction of the Dietary Guidelines into a more concrete visual of how the food groups fit into a meal pattern. This symbol outlines a plate with 1/2 fruits and vegetables that conveys to support the “5 a day” motto for intake of these nutrient rich foods, and the other half of the plate is taken up by grains and a smaller protein portion. Dairy is highlighted on the side in the common cup form for serving milk products. There are many choices with additional recommendations within each group, but this simple image conveys the most important pieces of dietary advice for Americans by encouraging increased fruit and vegetable intake.

Guideline Two

Focus on Variety, Nutrient Density, and Amount

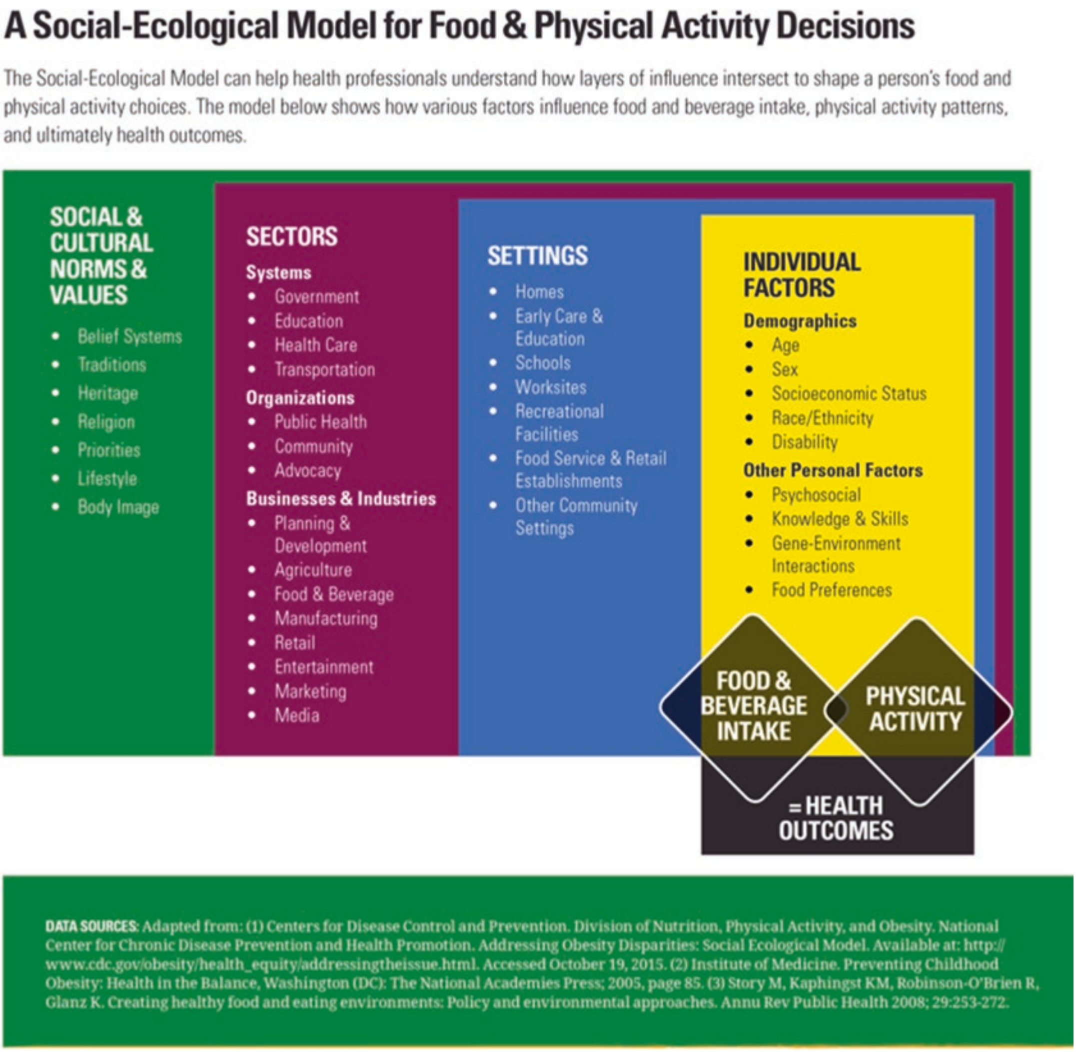
> *To meet nutrient needs within calorie limits, choose a variety of nutrient-dense foods across and within all food groups in recommended amounts.*—(U.S. Dept., 2015)

Most Americans do not follow the healthy eating pattern described in Guideline One as seen in lower intake in fruits, vegetables, and dairy categories and excess intake of sugars, sodium, and saturated fat as compared to the recommendations (U.S. Dept., 2015). In a dietary pattern where fruit and vegetable intake is low and refined grains are chosen over whole grains, the protective effects of fiber are reduced and subsequently overall diet quality is greatly diminished (McGill, Birkett, & Fulgonii, 2016). Guideline Two breaks down the Healthy Eating Pattern into more specific dietary advice within each food group and also addresses appropriate portion sizes. In addition to lower than recommended overall fruit and vegetable intake, the American diet low is specifically lacking in variety that provides the full array of micronutrients. Variety of intake refers to the selection of foods from an assortment of the food groups: fruits, vegetable, grains, proteins, and dairy and their subgroups.

Nutrient needs should be met primarily from food with a focus on nutrient-dense foods. Nutrient-dense foods are foods that are not diluted with calories from added fats, sugars, refined starches, or solid fats not naturally present in foods (U.S. Dept., 2015). Examples of nutrient-dense foods are fruits, vegetables, and whole grains that typically contain a variety of vitamins, minerals, phytonutrients, and fiber. A meal pattern that includes large amounts of juice drinks and processed grains is less nutrient dense than a diet that includes a variety of whole fruits and whole grains.

The majority of vegetables consumed by Americans are in the form of potatoes (French fries) and tomatoes (sauces and condiments), and the 2015 DGA now include recommendations for vegetable subgroups as a result (U.S. Dept., 2015). Recommended servings are based on a 2000 calorie diet (that will vary based on age, sex, and energy needs) as seen in the printable checklist, Fig. 54.2. The daily 2 1/2 cup-equivalent (c-eq) of vegetables in a 2000 calorie diet is broken down into subgroups for maximum nutrient density and fiber intake. Subgroups have weekly intake suggestions and include dark green (1 1/2 c-eq), red and orange (5 1/2 c-eq), legumes (beans and peas—1 1/2c-eq), starchy (5 c-eq), and other vegetables (4 c-eq) to supply maximum nutrient density with a wide array of vitamins, minerals, phytonutrients, and fiber (U.S. Dept., 2015). The fruit recommendation suggests the whole food form over fruit juices for improved fiber and potential for excess calories in juice. When juice is chosen within the fruit category, it should be 100% fruit juice (U.S. Dept., 2015).

Although over half of Americans meet the total protein and grain recommendations, they do not choose the healthful options recommended in these food subgroups (U.S. Dept., 2015). There has been a continuing shift in the DGA to include plant-based protein sources such as beans and legumes which also contain fiber and a multitude of vitamins and minerals. Animal protein sources are still favored in the typical American Diet but contain higher saturated fat and cholesterol without the beneficial fiber (U.S. Dept., 2015).



A Social-Ecological Model for Food & Physical Activity Decisions

The Social-Ecological Model can help health professionals understand how layers of influence intersect to shape a person's food and physical activity choices. The model below shows how various factors influence food and beverage intake, physical activity patterns, and ultimately health outcomes.

SOCIAL & CULTURAL NORMS & VALUES
- Belief Systems
- Traditions
- Heritage
- Religion
- Priorities
- Lifestyle
- Body Image

SECTORS

Systems
- Government
- Education
- Health Care
- Transportation

Organizations
- Public Health
- Community
- Advocacy

Businesses & Industries
- Planning & Development
- Agriculture
- Food & Beverage
- Manufacturing
- Retail
- Entertainment
- Marketing
- Media

SETTINGS
- Homes
- Early Care & Education
- Schools
- Worksites
- Recreational Facilities
- Food Service & Retail Establishments
- Other Community Settings

INDIVIDUAL FACTORS

Demographics
- Age
- Sex
- Socioeconomic Status
- Race/Ethnicity
- Disability

Other Personal Factors
- Psychosocial
- Knowledge & Skills
- Gene-Environment Interactions
- Food Preferences

FOOD & BEVERAGE INTAKE

PHYSICAL ACTIVITY

= HEALTH OUTCOMES

DATA SOURCES: Adapted from: (1) Centers for Disease Control and Prevention. Division of Nutrition, Physical Activity, and Obesity. National Center for Chronic Disease Prevention and Health Promotion. Addressing Obesity Disparities: Social Ecological Model. Available at: http://www.cdc.gov/obesity/health_equity/addressingtheissue.html. Accessed October 19, 2015. (2) Institute of Medicine. Preventing Childhood Obesity: Health in the Balance, Washington (DC): The National Academies Press; 2005, page 85. (3) Story M, Kaphingst KM, Robinson-O'Brien R, Glanz K. Creating healthy food and eating environments: Policy and environmental approaches. Annu Rev Public Health 2008; 29:253-272.

Fig. 54.2 The social ecological model (U.S. Dept., 2015)

Grains may be avoided because of the high-carbohydrate content in the refined, processed grains that are heavily consumed, and the DGA recommend that at least half of grain intake come from whole grains for increased dietary fiber and key nutrients including zinc, iron, folate, magnesium, and a variety of B vitamins (U. S. Dept., 2015). The nutrient content varies between grains and many refined grains have nutrients added back during processing. Whole grains contain the entire kernel and are single foods like oats, popcorn, and rice, but also present in breads, crackers, and other products that are labeled as 100% whole grain.

Although there is little research comparing eating patterns for individuals with intellectual disabilities to the DGA recommendations, a 2013 study compared the diet quality of the general population to a group of 70 overweight/obese adults with IDD (Ptomey et al., 2013). This comparison showed lower overall diet quality and even lower intake of fruits, whole grains, vegetables (dark green and orange) with higher amounts of dietary oils and sodium (Ptomey et al., 2013). Additionally, many nutrient deficiencies were noted in vitamin A, vitamin D, vitamin E, vitamin K, folate, potassium, fiber, and calcium (Ptomey et al., 2013).

USDA
United States Department of Agriculture

MyPlate Daily Checklist

Find your Healthy Eating Style

Everything you eat and drink matters. Find your healthy eating style that reflects your preferences, culture, traditions, and budget—and maintain it for a lifetime! The right mix can help you be healthier now and into the future. The key is choosing a variety of foods and beverages from each food group—*and making sure that each choice is limited in saturated fat, sodium, and added sugars.* Start with small changes—**"MyWins"**—to make healthier choices you can enjoy.

Food Group Amounts for 2,000 Calories a Day

Fruits	Vegetables	Grains	Protein	Dairy
2 cups	**2 1/2 cups**	**6 ounces**	**5 1/2 ounces**	**3 cups**
Focus on whole fruits	Vary your veggies	Make half your grains whole grains	Vary your protein routine	Move to low-fat or fat-free milk or yogurt
Focus on whole fruits that are fresh, frozen, canned, or dried.	Choose a variety of colorful fresh, frozen, and canned vegetables—make sure to include dark green, red, and orange choices.	Find whole-grain foods by reading the Nutrition Facts label and ingredients list.	Mix up your protein foods to include seafood, beans and peas, unsalted nuts and seeds, soy products, eggs, and lean meats and poultry.	Choose fat-free milk, yogurt, and soy beverages (soy milk) to cut back on your saturated fat.

Limit

Drink and eat less sodium, saturated fat, and added sugars. Limit:
- Sodium to **2,300 milligrams** a day.
- Saturated fat to **22 grams** a day.
- Added sugars to **50 grams** a day.

Be active your way: Children 6 to 17 years old should move **60 minutes** every day. Adults should be physically active at least **2 1/2 hours** per week.

Use SuperTracker to create a personal plan based on your age, sex, height, weight, and physical activity level.

SuperTracker.usda.gov

Guideline Three: Limit Calories from Added Sugars and Saturated Fats and Reduce Sodium Intake

> *Consume an eating pattern low in added sugars, saturated fats, and sodium. Cut back on foods and beverages higher in these components to amounts that fit within healthy eating patterns.*—(U.S. Dept., 2015)

While Guideline Two focuses on the nutrients that Americans need to increase and improve, Guideline Three clarifies the need to limit foods containing harmful components that are in excess in the diet. Recommendations suggest limiting both added sugars and saturated fat to less than 10% of daily calorie and limiting sodium intake to less than 2300 mg/day.

The consumption of sugar-sweetened beverages (SSBs) is the leading source of added sugar in the American diet (Park et al., 2016). Beverages classified as SSB include, but are not limited to, fruit drinks and beverages (100% fruit juice is excluded), regular soda, energy and sports drinks, sweetened flavored waters, and coffee and tea beverages with added sugars (Park et al., 2016).

Consumption of SSBs which provide little or no nutritional value is correlated to an increase in risks of overweight and obesity, cardiovascular disease, type 2 diabetes, kidney disease, and asthma (Rosinger, Herrick, Gahche, & Park, 2017).

Regular consumption of SSB by adults with disabilities contributes excess sugar and calories to the diet. Increased caloric intake can add to weight gain when physical activity is limited as is often the case in persons with disabilities. Excessive weight gain leads to an increase in the risk of chronic disease states such as diabetes and cardiovascular disease (Kim, Park, Carroll, & Okoro, 2017). Statistics comparing consumption of SSBs by persons with disabilities to the overall population show a slightly higher intake for persons with disabilities and a need for increased education and strategies to decrease this source of added sugars (Kim et al., 2017).

Other primary sources of added sugar in the American diet come from refined, processed treats like cookies, cakes, and ice cream (U. S. Dept., 2015). On a daily basis Americans consumed 270 calories (over 13%) of daily intake from added sugar (U. S. Dept., 2015). Another chunk of added calories that are disease promoting comes from saturated fat.

The largest contributor to saturated fat in the American diet comes from animal products that are part of menu items with cheese and meat including pizza, burgers, and tacos (U. S. Dept., 2015). Dessert items (that also contain high added sugars), full fat dairy products, and animal proteins account for most of the remaining saturated fat intake. Most Americans exceed the 10% of daily calories limit for saturated fat with average intake at 11% of caloric intake (U. S. Dept., 2015). Many of these foods also contain high levels of sodium which is another nutrient of concern.

Although sodium does not provide unnecessary calories, excessive intake can lead to hypertension and increased risk of cardiovascular disease. American sodium intake greatly exceeds the recommended maximum of 2300 mg/day (U. S. Dept., 2015). Mixed menu items containing meat and cheese (like the pizza, burgers, and tacos with high saturated fat) also represent the highest proportion of sodium in the United States diet (U. S. Dept., 2015). All commercially processed and prepared foods are potentially high in sodium.

Alcohol is included in the category of other items to limit as part of a healthy eating pattern because it is a source of extra calories that do not provide nutrients. Moderate consumption of alcohol is often misinterpreted and 2 out of 3 drinkers exceed the recommendations (U.S. Dept., 2015). If alcohol is consumed it should be limited to one drink per day for women and two drinks per day for men, and only by adults of the legal drinking age.

Guideline Four: Shift to Healthier Food and Beverage Choices

> *Choose nutrient-dense foods and beverages across and within all food groups in place of less healthy choices. Consider cultural and personal preferences to make these shifts easier to accomplish and maintain.* (U.S. Dept., 2015)

Guideline Two discusses the rational for consuming a variety of foods, focusing on nutrient density and amounts needed to stay within energy requirements. Guideline Four expands on guideline two discussing the need of making gradual shifts to healthier food choices to reach the goal of a health promoting eating pattern. It is estimated that almost half of Americans suffer from at least one chronic disease state that are related to poor dietary intake (U.S. Dept., 2015). Americans are consuming excessive calories as reflected in the high percentage of Americans that are overweight or obese. To stay within energy requirements and meet nutritional needs, nutrient-dense food should be consumed. Nutrient-dense foods are classified as foods that are high in nutrients but low in calories. Foods typically consumed by Americans are not nutrient dense due to the added calories from fat and sugar. Recommended shifts towards a healthier eating pattern include switching from processed foods with added fats and sugars to more nutrient-dense foods. Simple recommendations can be seen in Table 54.2.

The new Nutrition Fact Label can guide consumers in choosing foods that are lower in saturated fats, sodium, and added sugars. The new label lists serving sizes that reflect the amount of food consumers customarily eat and has a new section identifying added sugars. Vitamin D and potassium are now listed on the label as these nutrients are often deficient in the American diet. More information on the nutrition fact label can be found at https://www.fda.gov/downloads/Food/LabelingNutrition/UCM511646.pdf.

Table 54.2 Simple food swaps to increase nutrient density

Instead of…	Try this…
White bread	100% Whole Wheat Bread
Soda, juice “drinks”	100% Fruit Juice or water
Fruit, fruit products with added sugar	Whole fruit
Salty processed snacks	Unsalted nuts or popcorn
Whole fat dairy	Low-fat dairy
Butter or solid fats	Healthy Oils

Guideline Five: Support Healthy Eating Patterns for All

> *Everyone has a role in helping to create and support healthy eating patterns in multiple settings nationwide, from home to school to work to communities (DGA, 2015).*

The Social-Ecological Model seen in Fig. 54.2 is a tool designed for health professionals to better understand the factors that contributes to an individual’s health outcome. The model examines the impact of social and cultural norms and values, sectors providing programs, the settings in which food and beverages are offered, and individual factors have on health outcomes. Knowledge of factors within the model can help the health professional and/or caregivers tailor an eating pattern that best fits an individual’s profile and personal preferences. Individual eating patterns based profile and preferences are more likely to be adapted and maintained over time. Incorporating the recommendations in a personalized fashion may result in meaningful shifts in food consumption resulting in improved health outcomes. Evidence supports improved eating behaviors when changes are made at various levels of the Social-Ecological Model (U.S. Dept., 2015).

Barriers and Strategies: Following a Healthy Eating Pattern Across the Life Span

There are many barriers to a healthy eating pattern noted among the general population including psychological beliefs, environmental factors, cultural conditions, and nutrition knowledge. Lack of nutrition knowledge with confusion over current research and a preference for fad diets and busy schedules that limit time for food preparation and increase reliance on convenience foods and fast/casual dining, and higher cost of fresh foods over processed foods are the barriers still present for individuals with intellectual disabilities and their families but are greatly exacerbated by the increased difficulties of daily life and disability specific nutritional needs and conditions.

Barrier: *Competing High Urgency Needs.*

When children are diagnosed with an intellectual disability at a young age, parents are often devastated and overwhelmed. Many families spend their days rushing between evaluation and intake appointments, therapy sessions, and meetings with a variety of agencies involved in their child's treatment and care. This loaded schedule is in addition to the requirements of daily life—working, managing a household, and caring for other family members. Nutrition and food often end up at the bottom on the priority list and contributes to greater reliance on fast food and convenience foods. This barrier continues to be problematic across the life span when there is a poor nutritional foundation in early childhood.

Strategies: *Encourage nutrition as a priority*

- Educate parents and caregivers on the impact of food choices to overall health and wellness. The connection between nutrition and brain health is complex, but there is sufficient evidence to support the importance of food choices and healthy body weight. See Research Highlight 1.
- Parents and caregivers can be supported with easily implemented nutrition tips and resources as offered throughout this chapter and available at http://choosemyplate.gov.

Research Highlight 1: Education on the Impact of Nutrition on Learning

A 2014 review of the literature on the impact of obesity on learning notes that obese children (without ID) have lower task performance than normal weight peers, and that excess abdominal weight and insulin resistance as found in Type 2 Diabetes also reduces cognitive control (Hillman, Khan, & Kao, 2015). These impacts would likely have an even greater impact on children with ID. Parents of 22 children with ID and autism participating in a 10-week school-based nutrition and physical activity program reported decreased hospital visits and illness related school absences (Hinckson, Dickinson, Water, Sands, & Penman, 2013). Parents and caregivers can be motivated by the understanding that nutrition is not a lower priority item, and supported with resources and tools to make food part of the therapeutic experience to improve quality of life for individuals with intellectual disabilities. This education is ongoing for all direct care staff throughout the life span. Size was relatively small and included younger students, this study offers several workable options for creating healthful food environments for all ages. In order to undertake these changes, residential and direct care staff will need a basic food and nutrition knowledge base and support system.

Barrier: *Sensory Perception Difficulties.*

Most people can recall a time when they were coerced into trying a food that was unappealing due to visual sight, smell, or imagined taste. Many adults have vivid memories of sitting at the dinner table for hours in refusal of detested foods like slimy Brussels Sprouts, spicy chili, or funky fish. At the other end, there are foods that tasted bland and pasty and had to be choked down. Recalling traumatic sensory experiences of food can lead to refusal to try the same of similar foods for decades, or even a lifetime. Many children with ID are hypo-sensitive (have reduced sensation) or hyper-sensitive (are more sensitive), and may experience combined perceptual differences related to food stimuli. For individuals with intellectual disabilities, this heightened level of sensation impacts every meal and continues past childhood through adulthood.

Feeding programs that use the techniques of Applied Behavior Analysis have been used to reduce the sensory sensitivity of children with autism. Although this can be critical for proper nourishment, the ethics of these programs may be questionable. Individuals are "offered" a small bite of non-desirable food that must be eaten in order to receive a highly preferred food. Some parents of children who complete the programs cite their success as their child will eat anything after completion of the program. However, there are many other cases where individuals are traumatized and further restrict eating after being force-fed foods and then vomiting repeatedly during desensitization.

Individuals with disabilities may also restrict foods because of how they feel after eating a certain food and this can be mistaken for a sensory intolerance. Innate awareness of digestion difficulties due to undetected food allergies or intolerances is often overlooked by caregivers. Again, the communication challenges of individual with intellectual disabilities can inhibit expression of pain and discomfort. What might be perceived as an unprovoked aggression or irrational meltdown may be a result of eating aggravating foods. When individuals with intellectual disabilities severely restrict food choices, parents and professionals alike may be at a complete loss when it comes to deciphering an individual's food rules and nutrition suffers.

Strategies: *Shape sensory tolerances systematically*

- Assess food preferences and encourage new choices by presenting a variety of options in a "grazing bar." Small bite sized portions of fruits, vegetables, whole grains, and nuts/seeds can be placed in an ice cube or other divided tray. Note which types of foods are selected while paying close attention to the accompanying color, texture, flavor, and smell. Collect data and offer similar choices in future sessions.
- Once food preferences have been assessed and types of preferred tastes or textures identified, slowly shape choices into more nutrient-dense choices.
- When sensory perception is related to texture and before shaping is successful, change the texture of the food itself. Crunchy carrots can be cooked and pureed for a smooth texture. If a crunchy texture is preferred, offer crisp fruits, vegetables, and whole grain crackers.
- Serve food from a bowl or plate instead of a package or a box. In addition to controlling portion size this also disconnects the visual identification of food by the brand logo. This can decrease resistance to trying new foods.

Barrier*: Confusion of "fad" diets and cures.*

Additionally, families may receive restrictive dietary advice from other parents or professionals. One of the first pieces of nutrition related input for young children with autism is a regime eliminating all gluten (a protein found in wheat products) and casein (a protein in dairy products). The hypothesis is that children with autism symptoms may actually have a "leaky gut" that cannot absorb nutrients and that gluten and casein are not processed, turning into morphine-like chemicals that result in the repetitive and stereotypical behaviors seen in this disorder. There is little evidence to support this theory, although many families report anecdotal success (Piwowarczyk, Horvath, Łukasik, Pisula, & Szajewska, 2018). When gluten and/or casein are eliminated from the diet, dairy and whole grains must be removed and can further restrict healthy eating patterns unnecessarily.

Strategies*: Promote a healthy eating pattern to ensure adequate nutrient intake.*

- Professionals should educate parents and caregivers on the research and explain the difficulties with anecdotal support for dietary changes and advocate for a healthy eating pattern that includes all food groups.
- Professionals should share the MyPlate resources to encourage a healthy eating pattern.
- If parents or caregivers would still like to trial a specialized diet, they should be encouraged to work with a Registered Dietitian to ensure nutrient adequacy and avoid malnourishment and vitamin/mineral deficiencies.
- Professionals can also support diet trials through data collection that would provide information on successful outcomes.

Barrier: *Use of food rewards.*

Another common nutrition interference for young children with IDD who are followed any type of Applied Behavioral Analysis (ABA) program is the use of food rewards. This is also seen more commonly in children with a coexisting autism spectrum disorder, but food-based reinforcement is seen across the general population and likely more common in children with IDD. The use of favorite food rewards is successful for increasing skills, and it can be more difficult to find other motivators for individuals with intellectual disabilities. When children become adapted to grazing on their favorite treats all day during instruction/therapy sessions, it interferes with current intake of more healthful fruits and vegetables and also conditions the palate to prefer sweet/salty/high fat food choices.

Strategies: *Create a healthy food environment*

- Encourage non-food rewards by utilizing positive social interactions and games.
- Create a healthy food environment by making fruits and vegetables easily accessible and storing sweet/salty/high fat food choices out of sight. See Research Highlight 2.
- Assess for healthy food preferences using “grazing bar” method.
- Use of stoplight (slow, whoa, and go system) using “go” foods (fruits and vegetables) for rewards.

Research Highlight 2: Creating a Healthy Food Environment

In addition to educating caregivers and staff, changes to the food environment have also been shown to have a positive impact on adolescents and young adults ($n = 43$) with intellectual disabilities living in a residential school setting (Hubbard et al., 2015). This quasi-experimental study used digital photography to assess food selection and plate waste before and after a lunchroom intervention (Hubbard et al., 2015). The Smarter Lunchroom strategy encourages more healthful choices by moving energy-dense choices (i.e., desserts, peanut butter and jelly sandwiches) out of sight or later in the line and placing fruits and vegetables within easy reach and earlier in the line. While this intervention has been found successful in typical school settings, this study also adapted the intervention to the students’ needs by providing social stories and visual supports for communication.

The results showed statistically significant increases in whole grain selection and consumption with reduced refined grain selection and consumption, and increased fruit consumption (Hubbard et al., 2015). Another notable positive of this study is that autonomy was encouraged by providing free choice of side dishes instead of bundling sides into the main entrée without alternatives (Hubbard et al., 2015). Participants also had some say in menu planning through taste testing activities in the classroom (Hubbard et al., 2015). Although the sample size was relatively small and included younger students, this study offers several workable options for creating healthful food environments for all ages. In order to undertake these changes, residential and direct care staff will need a basic food and nutrition knowledge base and support system.

Barrier: *Lack of nutrition knowledge for individuals with disabilities and parents/caregivers.*

The plethora of nutrition information in the media is confusing for the general population, and even more so for individuals with intellectual disabilities. Our food environment offers a multitude of choices that are high in sugar and fat and low in nutrients, and these choices are promoted heavily with colorful and attractive packaging and commercials. Additionally, claims made on packaged foods such “high fiber,” “low fat,” or “less sugar” can make these products sound healthier than they are in reality. There is an abundance of inaccurate information on “quick fixes” for our dietary problem that do not work in the long term. The importance of a healthy eating pattern with shifts to increase nutrient density and decrease nutrients of concern is an evidence-based solution that should be clearly communicated to individuals and caregivers.

Strategies: *Educate and support with evidence-based resources*

- Adapt nutrition education for individual needs through interactive, hands-on lessons (see Research Highlight 3).
- Educate using MyPlate model with divided plate system (Research Highlight 4).
- Offer resources similar to MENU-AIDDS.
- http://www.goodnutritionideas.com/products/menu.html
- MyPlate10TipsSeries https://www.choosemyplate.gov/myplate-tip-sheets

Research Highlight 3: Interactive, Adapted Nutrition Education for Individuals with ID

A 2013 study to investigate if implementation of an appropriately planned nutrition education program for young adults with ID would result in changes in food choices to improve the nutrient adequacy of the diet revealed positive outcomes (Subach, 2017). The program design included active participatory lessons incorporating observation and modeling of healthy eating from both the researcher and the program peers. Food logs were used to record all food and drink consumed for a 3-day period at the start and end of the study. Averages of the 3 days were calculated, extracting total calories, fat, saturated fat, cholesterol, sodium, sugar, fiber, and vitamins A, C, and D.

Five nutrition lessons were developed utilizing the U.S. Department of Agriculture's (USDA) MyPlate, designed to meet the participants' needs, based on relevant literature review and on parental input. Nutrition instruction was added to the weekly exercise sessions. Participants were given their own plastic MyPlate for use at home as a visual aid and motivator. Each week featured a lesson and activity on a specific food group. Participants were given take-home food boxes, a corresponding activity, and standardized homework to be returned the following session. Analysis of the pre and post study food logs revealed a reduction of negative nutrients (fat, saturated fat, and sugar) and an increase in positive nutrients (vitamin A, C), although values were not statistically significant. Program reception was positive from parents and participants, as measured by the active participation in group activities, discussions and end of program surveys. There was an overwhelming request for expansion of the program to include shopping and cooking instruction.

Research Highlight 4: Nutrition Education for Direct Care Staff—Divided Plate System

A 2012 study focused on improving weight status for youth with developmental disabilities living in group homes that targeted staff knowledge of dietary guidelines and nutrition knowledge demonstrates the importance of the staff/client relationship (Gephart, 2012). The 4-month study included education on weight status and goal setting procedures and showed significant improvement in mean body mass index (Gephart, 2012). Improving the health literacy of direct care staff and caregivers is a necessary first step to shaping the food choices of individuals with disabilities.

A community-based nutrition intervention program sought to increase the nutrition knowledge of group home staff and subsequent meal adequacy for the 21 residents in four group homes (Edwards, Holder, Baum, & Brown, 2014). All staff attended two nutrition education sessions conducted by a registered dietitian on meal planning, grocery shopping, food preparation, and guidelines for food groups and serving sizes (Edwards et al., 2014). Additionally, portion divided plates were

(continued)

Research Highlight 4 (continued)

used in the homes with daily photographs to record the application of knowledge surrounding food groups and serving sizes before, during, and after the staff training (Edwards et al., 2014). The changes in meal adequacy were significantly positive based on the photographs, and demonstrate that the nutrition education program enhanced staff knowledge and also overcame perceived barriers of increased workload and negative behavior changes (Edwards et al., 2014).

Barrier: *Physical health issues.*

Physical health issues can complicate following a healthy eating pattern for many individuals with ID. These added health impairments may be part of the disability itself, such as feeding issues that occur with cerebral palsy and Down syndrome. Another possibility is that physical health declines due to aspects of the disability such as poor oral health, sensory dysfunction, or food intolerances. The physical health may result from the poor nutritional outcomes as a result of disability in the case of constipation, gastroesophageal reflux disease, nutrient deficiencies, and chronic disease. Detailed information on physical health needs for specific disabilities that may occur in conjunction with ID will be discussed at the end of this chapter.

Poor oral health care is a known nutrition risk factor for individuals with ID due to oral hypersensitivity limiting brushing/flossing/dental visits, reliance on caregivers for oral hygiene, food consistency aversions restricting food textures, and medication side effects impacting taste and saliva production. Access to routine dental visits due to limited transportation, competent providers, and insurance restrictions further exacerbates poor oral health (Ptomey & Wittenbrook, 2015).

Strategies*: Assess and treat underlying medical complications*

- Determine if there are underlying medical issues that need to be resolved to improve nutrition status and refer out to appropriate professional.
- Maintain education on disability specific strategies to ensure appropriate intervention.
- Consult with Registered Dietitian for nutrition assessment and development of care plan.

Barrier: *Communication Deficits.*

Communication difficulties impact following a healthy eating pattern in several ways. Children with ID may not be able to communicate the reason for refusing foods and caregivers must make assumptions on why food is uneaten. These may be incorrect. A food might be refused based on any number of reasons and it could be that the reason is not even related to the disability. Young children often react to new foods with disgust the first several times and the resistance to try new tastes may be heightened for kids with ID. Parents are in a difficult position and may be more inclined to placate their children with ID by offering preferred tastes of salty, sweet, and fatty foods when fruits, vegetables, whole grains, and lean proteins are initially refused.

Strategies*: Support communication and choice*

- Make sure that individual with ID has easy access to adaptive communication as needed to request food appropriately. While teaching communication, reduce access to foods so that they cannot be obtained without asking.
- Food preference assessments as described above can help ensure that individuals have correct vocabulary in any augmentative communication system (Picture exchange, sign language, technology-based apps and devices) to request food choices.
- Offer foods on numerous occasions to determine if they are truly not preferred.
- Collaborate with speech therapist and educate on importance of texture variety of foods for development of muscles for speech when appropriate.

Barriers*: Financial limitations.*

Financial resources and limited access to healthy foods can be a significant barrier to eating a healthy meal pattern in the general population but financial stressors associated with disabilities greatly increase this barrier for families affected by ID. Families often pay more out-of-pocket expenses and higher premiums for health insurance, reduce work hours due to caregiving demands, and have lower household income due to divorce from psychological stress (Goudie, Hall, Narcisse, & Kuo, 2014). Financial limitations continue across the life span for individuals moving into shared or independent living arrangements due to lower compensation, limited employment rates, and reliance on government funding. These excessive burdens are another factor that decreases the budget for nourishing, fresh foods.

Strategies: *Refer to External Resources*

- Refer to Nutrition Assistance Programs as Appropriate: Individuals with ID and their caregivers may not realize that they qualify for Supplemental Nutrition Assistance Programs (SNAP) https://www.fns.usda.gov/snap/supplemental-nutrition-assistance-program-snap . SNAP-Ed is another resource available to individuals and families utilizing Supplemental Nutrition Assistance that provides information on healthy eating with a limited food budget. Additional information on the program and eligibility can be found at https://www.fns.usda.gov/snap/supplemental-nutrition-assistance-program-education-snap-ed. SNAP also offers Farmer's Markets programs: www.fns.usda.gov/snap/supplemental-nutrition-assistance-program-education-snap-ed
- Offer additional resources from the Thrifty Meal Plan, MyPlate on a Budget, and Steps to Healthy Economical Meals: https://whatscooking.fns.usda.gov/sites/default/files/featuredlinks/MeetingYourMyPlateGoalsOnABudget.pdf, https://choosemyplate-prod.azureedge.net/sites/default/files/budget/StepsToHealthyEconomicalMeals.pdf
- Encourage the use of food in all forms according to seasonal availability and pricing. In the summer, fresh foods are often the more cost-effective choice, while frozen, canned, and dried foods can be inexpensive options for out-of-season produce. https://www.fruitsandveggiesmorematters.org/fresh-frozen-canned-dried-and-100-juice

Barriers: *Medication.*

Many individuals with ID require medication(s) that inhibit the ability to follow a healthy eating pattern or block nutrient absorption. Weight is altered through medications through increased or decreased appetite, carbohydrate cravings, changes in hormone production, metabolism, and thyroid function (Grondhuis & Aman, 2014). Common medications include atypical antipsychotics, antidepressants, anticonvulsants, and psychostimulants (Grondhuis & Aman, 2014). Many of these drugs can also interact negatively with the absorption of nutrients and increase the risk of other diseases (Ptomey & Wittenbrook, 2015). This is seen with prolonged use of anticonvulsants for epilepsy negatively affecting bone metabolism and heightening risk of fractures as well as acid reflux medications causing B12 deficiencies due to reduced stomach acid (Ptomey & Wittenbrook, 2015). Specific disabilities may also require additional individual medication for comorbidities increasing the likelihood of multiple medication impacts and interactions.

Strategies: *Cross-collaborate to reduce medication-related nutrition interference*

- Collaborate with health professionals and prescribing physician to understand interactions between medications and potential side effects related to appetite, weight, and nutrient interference.
- Consult with a Registered Dietitian to prevent nutrient deficiencies and determine appropriate nutrient and calorie intake.
- Create a healthy food environment by making fruits and vegetables easily accessible and storing sweet/salty/high fat food choices out of sight.

Barrier: *Environmental Systems.*

Environmental systems like schools, day programs, churches, work sites, and residential homes face competing priorities with limited funds, and nutrition is often the lowest priority need.

Organizations that provide support services to individuals are typically reliant on government funding sources that are continually cut and barely meet the minimum of service needs. Direct care staff are underpaid which results in a high turnover rate of under-qualified and often unmotivated support staff. It can be challenging to develop consistent nutrition education protocols with constantly changing faces among workers. As life expectancy increases due to medical interventions, it is even more critical that environmental systems reduce the healthcare burden through preventive efforts including improvement of nutritional health.

Strategies: *Implement Integrated Wellness Programs*

- Educate on importance of nutrition for reducing negative behaviors, chronic disease, and improving learning and skills acquisition.
- Develop training and systems that promote wellness adapted to the needs of individuals.
- Ongoing nutrition assessment protocols in schools, adult programs, etc.
- Develop a "Train-the-Trainer" for easy implementation of system wide wellness programs (Research Highlight 4).

.

Barrier: *Self-determination and Independent Living.*

Research Highlight 5: Health Matters: Train the Trainer

The "Train-the-Trainer" Model to improve health status of individuals with intellectual disabilities ($n = 67$) was investigated in a 12-week randomized intervention with a specific program, Healthmatters, to determine the efficacy of this approach for providers ($n = 34$) in Community Based Organizations (CBOs) (Marks, Sisirak, & Chang, 2013). The study sought to determine if training the direct care staff impacted the health of adults with intellectual disabilities as demonstrated by improved physiological and psychosocial health status, improved nutrition and exercise knowledge and skills, and improved fitness status (Marks et al., 2013). A coin flip method was used for the 6 CBOs to randomize staff into either and intervention group receiving immediate training or a control group that was "wait-listed" for training after 6 months (Marks et al., 2013). The Health Matters: The Exercise, Nutrition, and Health Education Curriculum for People with Developmental Disabilities, based on the Transtheoretical Model of Behavior Change and Bandura's Social Cognitive Theory, was used in the protocol to train direct care staff to develop a structured program for the adults they support (Marks et al., 2013). After an 8-h, small group staff training, the direct card staff participants initiated the 5 modules of the Health Matters curriculum within the intervention groups (Marks et al., 2013).

The Health Matters: Train the Trainer approach showed promising results as seen in the outcome categories (Marks et al., 2013). Significant improvements for the intervention group were seen in psychosocial measures including Social/Environmental Supports for Exercise and Nutrition and health behaviors, which was a primary target (Marks et al., 2013). This study provides a methodology for expanding the knowledge and skills of direct care staff to support improved health status of the individuals with intellectual disabilities that they support in community settings (Marks et al., 2013)

As individuals with disabilities enter adolescence, new challenges emerge. Throughout adulthood and aging, the quest for independence and self-determination continues and further impacts healthy eating patterns among adults with ID. When individuals with intellectual disabilities reach adulthood, supportive caregivers confront a dilemma between supporting self-determination and ensuring health and wellness. In this situation, "self-determination" refers to the right of individuals with disabilities to make their own choices regarding their budgets, services, and overall direction of life (Nonnemacher & Bambara, 2011). This essential freedom is necessary for all adults to have a high quality of life (Nonnemacher & Bambara, 2011). Staff and caregivers may compromise responsibilities for health to ensure that the individual has the right to choose unhealthy eating patterns and sedentary behavior or vice versa (Gill & Fazil, 2013).

Adults can and should have independence to make their own food choices, but those with disabilities may need extra support and education on healthy eating and cooking. Those who live in group homes or congregate living facilities are at even greater nutritional risk because nutritionally inadequate meals may be provided for them as a result of staff lacking food and nutrition relation knowledge and reduced food budgets (Edwards et al., 2014). The lifestyle, socioeconomic status, and personal preferences of staff influence how they support healthy eating guidelines and are potential barriers to nutritional health of residents (Gill & Fazil, 2013). There is also confusion for residential staff directed to follow a person-centered approach and still find balance between supporting an individual's right to choose their own food and ensuring the presence of healthful choices (Gill & Fazil, 2013).

Strategies: *Active Participation and Skills Instruction for Individuals with ID*

- Incorporate a nutrition assessment as part of the intake process to determine all preferred food choices for all day and residential programs.
- Utilize resources on MyPlate Website: The consumer website for MyPlate helps guide food choices at a more accessible level and offers many colorful resources with graphics that can support a variety of learning styles.
- Residents/consumers should actively participate in menu planning and grocery shopping.
- Use cooking instruction as a social activity to promote healthier eating at home and in the community. See Research Highlight 6.

Research Highlight 6: Cooking Instruction

Numerous research studies on improving food choices in young adults with disabilities have indicated a need for cooking instruction and socialization of persons with ID. In continuation of the adapted nutrition education program seen in Research Highlight 2, twenty participants took part in a pilot 6-week cooking series designed to increase their fruit, vegetable and whole grain intake. Participants were first educated on food safety using the "Clean, Separate, Cook, Chill" program by the USDA (U.S. Dept., 2015). Adherence to food safety guidelines is crucial to decease incidences of food borne illness which is addressed in Guideline Five.

Weekly sessions were held in the University's foods laboratory where participants were given basic food preparation instruction. Equipment modification and one-on-one recipe coaches were used to accommodate recipe execution. Participants followed pictorial recipes using pre-portioned, pre-cut, and frozen food items to ease in recipe completion. Participants took home food items to be used as part of their meal pattern for the following week. Foods prepared included oatmeal bowls, vegetable soup, fruit smoothies, baked potatoes and broccoli, rice bowls and chicken tenders. Program reception was positive with participants verbalizing confidence in their cooking ability and enjoyment of cooking with their peers. This led to the creation of a "coffee house" program that allowed participants to gather and prepare a meal at a local coffee roaster who donated space for the event. Feedback was positive from participants and their families, but lack of funding prohibited the continuation of the program. Outcomes from both programs indicate a need for further research and funding to support program that support healthy eating patterns in social situations.

Specific Disabilities and Case Studies

Specific disabilities that may have a comorbid intellectual disability also have individual physical health issues. Individuals with cerebral palsy (CP) have difficulty with muscle control and/or coordination affecting different parts of the day and are also often diagnosed with ID (Ptomey & Wittenbrook, 2015). The physical limitations of CP can result in over- or undernutrition due to feeding difficulties. Individuals who use a wheelchair for ambulation will have a reduced need for calories and may struggle with weight gain and higher body fat percentage (Ptomey & Wittenbrook, 2015). Individuals with CP may have trouble swallowing (dysphagia) and need enteral nutrition (tube feeding) that can result in inadequate fluid and fiber intake or over/undernutrition (Ptomey & Wittenbrook, 2015). When motor function is limited, the ability to independently eat and drink can result in a myriad of additional physical problems including gastroesophageal reflux disease, constipation, mouth and dental disease, and malnutrition (Ptomey & Wittenbrook, 2015).

AV Case Study

AV, a 25-year-old female, has a deletion on the fifth chromosome (-5q) and mild cerebral palsy. The chromosomal break left her genetically preprogrammed to a disease known as FAP (familial adenomatous polyposis). She was full-term, with a birth weight of 7lbs. 6 oz. She had no feeding challenges as an infant but was failure-to-thrive at 3 months, due to excessive sleeping. AV was diagnosed with low muscle-tone and mild cerebral palsy by age 1. AV consulted with a speech therapist for strategies to increase muscle strength in her mouth as a strategy to promote adequate nutrition intake. She was nonverbal until the age of six. At age 3 she was diagnosed with a B-12 deficiency which subsided after a 6-month regiment of B-12 injections. AV currently has a BMI 21.5 putting her within the normal weight classification. She had an ileostomy at age 12 and wears a stoma to collect the waste 24 h/day. Amy is an avid exerciser and swims competitively. Adequate hydration is a constant challenge due to the constant drainage of her GI system. Hot weather and exercise lasting over 1 h presents a challenge of maintaining hydration and electrolyte balance. She frequently suffers from dehydration and electrolyte imbalance during sporting events occurring in the absence of her parents. Dietary intervention for electrolyte imbalance includes Gatorade, bananas, and potato chips. AV's parents try to educate her on proper hydration practice, but due to the ID she often fails to consume adequate amounts of fluid. Gatorade is used on a regular basis to aid in hydration, but parents try to limit intake due to the sugar content. Food intake is monitored, excluding beans, nuts, and seeds and limiting fiber. Excessive fiber causes extreme pain and has resulted in a blockage and hospitalization. Citrus fruits cause skin irritation and are excluded. Dairy, carbonated beverages, and high sugar intake causes excessive bloating and pain. AV's current diet is based on trial and error, never receiving a nutrition consultation post ileostomy. AV faces challenges when in social settings where excessive food consumption is encouraged. Special attention is paid to the quantity and types of food consumed prior to limit odor from and bloating of the stoma.

Strategies for the Health Professionals

- Early intervention of nutrition consultations with a Registered Dietitian, providing the family with nutrition guidelines may prevent flare-ups and hospitalization.
- Education on hydration strategies, providing alternate beverage choice to decrease Gatorade use, however the RD should reinforce that occasional use does not have a significant negative impact on the overall quality of AV's relatively healthy diet.
- Education for AV on safe foods, appropriate portions and culinary instruction may increase her independence in making food choices.
- Education to caregivers and peers on AV's food restrictions.

Down Syndrome

Down syndrome is a common chromosomal disorder affecting 1 in 700–1000 live births. Individuals with Down syndrome typically have congenital heart disease, intellectual delays, shorter height, gastrointestinal problems, and low muscle tone that can negatively affect the ability to follow a healthy eating pattern. Feeding problems often begin during infancy with a weak ability to suck and poor weight gain. Constipation is common in persons with Down syndrome due to decreased gastric motility. A diet rich in fruits, vegetables, and whole grains can aid in improving gastric motility (Ptomey & Wittenbrook, 2015). Celiac disease or gluten sensitivity, which prohibits ingestion of gluten, a protein found in wheat is common and warrants adherence to a gluten-free diet. Persons with Down syndrome often have a high rate of overweight and obesity, with over half of this population being classified as overweight and up to 31% as obese (Ptomey & Wittenbrook, 2015).

Being overweight or obese is a risk factor in the development of cardiovascular disease and diabetes. Persons with Down syndrome have an increased risk of developing cardiovascular disease. Adelekan, Magge, Shults, Stallings, & Stettler (2012) compared lipid levels of 27 children with Down syndrome and their non-affected siblings with a hypothesis that children with Down syndrome would have higher levels of total cholesterol, low-density lipoprotein, triglycerides, and lower high-density lipoprotein levels than their non-affected siblings. Analysis concluded that the children with Down syndrome had a less desirable lipid profile than their non-affected siblings, independent of weight. Due to the increased risk of heart disease in the Down syndrome population, the results provide a significant contribution to the development of screening and intervention within this group.

Leptin Levels and Down Syndrome

Leptin, a satiety-regulating hormone that promotes energy homeostasis through energy expenditure, controls food intake. In the general population, high leptin levels have been associated with obesity and are now being linked to obesity levels in children with Down syndrome. Excessive amounts of leptin cause desensitization to the hormone, making satiety harder to achieve. Decreased satiety increases appetite, leading to overeating, which is a causal factor to weight gain. A clinical study comparing the leptin levels of 25 children with Down syndrome and 25 normal children found that the children with Down syndrome had higher levels of serum leptin levels for their percent body fat compared to normal children. Serum leptin levels were reported higher in females with Down syndrome than males (Sridevi et al., 2016)

Combating Obesity in Persons with Down Syndrome

A nutrient-dense diet consisting of low-energy-density foods such as fruits and vegetables is one strategy used to overcome the negative effect of the medical conditions faced by persons with Down syndrome. Nutrition education with an emphasis on proper portion sizes, healthy food choices, and meal planning is a suggested practice in controlling or combating overweight and obesity (National Down Syndrome Society, 2013). It is recommended to include persons with Down syndrome in menu planning, shopping, and meal preparation as a way of improving their nutritional status through self-determination.

SB Case Study

SB is a 28-year-old young woman who has Down syndrome (DS), autistic spectrum disorder (ASD), a bilateral sensorineural hearing loss, celiac disease, and an allergy to natural rubber latex. Her current BMI is 22.2 putting her within the normal weight classification. She was diagnosed with pituitary dwarfism and tested positive for celiac disease at age 12. She currently supplements with 250 mg calcium and 1000 IU vitamin D3.

SB consulted with a Registered Dietitian at the onset of diagnosis of celiac disease. Initial nutrition education was limited to routine medical visits and was not extensive. SB faces many challenges in meeting her nutritional needs while complying with a gluten-free diet. When she does not comply with her diet, she experiences the natural consequences of an upset stomach, nausea, and fatigue.

SB had been educated on the gluten-free diet, but due to her intellectual developmental disabilities (IDD) it is difficult for her to remember the variety of possibilities. The family focuses on eating naturally gluten-free foods, such as fruits, protein and vegetables and gluten-free grains such as rice and quinoa.

SB has difficulties in social situations communicating her need for food that is gluten free and also free from cross-contamination. Her hearing loss also complicates her ability to adequately communicate in social situations. She cannot eat foods that have been handled by someone wearing latex gloves, nor eat in a restaurant where latex gloves are routinely used for food preparation and handling. SB's participation in social events is often limited by these needs. Although SB has learned about the various food groups and examples of foods in each, she is not able to independently plan a diet that meets her nutritional needs. She requires prompting and assistance to plan, shop for, and prepare many foods, as well as communicate her various medical needs in other environments.

Strategies for Health Professionals

- Provide resources for health professionals and caregivers on the gluten-free diet can be found at the Celiac Foundation's website: https://celiac.org/marketplace/gf-products/ and at the National Celiac Association's website: https://www.nationalceliac.org
- Education of foods to avoid for individuals with a latex allergy. Foods commonly associated with allergic reaction to latex include apples, avocado, bananas, carrots, celery, chestnuts, kiwi, melons, papaya, raw potatoes, and tomatoes. Additional information on latex allergies can be found at The American College of Allergy and Asthma & Immunology website: https://acaai.org/allergies/types/skin-allergies/latex-allergy.
- Education for SB on safe foods and appropriate portions may increase her independence in making food choices. Use of APPs for smartphone to improve communication for SB when in restaurant and social settings. Professional and consumer resources on celiac disease can be found at https://www.eatrightstore.org/product-subject/celiac-disease
- Connect families with practitioners who specialize in celiac disease using the CDF Healthcare Practitioner Directory. The directory of a free listing of physicians, dietitians, mental health professionals and allied health providers who specialize in the treatment of celiac disease and other gluten-related disorders can be found at: https://celiac.org/celiac-disease/resources/provider-directory/#bTsbcyjhKEEvCPy
- Recommend caregivers complete the National Restaurant Association's Servsafe Allergen Training program. The program educates laymen and professional on safely providing food to populations sensitive to allergens. Information on this certification can be found at: https://www.servsafe.com/ServSafe-Allergens/The-Course

Autism

Individuals with autism commonly also have an intellectual disability and face additional nutrition risk factors related to social isolation, sensory-based food selectivity, increased sedentary activities, and common regimens of appetite increasing medications (Presmanes Hill et al., 2015). Additional health-related problems coinciding with autism include mood disorders, attention deficit disorders, and gastrointestinal conditions and these may exacerbate weight gain and chronic disease (Presmanes Hill et al., 2015).

Selective eating patterns seen in autism are a likely contributor to poor nutrition status. A 2015 study found higher rates of nutritional deficiencies despite higher body mass index (BMI) values in a group of young children with autism as compared to an age matched control group of typically developing peers (Shmaya et al., 2015). This pattern of overnutrition and undernourishment is connected to higher rates of chronic disease in the general population.

Aging Population

As the number of individuals diagnosed with autism increases, the aging population with the disorder also expands and with it, the potential for nutrition and weight related disease. Although there is limited data on the health status of adults with autism, a 2015 case-control study found significantly higher rates of almost all medical conditions among adults with autism ($N = 1507$) as compared to the control group including diet and lifestyle related conditions of obesity, dyslipidemia, hypertension, and diabetes (Croen et al.). Additional medical and psychiatric conditions were also found at significantly higher rates in the adult autism population. There are many potential reasons for the increased prevalence of secondary health issues in this group including restricted eating patterns due to sensory issues, medications that promote weight gain, lack of access to recreation and physical activity, and communication and social impairments (Croen et al., 2015).

Case Study: Autism and Vitamin B-12 Deficiency

MK, an active 16-year-old male with autism, consumed a self-restricted diet that contained no meat, dairy, eggs, or other animal products. After an uncomplicated appendectomy with pathology positive for a small tumor, MK also required resection of part of the small and large intestines. The surgeon did not refer to a dietitian and said to continue with "normal" diet. MK recovered well, but continued with a selective and limited diet. He did begin eating additional fruits and vegetables when his mother explained that these would help him not get any more tumors and stay healthy.

His parents sought a neurology consult after a suspected seizure when they found him lying on the ground. Seizure activity was not evident, but lab work to rule out other conditions determined that he had a severe B-12 deficiency with a serum level in the blood of 50 pg/mL. Values of less than 200 pg/mL are a sign of deficiency and will often be symptomatic (ADAM, 2018). Symptoms of B-12 deficiency can be psychiatric, hematological, and neurological and are often misdiagnosed. Due to the inability to communicate bodily sensations that may have been abnormal, the extent to which the B-12 deficiency enhanced other medical issues is unknown. It is possible that the increased weight loss, anxiety, and depression that MK had been experiencing were partially due to the B-12 deficiency. It is incredibly fortunate that blood work was checked before any additional neurological damage occurred. Intramuscular injections of B-12 were initiated and continued for 6 months with quarterly monitoring to ensure that serum B-12 increased. Oral supplementation continues to address this dietary deficiency with regular monitoring. B-12 supplementation is recommended for anyone who does not consume animal products.

Strategies for Health Professionals

- Suggest that a comprehensive panel of blood work be completed to check for nutrient deficiencies when there is a sudden change in behavior.
- Recommend consultation with a Registered Dietitian for any chronic digestion issues, and especially after gastrointestinal surgery.
- Understand that a "normal diet" may be a severely limited diet in individuals with autism and intellectual disabilities.
- Recognize the potential need for some supplementation under guidance of a medical professional when diet or surgical resection reduces nutrients to a level that can cause health complications.

Chapter Summary

It is evident that individuals with ID face extraordinary barriers on all fronts and nutritional health is no exception. Best practice recommendations include a comprehensive approach across home, school, and community environments, and one that involves individuals with ID as well as their caregivers and service providers in order to improve diet quality and nutritional outcomes across the life span (Steenbergen, Van der Schans, Van Wijck, De Jong, & Waninge, 2017) (Table 54.3).

Table 54.3 Lists a summary of barriers and potential strategies to be used to help individuals with ID meet the recommendations of the Dietary Guidelines for Americans to promote health and reduce risk of chronic diseases

Barriers and strategies: Following a healthy eating pattern across the life span	
Barriers	Strategies
Competing high urgency needs	Education on potential impact of nutrition on behavior, learning, and overall health Support with easy to implement tips and resources
Sensory perception difficulties	Continuous assessment of food preferences Slow, systematic shaping of food choices for nutrient density Use of texture modified foods Disconnection of link between food and brand
Fad diet/“cure” confusion	Education on lack of evidence Education on healthful eating patterns Professional support and data collection for trials of “special” diets
Use of food rewards	Assessment and use of non-food rewards Creation of healthy food environment Assessment of healthy food preferences Use of stoplight systems
Lack of nutrition knowledge	Education using MyPlate Model, MENU-AIDDS approach, MyPlate 10 Tips series Use of divided plate system Education on healthy choices for eating out
Physical health issues	Collaboration with health professionals on underlying medical issues Professional continuing education on disability specific needs Consultation with Registered Dietitian for nutrition assessment
Communication deficits	Adaptive communication Food preference assessments Repeated food trials Collaboration with speech therapist Education on importance of texture variety for speech development
Financial limitations	Education on Resources: SNAP and SNAP-Ed: https://www.fns.usda.gov/snap/supplemental-nutrition-assistance-program-snap https://www.fns.usda.gov/ebt/snap-and-farmers-markets www.fns.usda.gov/snap/supplemental-nutrition-assistance-program-education-snap-ed Thrifty Meal Plan Resource and MyPlate on a Budget: https://whatscooking.fns.usda.gov/sites/default/files/featuredlinks/MeetingYourMyPlateGoalsOnABudget.pdf Utilization of food in all forms: fresh, frozen, canned, and dried: https://www.fruitsandveggiesmorematters.org/fresh-frozen-canned-dried-and-100-juice Steps to Healthy Economical Meals. https://choosemyplate-prod.azureedge.net/sites/default/files/budget/StepsToHealthyEconomicalMeals.pdf
Medication	Collaboration with health professionals to ensure proper dosage Consultation with Registered Dietitian to adapt for any related nutrient deficits Creation of Healthy Food Environment

(continued)

Table 54.3 (continued)

Barriers and strategies: Following a healthy eating pattern across the life span	
Barriers	Strategies
Environmental systems	Education on potential impact of nutrition on behavior, learning, and overall health Development of training systems that promote wellness adapted for ID Use of ongoing environmental nutrition assessment protocols Implementation of cooking instruction for improved self-efficacy and social connectedness
Self-determination and independent living	Education and training for direct care staff Consideration of food-related vocational opportunities Implementation of cooking instruction

Acknowledgments A special note of thanks to Mary Kay Martin for her contribution to the section on macronutrients.

References

ADAM. (2018). *Vitamin B12*. https://medlineplus.gov/ency/article/002403.htm.

Adelekan, T., Magge, S., Shults, J., Stallings, V., & Stettler, N. (2012). Lipid profiles of children with Down syndrome compared with their siblings. *Pediatrics, 129*(6), e1382–e1387. https://doi.org/10.1542/peds.2011-1262

Croen, L. A., Zerbo, O., Qian, Y., Massolo, M. L., Rich, S., Sidney, S., & Kripke, C. (2015). The health status of adults on the autism spectrum. *Autism: The International Journal Of Research & Practice, 19*(7), 814-823. https://doi.org/10.1177/1362361315577517

Edwards, M., Holder, M., Baum, N., & Brown, R. (2014). Targeting health improvement via a nutritional intervention program for adults with developmental disabilities and challenging behaviors. *Journal of Policy & Practice in Intellectual Disabilities, 11*(1), 62–67. https://doi.org/10.1111/jppi.12067

Gephart, E. (2012). Use of prevention and prevention plus weight management guidelines for youth with developmental disabilities living in group homes. *Western Journal of Nursing Research, 34*(8), 1076–1077. 2p. https://doi.org/10.1177/0193945912453691

Gill, J., & Fazil, Q. (2013). Derogation of "duty of care" in favour of "choice"? *Journal of Adult Protection, 15*(5), 258–270. https://doi.org/10.1108/JAP-12-2012-0028

Goudie, A., Hall, D. E., Narcisse, M., & Kuo, D. Z. (2014). Financial and psychological stressors associated with caring for children with disability. *Families, Systems & Health: The Journal of Collaborative Family Healthcare, 32*(3), 280–290. https://doi.org/10.1037/fsh0000027

Grondhuis, S. N., & Aman, M. G. (2014). Overweight and obesity in youth with developmental disabilities: a call to action. *Journal of Intellectual Disability Research, 58*(9), 787–799. https://doi.org/10.1111/jir.12090

Hillman, C. H., Khan, N. A., & Kao, S. (2015). The relationship of health behaviors to childhood Cognition and Brain Health. Annals of Nutrition & Metabolism, *66*1–4. doi:https://doi.org/10.1159/000381237

Hinckson, E. A., Dickinson, A., Water, T., Sands, M., & Penman, L. (2013). Physical activity, dietary habits and overall health in overweight and obese children and youth with intellectual disability or autism. *Research in Developmental Disabilities, 34*(4), 1170–1178. https://doi.org/10.1016/j.ridd.2012.12.006

Hubbard, K. L., Bandini, L. G., Folta, S. C., Wansink, B., Eliasziw, M., & Must, A. (2015). Impact of a smarter lunchroom intervention on food selection and consumption among adolescents and young adults with intellectual and developmental disabilities in a residential school setting. *Public Health Nutrition, 18*(2), 361–371. https://doi.org/10.1017/S1368980014000305

Institute of Medicine. (2010). Dietary Reference Intake for Calcium and Vitamin D. Retrieved from http://www.nationalacademies.org/hmd/~/media/Files/Report%20Files/2010/Dietary-Reference-Intakes-for-Calcium-and-Vitamin-D/Vitamin%20D%20and%20Calcium%202010%20Report%20Brief.pdf.

Kim, S., Park, S., Carroll, D. D., & Okoro, C. A. (2017). Daily sugar-sweetened beverage conumption, by disability status, among adults in 23 states and the District of Columbia. *Preventing Chronic Disease, 14*, E132. https://doi.org/10.5888/pcd14.160606

Lagua, R., & Claudio, V. (1995). *Nutrition and diet therapy reference dictionary*. New York, NY: Champan & Hall.

Marks, B., Sisirak, J., & Chang, Y. (2013). Efficacy of the health matters program train-the-trainer model. *Journal of Applied Research in Intellectual Disabilities, 26*(4), 319–334. https://doi.org/10.1111/jar.12045

McGill, C. R., Birkett, A., & Fulgonii, V. L., III. (2016). Healthy eating index-2010 and food groups consumed by US adults who meet or exceed fiber intake recommendations. NHANES 2001–2010. *Food & Nutrition Research, 60*. https://doi.org/10.3402/fnr.v60.29977

National Down Syndrome Society (2013). Healthy eating habits in children with Down syndrome. http://www.ndss.org/.

National Institutes of Health. (2010). Vitamin D. https://ods.od.nih/gov/factsheets/VitaminD-HealthProfessional/#ref.

Nonnemacher, S. L., & Bambara, L. M. (2011). "I'm supposed to be in charge": Self-advoates' perspectives on their self-determination support needs. *Intellectual & Developmental Disabilities, 49*(5), 327–340. https://doi.org/10.1352/1934-9556-49.5.327

Office of Dietary Supplements. (2018). Vitamin B12. https://ods.od.nih.gov/factsheets/list-all/VitaminB12/.

Park, S., Xu, F., Town, M., & Blanck, H. M. (2016). Prevalence of sugar-sweetened beverage intake among adults — 23 States and the District of Columbia, 2013. *MMWR Morbidity and Mortality Weekly Report, 65*, 169–174. http://dx.doi.org/10.15585/mmwr.mm6507a1externalicon.

Piwowarczyk, A., Horvath, A., Łukasik, J., Pisula, E., & Szajewska, H. (2018). Gluten- and casein- free diet and autism spectrum disorders in children: a systematic review. *European Journal of Nutrition, 57*(2), 433–440. https://doi.org/10.1007/s00394-017-1483-2

Presmanes Hill, A., Zuckerman, K. E., & Fombonne, E. (2015). Obesity and Autism. *Pediatrics, 136*(6), 1051–1061. https://doi.org/10.1542/pedS.2015-1437

Ptomey, L., Goetz, J., Lee, J., Donnelly, J., & Sullivan, D. (2013). Diet quality of overweight and obese adults with intellectual and developmental disabilities as measured by the healthy eating Index-2005. *Journal of Developmental & Physical Disabilities, 25*(6), 625–636. https://doi.org/10.1007/s10882-013-9339-z

Ptomey, L. T., & Wittenbrook, W. (2015). Position of the academy of nutrition and dietetics: nutrition services for individuals with intellectual and developmental disabilities and special health care needs. *Journal of The Academy of Nutrition & Dietetics, 115*(4), 593–608. https://doi.org/10.1016/j.jand.2015.02.002

Rosinger, A., Herrick, K., Gahche, J., & Park, S. (2017). *Sugar-sweetened beverage consumption among U.S. adults, 2011–2014. NCHS Data Brief. No 270.* Hyattsville, MD: National Center for Health Statistics.

Shmaya, Y., Eilat-Adar, S., Leitner, Y., Reif, S., & Gabis, L. (2015). Nutritional deficiencies and overweight prevalence among children with autism spectrum disorder. *Research In Developmental Disabilities*, 381–6. https://doi.org/10.1016/j.ridd.2014.11.020

Sridevi, S., Adalarasan, N., Narayana Babu, R., Arasar Seeralar, A. T., Srilakshmi, R., & Anadan, H. (2016). Serum leptin levels in Down syndrome versus normal children. *International Journal of Scientific Study, 4*(2). https://doi.org/10.17354/ijss/2016/279

Steenbergen, H. A., Van der Schans, C. P., Van Wijck, R., De Jong, J., & Waninge, A. (2017). Lifestyle approaches for people with intellectual disabilities: A systematic multiple case analysis. *Journal of the American Medical Directors Association, 18*(11), 980–987.e3. https://doi.org/10.1016/j.jamda.2017.06.009

Subach, R. (2017). Improving food choices and nutrient adequacy in adolescents/young adults with developmental disabilities. *Journal of Disability and Health, 11*(1), 139–142. https://doi.org/10.1016/j.dhjo.2017/05.003

Tripkovic, L., Lambert, H., Hart, K., Smith, C. P., Bucca, G., Penson, S., … Lanham-New, S. (2012). Comparison of vitamin D2 and vitamin D3 supplementation in raising serum 25-hydroxyvitamin D status: Systematic review and meta-analysis. *American Journal of Clinical Nutrition., 95*(6), 1357–1364. https://doi.org/10.3945/ajcn.111.031070

U.S. Department of Health and Human Services and U.S. Department of Agriculture 2015–2020 Dietary Guidelines for Americans. (2015, December). 8th ed. Retrieved from https://health.gov/dietaryguidelines/2015/guidelines/.

Wimalawansa, S. J. (2016). Vitamin D adequacy and improvements of comorbidities in persons with intellectual developmental disabilities. *Journal of Child Development and Disorders, 2*(3). https://doi.org/10.4172/2472-1786.100030

Reference Website Links

The American College of Allergy and Asthma & Immunology

https://acaai.org/allergies/types/skin-allergies/latex-allergy.

Celiac Foundation

https://celiac.org/marketplace/gf-products/

CDF Healthcare Practitioner Directory

https://celiac.org/celiac-disease/resources/provider-directory/#bTsbcyjhKEEvCPy

Celiac Disease Professional and Consumer Resources

https://www.eatrightstore.org/product-subject/celiac-disease

Fruits and Veggies More Matters

https://www.fruitsandveggiesmorematters.org/fresh-frozen-canned-dried-and-100-juice

National Celiac Association

https://www.nationalceliac.org

SNAP and SNAP-Ed

https://www.fns.usda.gov/snap/supplemental-nutrition-assistance-program-snap https://www.fns.usda.gov/ebt/snap-and-farmers-markets

www.fns.usda.gov/snap/supplemental-nutrition-assistance-program-education-snap-ed

Menu-Aidds

http://www.goodnutritionideas.com/products/menu.html

Myplate Steps to Healthy Economical Meals

https://choosemyplate-prod.azureedge.net/sites/default/files/budget/StepsToHealthyEconomicalMeals.pdf

Nutrition Facts Label

https://www.fda.gov/downloads/Food/LabelingNutrition/UCM511646.pdf.

Servsafe Allergen

https://www.servsafe.com/ServSafe-Allergens/The-Course

Steps to Healthy Economical Meals

https://choosemyplateprod.azureedge.net/sites/default/files/budget/StepsToHealthyEconomicalMeals.pdf

Thirty Meal Plan and MyPlate on a Budget

https://whatscooking.fns.usda.gov/sites/default/files/featuredlinks/MeetingYourMyPlateGoalsOnABdget.pdf

55

Diabetes

Lisa C. Whitehead and Henrietta T. Trip

Diabetes in the General Population

Diabetes is a global issue with 8.5% of the population estimated to be living with diabetes (World Health Organization [WHO], 2016) with increasing prevalence in low-middle income countries. Three-quarters of those living with diabetes are living in low and middle income countries (International Diabetes Federation [IDF], 2017a, 2017b, 2017c). An estimated 1.6 million deaths were directly attributable to diabetes in 2015 (WHO, 2017) and range from 6% in Africa, 9% in Europe, 11% Western Pacific, South and Central America, 13% Middle East and North Africa, 14% South East Asia, North America and Caribbean of all mortality in the 20–79-year-old age group (IDF, 2017a, 2017b, 2017c): Type 1 diabetes (T1D) accounts for between 5% and 10% of total cases of diabetes worldwide (American Diabetes Association, 2008) and therefore type 2 (T2D) makes up to 90% (IDF, 2017a, 2017b, 2017c) .

Diabetes in People with Intellectual Disability

The epidemiology and prevalence of diabetes in people with intellectual disabilities (ID) is emerging. Genetic predisposition is linked to a higher risk of developing T1D. Type 1 diabetes has been reported to be more prevalent in individuals with Down syndrome at 10.6% compared with the general population (Anwar, Walker, & Frier, 2004). Turner syndrome, Klinefelter syndrome, Prader–Willi syndrome, Noonan syndrome and Williams syndrome have also been associated with an increased risk of endocrine conditions such as T1D (Hoybye, 2004; Botero & Fleischman, 2006). The prevalence of T2D in people with intellectual disabilities (ID) compared to the wider population is unclear. However, the most recent review of diabetes prevalence rates in people with ID (MacRae et al. (2015) identified 22 studies and a mean prevalence rate of 8.3%. Prevalence rates varied widely across studies, ranging from 0.4% to 25%. The variance likely reflects diagnostic as well as reporting issues; however, overall, the evidence suggests that prevalence rates may be higher for people with ID than for the wider population.

The prevalence of obesity in adults with ID is greater than that found in the general population (Hsieh, Rimmer, & Heller, 2014; Melville et al., 2008; Rimmer & Yamaki, 2006; Stedman & Leland Jr, 2010). Obesity is strongly linked to the

L. C. Whitehead (✉)
School of Nursing and Midwifery, Edith Cowan University, Joondalup, WA, Australia
e-mail: l.whitehead@ecu.edu.au

H. T. Trip
Centre for Postgraduate Nursing Studies, University of Otago Christchurch, Christchurch, Canterbury, New Zealand

J. L. Matson (ed.), *Handbook of Intellectual Disabilities*, Autism and Child Psychopathology Series, https://doi.org/10.1007/978-3-030-20843-1_55

development of T2D. Using body mass index (BMI) as a measure, obesity has been linked to gender in adults with ID (Emerson, 2005; Hsieh et al., 2014; Yamaki, 2005), the severity of ID (Emerson, 2005), place of residence (Bhaumik, Watson, Thorp, Tyrer, & McGrother, 2008; Hsieh et al., 2014), use of psychotropic medication (Hellings, Zarcone, Crandall, Wallace, & Schroeder, 2013; Hsieh et al., 2014) and cause of ID (Bhaumik et al., 2008; Hsieh et al., 2014). It is likely that the development of obesity-induced insulin resistance involves a complex interplay of genetic and environmental factors (Samuel & Shulman, 2012). Obesity in turn is related to a more sedentary lifestyle and poorer diet (Taggart, Brown, & Karatzias, 2014) As a population, people with ID have been found to have higher rates of cardiovascular disease, hypertension and obesity; risk factors for developing diabetes (Havercamp, Scandlin, & Roth, 2004; Merrick et al., 2004; Shireman, Reichard, Nazir, Backes, & Greiner, 2010; Taggart et al., 2014).

It is clear that, as with the general population, the number of people with ID affected by diabetes is likely to increase (Sohler, Lubetkin, Levy, Soghomonian, & Rimmerman, 2009).

What Is Diabetes?

Diabetes is a condition that develops over many years and the aetiology is a combination of ineffective insulin and not enough insulin. The body becomes resistant to the normal effects of insulin and gradually loses the capacity to produce enough insulin in the pancreas. Insulin is vital in converting glucose into energy and in T2D, the result is commonly hyperglycaemia (too much glucose in the blood). As a result of insulin resistance, the pancreas responds by producing greater amounts of insulin in an attempt to regulate blood glucose levels; over time, these cells become dysfunctional. At diagnosis, it is estimated that an individual will have “lost” 50–70% of their insulin producing cells (Diabetes Australia, 2015).

A healthy lifestyle with daily routines is essential in controlling blood glucose; however, many people living with diabetes in the general population are unable to maintain glycaemic control within the recommended levels (4–7%; 20–53 mmol/mol) (Peyrot et al., 2013). Diet, exercise, stress reduction and medication management are the key mediators of glycaemic control (National Institute for Health and Care Education [NICE], 2015) and are strongly influenced by self-management through individual behaviour and action (Whitehead, Trip, Hale, & Conder, 2016; Wilkinson, Whitehead, & Ritchie, 2011). The short and long-term effects of hyperglycemia are multiple and include microvascular changes (e.g. retinopathy, nephropathy, and neuropathy) and macrovascular changes (e.g. heart disease) (D’Elia, Barba, Cappuccio, & Strazzullo, 2011; World Health Organization [WHO], 2016). Neurovascular changes linked to diabetes include peripheral arterial disease and peripheral neuropathy. Symptoms may include a person not registering the lack of sensation in the feet resulting in decreased recognition of pain, pedal deformities and/or injury, impaired circulation, ulcer formation and possibly gangrene (IDF, 2017b).

Experience of Living with Diabetes-Perspectives from People with ID, Carers and Families and Service Providers

Nine papers have explored the experience of managing diabetes from the viewpoint of people living with ID and/or carers or service providers. These studies provide important insight into living with diabetes, current support and service provision. Two of the eight studies were conducted in the Netherlands: Cardol, Rijken, and van Schrojenstein Lantman-de Valk (2012a, 2012b); three in the UK: Brown et al. (2017); Dysch, Chung, and Fox (2012) and Rouse and Finlay (2016); three in New Zealand: Hale, Trip, Whitehead, and Conder (2011), Trip, Conder, Hale, and Whitehead (2016), and Whitehead et al. (2016) and one in Australia: Rey-Conde, Lennox, and McPhee (2005). Three of the studies were drawn from the same sample of people with ID (Hale et al., 2011; Trip et al., 2016; Whitehead et al., 2016); however, all three of the studies had a different focus with the analyses yielding different results.

The majority of participants were diagnosed with T2D, a mild or moderate level of ID, resided in residential or community housing settings and Down syndrome was the most commonly reported co-existing condition. The main themes identified were participants' understanding of diabetes, challenges to managing diabetes and factors that supported the experience of managing diabetes.

Across the studies people with ID reported a basic understanding of their condition, notably its cause and complications (Dysch et al., 2012; Hale et al., 2011), awareness of changes in their blood glucose level (Dysch et al., 2012; Hale et al., 2011), familiarity with blood tests and of the need to take medication (Cardol et al., 2012a), the role of diet and physical activity (Hale et al., 2011) and the importance of attending medical appointments for review (Cardol et al., 2012a).

Areas identified by participants as challenging were; diabetes was only viewed as "serious" when insulin was required (Cardol et al., 2012a), difficulty differentiating between diabetes symptoms and symptoms arising from other comorbid health conditions (Cardol et al., 2012a) and the complexity of managing a number of conditions at the same time (Dysch et al., 2012; Hale et al., 2011). A difficult relationship with diabetes was described where participants reported feeling frustrated with the restrictions and inconveniences of diabetes (Dysch et al., 2012; Hale et al., 2011), a sense of loss in relation to food choices (Cardol et al., 2012a) and at times feeling fear, insecurity, and being overwhelmed (Rey-Conde et al., 2005). Diabetes was described as sometimes socially stigmatising (Dysch et al., 2012). Hale et al. (2011) also reported that the majority of participants could not recall receiving information about diabetes when they were first diagnosed and expressed a desire to learn more about their condition.

Support from family and caregivers was described as both facilitatory and as creating dependence. People living with ID and diabetes described getting more out of health review appointments when supported by family members or professional carers (Cardol et al., 2012a). Three studies described the action of family and caregivers as supportive and promoting independence and autonomy (Hillege, Gallagher, & Evans, 2013; Rey-Conde et al., 2005; Whitehead et al., 2016) with an increase in support only when assessed as necessary to maintain glycaemic control, such as illness or a change in medication regime (Whitehead et al., 2016). Cardol et al. (2012b) described a more prohibitive approach by carers through the restriction of dietary choices and provision of healthy alternatives on special occasions such as birthday parties.

Other findings indicated that where educational resources were available, they were not appropriate for people with ID due to being in formats that were not accessible (Hale et al., 2011). Rey-Conde et al. (2005) also reported that information on diet was insufficient leading to confusion. It is therefore important that educational resources are appropriately tailored for people with ID.

Limited training and knowledge of diabetes was described by families and carers involved in supporting people with ID. Both Rey-Conde et al. (2005) and Brown et al. (2017) reported staff training to be a barrier to effective self-management support with some issues related to high levels of staff turnover. Levels of knowledge varied according to professional background where nurses had either general or specialised training diabetes but social workers and care workers reported variable levels of training and for some, this was only as a consequence of them seeking out opportunities to improve their knowledge themselves (Cardol et al., 2012b). Trip et al. (2016) reported that some carers were unaware of diabetes management plans and Cardol et al. (2012b), Hillege et al. (2013) and Trip et al. (2016) reported an overall inconsistency in staff knowledge and training. Caregiver dilemmas also reflected a potential need for further training of caregivers. These dilemmas were between enabling autonomy and reducing risk, and were described in four studies (Cardol et al., 2012b; Rouse & Finlay, 2016; Trip et al., 2016; Whitehead et al., 2016).

Guidelines

A global guideline on diabetes management exists (IDF, 2012), along with specific country based guidelines. All have a consistent focus on patient education, dietary advice, managing cardiovascular risk, managing blood glucose levels and managing the risk of long-term complications (e.g. Diabetes Australia, 2018; Diabetes Canada Clinical Practice Guidelines Expert Committee, 2018; Harkins, 2008; IDF, 2017a; New Zealand Guidelines Group [NZGG], 2011; The National Institute for Health and Care Excellence [NICE], 2015; The Society of Endocrinology, Metabolism and Diabetes of South Africa [SEMDSA], 2017; UNITE for Diabetes Philippines, 2014). Support to achieve optimal management, however, is thought to only be reaching the minority (IDF, 2012) due to the size and complexity of the evidence-base, the complexity of diabetes care itself, a lack of proven cost-effective resources for diabetes care and the diversity in standards of clinical practice driving disparities in clinical care.

The guidelines, regardless of country of origin reflect the same key concepts. Diabetes UK (2017a) recently released guidelines setting out 15 essential indicators, also referred to as the 15 Healthcare Essentials which, if achieved by adults with T2D, underpin good quality care. The 15 Healthcare Essentials are the minimum level of healthcare everyone with T2D require to have in place and in Western countries, expect from their healthcare team. These are:

1. Getting your blood glucose levels measured.
2. Have your blood pressure measured.
3. Have blood fats (cholesterol and triglycerides) measured.
4. Ensure eyes are screened.
5. Having your feet and legs checked.
6. Have your kidney function monitored.
7. Get individual dietary ongoing advice.
8. Getting emotional psychological support.
9. Being offered a group education course.
10. See specialist diabetes healthcare professionals.
11. Receive good care if admitted to hospital.
12. Get a free flu vaccine.
13. Have the opportunity to talk about any sexual problems.
14. If you smoke, get support.
15. Get information and specialist care if you are planning to have a baby.

Little guidance exists on diabetes assessment and management for people with ID. Of note, national or international guidelines for T2D vary in regard to whether people with ID are included and/or how they are represented. There may be similarities in reference to other vulnerable populations with cognitive impairment and communication difficulties such as people with acquired-brain injury, stroke, severe mental illness, and dementia for example. Whilst the concept of *Reasonable Adjustments* within employment legislation is not new, the Convention on the Rights of Persons with Disabilities (United Nations, 2006) requires this to be readily implemented across life domains for people with a disability—including healthcare. Diabetes UK (2017b) recently released a *Guideline on How to Make Reasonable Adjustments to Diabetes Care for Adults with Learning Disability* and advocates the following service considerations:

1. Make information accessible and user friendly.
2. Provide training for all staff.
3. Address social barriers.
4. Involve supporters.
5. Plan for and make reasonable adjustments.

There is no evidence, however, that people with ID should attend reviews, be screened or assessed any more or less frequently than the general population. In this respect all of the general guidelines around frequency of assessment apply: What does differ, however, is the need for health and social service providers to frame the encounter to ensure equity of access, maximum benefit and impact on health outcomes for the person with ID. The following suggestions are reasonable adjustments to improve communication and support:

- Ongoing education and training for the self-monitoring of blood glucose.

- Attend annual health checks for HbA1c blood glucose levels, blood pressure, eye checks and flu vaccination.
- Having the support of a family/paid carer with the person with ID at an annual health check and when blood is taken, blood pressure is measured, eyes are checked and the flu-vaccination is given.
- Use of a easy read book with accessible visuals to help explain what is involved in taking blood, taking blood pressure, having eyes checked, receiving a vaccination, how to cook healthy meals, reading food labels and making healthy choices.
- Referral to a dietician and a cooking program to demonstrate how to plan meals, how to cook basic healthy meals and how to manage barriers such as feeling hungry.

A number of plain language resources exist that can be freely accessed and cover a range of topics related to self-management including diet, exercise, blood tests, eye tests, and foot care. These can be downloaded from www.easyhealth.org.uk, www.diabetes.org, www.diabetesaustralia.com.au, www.diabetes.org.uk, and www.safefood.eu.

Self-Management and Self-Management Interventions

Diabetes is a condition in which lifestyle can impact insulin and other medication requirements and glucose concentrations. The self-management of diet, exercise, and, for many, medication are important in managing both type 1 and type 2 diabetes (Reddy, Rilstone, Cooper, & Oliver, 2016; Whitehead et al., 2017). The principles of diabetes management are relevant to all populations. However, where people with ID are screened less often than recommended in national guidelines (Shireman et al., 2010; Taggart, Coates, and Truesdale-Kennedy, 2012), and structured education programmes have not recognised or addressed the specific challenges faced by people with ID, potential opportunities for health promotion are likely being missed (Taggart et al., 2014). Autonomy, the individual's capacity to govern him or herself and decision-making underpin self-management.

Internationally, it is recommended that education is ongoing both in terms of diabetes itself as well as concepts and the practice of self-management for the people themselves and those in their network of support (American Diabetes Association, 2018; SEMDSA, 2017; UNITE for Diabetes Philippines, 2014). Undertaking such education in groups can be beneficial (Diabetes Australia, 2015) as well as identifying peer-led opportunities to encourage those with diabetes to engage with education (Diabetes Canada Practice Guidelines, 2018; Hale et al., 2011), and in the practicalities of peer-led shopping, exercise and growing a garden for example (Hale et al., 2011). In supporting people diagnosed with T2D, a number of countries have developed structured self-management education programs. For example, developed and implemented in the UK, Western Australia and in New Zealand, DESMOND (Diabetes Education and Self Management for Ongoing and Newly diagnosed Diabetes) is a funded self-management diabetes education programme for the general population. This program is theoretically underpinned and has clearly been shown to improve biomedical and psychosocial benefits, as well as the increase in self-management strategies for those people who attend (Khunti et al., 2008; Khunti et al., 2012; Skinner et al., 2006). There may be other structured self-management diabetes education programs in other jurisdictions.

People with intellectual disability rarely access structured self-management education programs for diabetes that are routinely offered to the general population (MacRae et al., 2015; Taggart et al., 2014). The specific challenges related to cognitive limitations, communication difficulties, low levels of literacy skills and learning styles have been neither recognised nor addressed in the mainstream programs. In addition, little consideration of the support and information needs of paid carers has been given to understand and employ self-management concepts (Trip et al., 2016). Self-management of diabetes is recommended by health services across the world for people without disabilities (WHO, 2016). People with diabetes are encouraged, where possible, to attend structured self-management education programmes, such as Dose Adjustment for Normal Eating (DAFNE) for adults with T1D (www.dafne.uk.com) or DESMOND for adults with T2D (www.desmond-project.org.uk). Until recently, no intervention program developed or adapted for people with ID and diabetes was available. There is now some evidence of an initiative to actively screen for prediabetes and diabetes, the STOP program and the adaptation of a major program, DESMOND.

The STOP program, developed in the UK (Dunkley et al., 2017) involved a diabetes screening programme to determine the prevalence of previously undiagnosed T2D and impaired glucose regulation in people with ID. Screening was conducted in a variety of community settings (general practices, ID psychiatric services) and through publicity about the study. Screening involved the collection of anthropometric, biomedical and questionnaire data. Type 2 diabetes and impaired glucose regulation were defined according to (venous) fasting plasma glucose or HbA1c, following WHO criteria. Of the 930 adults (29% of those approached) participated. Mean age was 43 years, 58% were male and 16% of South Asian ethnicity. Most participants were either overweight or obese (68%). Diabetes status was successfully assessed for 675 (73%) participants: Nine (1.3%, 95% confidence interval 0.6 to 2.5) were found to have undiagnosed T2D, and 35 (5.2%, 95% confidence interval 3.6 to 7.1) had impaired glucose regulation. Key factors associated with abnormal glucose regulation included the following; non-white ethnicity and a first degree family history of diabetes. The results from this large multi-ethnic cohort suggest a low prevalence of screen-detected (previously undiagnosed) T2D and impaired glucose regulation in adults with ID. However, the high levels of overweight and obesity detected emphasised the need for targeted lifestyle prevention strategies, specifically tailored for the needs of people with ID.

The DESMOND program is an established diabetes self-management program that has recently been adapted for people with ID, the DESMOND-ID (Taggart et al., 2018). A pilot feasibility study of a structured self-management diabetes education program targeting HbA1c for adults with ID and Type 2 diabetes mellitus was conducted using a two-arm, individually randomised design. A total of 66 adults with disabilities across the UK met the eligibility criteria. Of these, 39 agreed to participate and were randomly assigned to either the DESMOND-ID programme ($n = 19$) or a control group ($n = 20$). The program consisted of seven weekly educational sessions. The primary outcome was HbA1c level, and secondary outcomes included BMI, diabetes illness perceptions, diabetes severity, quality of life, and attendance rates.

The DESMOND-ID program was reported as feasible to deliver and, with reasonable adjustments, participants could provide consent, complete the outcome measures, be randomised to the groups and attend most of the sessions, with minimal loss to follow-up. Using the fixed-effects model, the interaction between occasion (time) and condition resulted in a statistically significant decrease in HbA1c ($p < 0.05$); however, the CI was large. This is the first published study to adapt and pilot a national structured self-management diabetes education program for adults with intellectual disability. This study demonstrates that it is possible to identify, recruit, consent and randomise adults with intellectual disabilities to an intervention or control group and further studies involving adequately powered trials are recommended.

A newly developed intervention for adults with obesity, T2D and an intellectual disability based on standardised supported self-management delivered by diabetes specialist nurses plus treatment as usual was piloted by (House et al., 2018). This feasibility randomised controlled trial included adults aged >18 years with a mild or moderate intellectual disability, living in the community with T2D, on any therapy other than insulin. Most participants were recruited through referral by a primary care physician. Participants were assessed on their mental capacity to consent to research and the intervention. Inclusion criteria included

HbA1c > 48 mmol/mol (6.5%), BMI >25 kg/m^2, or self-reported physical activity below national guideline levels. In this study, the intervention was based on modifying existing approaches to supported self-management, with adjustments made to respond to barriers to self-care in people with an intellectual disability. It emphasised realistic goal setting, identifying resources and barriers likely to influence success in reaching goals, and regular self-monitoring of goal attainment. For the "supported" element the authors chose to use face-to-face contact and employed two diabetes specialist nurses from the local diabetes service, with no prior experience in learning disability or involvement in research concerning trials of complex interventions.

The experimental intervention was compared with treatment as usual alone. Feasibility outcomes included: recruitment and retention; intervention acceptability and feasibility; data collection and completeness for physiological state and values for candidate primary outcomes (HbA1c and BMI). A total of 82 participants (89% of those contacted and eligible) were randomised. All supported self-management sessions were completed by 35/41 participants (85%); only four of whom completed no sessions. Data on the follow-up candidate primary outcomes HbA1c and BMI were obtained for 75/82 (91%) and 77/82 participants (94%), respectively. Primary outcomes were similar across trial arms at baseline and follow-up; however, when comparing within-participant reduction in HbA1c and BMI in individuals with measures at baseline and follow-up, an effect size in the supported self-management arm of 0.33 (0.5/1.5) was observed for BMI, and for HbA1c it was 0.30 (0.17/0.57), whilst minimal effects were observed in the group receiving treatment as usual. Adherence to a supported self-management program and willingness to have blood taken for outcome measurement was high and the authors conclude that a definitive randomised controlled trial is feasible in this population.

These are the first two studies to adapt an intervention for people with ID and diabetes. Differences existed in the delivery, such that for one group the intervention run in a public space (Taggart et al., 2018) and the other was a one-to-one intervention often delivered in the person's own home (Dunkley et al., 2017). Both studies described making reasonable adjustments. Detail on the adjustments made to the DESMOND-ID program were not clear; however, participants reported that the content and the delivery of the program were user-friendly, and the educators advised that the adapted programme content, structure, curriculum, length of sessions, resources, health action plans and interactive sessions were at the appropriate level for those with a range of cognitive impairments and communication difficulties. The opportunity to provide booster sessions was described as a potential to further reinforce the messages of the program. The educators also found that the first session, for the carers only, was a useful means of creating a relationship with the carers, and was supportive of them working through the programme together with the adults with intellectual disabilities. Only one issue was raised by the educators in relation to the increased preparation time needed prior to delivery of the program (Taggart et al., 2018).

The intervention by Dunkley et al., (2017) was described as starting with three principles: that the intervention should respond to known barriers to self-management reported by people with a disability, including practical problems such as transport, likely attrition through drop-out when multiple attendances are expected, and an inability to accommodate the presence of a supporter. The format of the intervention should encourage self-maintained change beyond an early supported element and in this study a focus on involving supporters linked to any aspect of lifestyle relevant to diabetes; and the intervention should be designed to be readily integrated into usual healthcare provision, to ensure sustainability. The intervention was planned to focus on practical aspects of self-care; use of simple (accessible) written materials and charts; supportive contact both with a professional and with a supporter if one could be identified; use of practical goal setting and self-monitoring. The intervention sought to avoid theoretical content on the nature of diabetes and food values, using booklets to cover factual information. Interestingly, in contrast to the Taggart et al. (2018) study Dunkley et al. (2017) decided against group-based interventions predicting that attendance would be low and the ability to meet the specific individual needs of a heterogeneous population harder in such settings.

Conclusion

The challenges of self-management of glycaemic control apply to those with ID in even greater measure when ID can impact on capacity and capability to follow a healthy diet, take physical activity, monitor blood sugar levels, and take medication as prescribed (Whitehead et al., 2016). This means people with intellectual disabilities may be more reliant on family, paid carers and others to support them in making healthy lifestyle choices and in glycaemic control (Cardol et al., 2012a; Hale et al., 2011; Rouse & Finlay, 2016).

For effective diabetes self-management, people with ID require structured education about diabetes in which the language and approach is tailored and adapted to the individual (Bell, 2005; Taggart et al., 2015, 2018), as well as feedback on what has been mastered (Maine, Dickson, Truesdale, & Brown, 2017). Such approaches develop the autonomy and confidence of a person with an intellectual disability and those who support them (Cardol et al., 2012b; MacRae et al., 2015).

Key considerations for future research include further development of interventions specifically for people with ID and diabetes and establishing the core elements of the intervention, for example the value of one to one engagement versus group intervention, and the role that carers provide in promoting self-management.

Key considerations for clinical practice include the provision of joint clinics between primary care, diabetes services and intellectual disability healthcare staff to inform service delivery, accessibility and the continuity of care (Brown et al., 2017). Furthermore, education about diabetes to healthcare staff and people with intellectual disability alike should include the promotion of practice translation the principles of self-management, thereby enabling the inherent and developing autonomy of individuals with diabetes (Trip et al., 2016; Whitehead et al., 2016).

References

American Diabetes Association. (2008). Nutrition recommendations and interventions for diabetes: A position statement of the American Diabetes Association. *Diabetes Care, 31*, S61–S78.

American Diabetes Association. (2018). Standards of medical care in diabetes-2018. *The Journal of Clinical and Applied Research and Education, 41*(1), S105–S118.

Anwar, A., Walker, D., & Frier, B. (2004). Type 1 diabetes mellitus and Down Syndrome: Prevalence, management and diabetes complications. *Diabetic Medicine, 15*, 160–163.

Bell, D. M. (2005). Over the sea and far away: Ways of overcoming some of the difficulties arising in work with adults with learning disabilities living in geographical isolation in the United Kingdom. *British Journal of Learning Disabilities, 33*, 180–197. https://doi.org/10.1111/j.1468-3156.2005.00312.x

Bhaumik, S., Watson, J. M., Thorp, C. F., Tyrer, F., & McGrother, C. A. (2008). Body mass index in adults with intellectual disability: distribution, associations and service implications: a population-based prevalence study. *Journal of Intellectual Disability Research, 52*(4), 287–298. https://doi.org/10.1111/j.1365-2788.2007.01018.x

Botero, D., & Fleischman, A. (2006). Endocrinology. In I. L. Rubin & A. C. Crocker (Eds.), *Medical care for children and adults with developmemal disabilities* (pp. 387–398). Baltimore, MD: Paul Brookes.

Brown, M., Taggart, L., Karatzias, T., Truesdale, M., Walley, R., Northway, R., … Davies, M. (2017). Improving diabetes care for people with intellectual disabilities: A qualitative study exploring the perceptions and experiences of professionals in diabetes and intellectual disability services. *Journal of Intellectual Disability Research, 61*(5), 435–449. https://doi.org/10.1111/jir.12369

Cardol, M., Rijken, M., & van Schrojenstein Lantman-de Valk, H. (2012a). People with mild to moderate intellectual disability talking about their diabetes and how they manage. *Journal of Intellectual Disability Research, 56*(4), 351–360. https://doi.org/10.1111/j.1365-2788.2011.01472.x

Cardol, M., Rijken, M., & van Schrojenstein Lantman-de Valk, H. (2012b). Attitudes and dilemmas of caregivers supporting people with intellectual disabilities who have diabetes. *Patient Education and Counseling, 87*, 383–388.

D'Elia, L., Barba, G., Cappuccio, F. P., & Strazzullo, P. (2011). Potassium intake, stroke, and cardiovascular disease a meta-analysis of prospective studies. *Journal of the American College of Cardiology, 57*(1), 1210–1219. https://doi.org/10.1016/j.jacc.2010.09.070

Diabetes Australia. (2015). *General practice management of type 2 diabetes—2014–15* (p. 2014). Melbourne: The Royal Australian College of General Practitioners and Diabetes Australia. Retrieved from https://static.diabetesaustralia.com.au/s/fileassets/diabetes-australia/5ed214a6-4cff-490f-a283-bc8279fe3b2f.pdf.

Diabetes Canada Clinical Practice Guidelines Expert Committee. (2018). Diabetes Canada 2018. Clinical practice guidelines for the prevention and management of diabetes in Canada. *Canadian Journal of Diabetes, 42*, S1–S325.

Diabetes UK (2017a). *15 Healthcare Essentials: Getting the care you need.* Retrieved from www.diabetes.org.uk/Guide-to-diabetes/Managing-your-diabetes/15-healthcare-essentials

Diabetes UK (2017b). Guide to how to make reasonable adjustments to diabetes care for adults with a learning disability. Retrieved from https://www.diabetes.org.uk/Professionals/Resources/shared-practice/for-people-with-learning-disability.

Diabetes Philippines. (2014). UNITE for Diabetes Philippines. Philippine practice guidelines on the diagnosis and management of diabetes mellitus. Compendium of Philippine Meicine, 16th edition.

Dunkley, A. J., Tyrer, F., Gray, L. J., Bhaumik, S., Spong, R., Chudasama, Y., … Khunti, K. (2017). Type 2 diabetes and glucose intolerance in a population with intellectual disabilities: The STOP diabetes cross-sectional screening study. *Journal of Intellectual Disability Research, 61*(7), 668–681. https://doi.org/10.1111/jir.12380

Dysch, C., Chung, M. C., & Fox, J. (2012). How do people with intellectual disabilities and diabetes experience and perceive their illness? *Journal of Applied Research in Intellectual Disabilities: JARID, 25*(1), 39–49. https://doi.org/10.1111/j.1468-3148.2011.00641.x

Emerson, E. (2005). Underweight, obesity and exercise among adults with intellectual disabilities in supported accommodation in Northern England. *Journal of Intellectual Disability Research, 49*, 134–143.

Hale, L. A., Trip, H. T., Whitehead, L., & Conder, J. (2011). Self-management abilities of diabetes in people with an intellectual disability living in New Zealand. *Journal of Policy and Practice in Intellectual Disabilities, 8*(4), 223–240.

Harkins, V. (2008). A practical guide to integrated type 2 diabetes care.. Retrieved from https://www.indi.ie/images/public_docs/539_A_Practical_Guide_to_Integrated_Type_II_Diabetes_Care[1].pdf.

Havercamp, S. M., Scandlin, D., & Roth, M. (2004). Health disparities among adults with developmental disabilities, adults with other disabilities, and adults not reporting disability in North Carolina. *Public Health Reports, 119*(4), 418–426. https://doi.org/10.1016/j.phr.2004.05.006

Hellings, J. A., Zarcone, J. R., Crandall, K., Wallace, D., & Schroeder, S. R. (2013). Weight gain in a controlled study of risperidone in children, adolescents and adults with mental retardation and autism. *Journal of Child and Adolescent Psychopharmacology, 11*(3), 229–238.

Hillege, S., Gallagher, S., & Evans, J. (2013). The challenges for families managing an adolescent with an intellectual disability and type 1 diabetes. *Australian Journal of Advanced Nursing, 30*(3), 26–32.

House, A., Bryant, L., Russell, A. M., Wright-Hughes, A., Graham, L., Walwyn, R., … Farrin, A. (2018). Randomized controlled feasibility trial of supported self-management in adults with Type 2 diabetes mellitus and an intellectual disability: OK Diabetes. *Diabetic Medicine., 35*, 776–788.

Hoybye, C. (2004). Endocrine and metabolic aspects of adult Prader-Willi syndrome with special emphasis on the effect of growth hormone treatment. *Growth Hormone and IGF Research, 14*, 1–15.

Hsieh, K., Rimmer, J. H., & Heller, T. (2014). Obesity and associated factors in adults with intellectual disability. *Journal of Intellectual Disability Research, 58*(9), 851–863. https://doi.org/10.1111/jir.12100

International Diabetes Federation. (2017a). *Recommendations For Managing Type 2 Diabetes In Primary Care −2017*. Retrieved from www.idf.org/managing-type2-diabetes

International Diabetes Federation. (2017b). *Clinical Practice Recommendation on the Diabetic Foot-2017: A guide for health care professionals.* ISBN: 978–2–930229-86-7

International Diabetes Federation. (2017c). *IDF diabetes atlas 8th edition*. Diabetes Atlas. Retrieved from http://www.diabetesatlas.org/.

International Diabetes Federation Clinical Guidelines Taskforce. (2012). *Global guideline for type 2 diabetes.* Retrieved from https://www.iapb.org/wp-content/uploads/Global-Guideline-for-Type-2-Diabetes-IDF-2012.pdf.

Khunti, K., Gray, L. J., Skinner, T., Carey, E., Realf, K., Dallosso, H., … Davies, M. J. (2012). Effectiveness of a diabetes education and self-management programme (DESMOND) for people with newly diagnosed type 2 diabetes mellitus: three-year follow-up of a cluster randomized controlled trial in primary care. *British Medical Journal, 344*, e2333. https://doi.org/10.1136/bmj.e2333

Khunti, K., Skinner, T. C., Heller, S., Carey, M. E., Dallosso, H. M., Davies, M. J., & DESMOND Collaborative. (2008). Biomedical, lifestyle and psychosocial characteristics of people newly diagnosed with Type 2 diabetes: baseline data from the DESMOND randomized controlled trial. *Diabetic Medicine, 25*(12), 1454–1461. https://doi.org/10.1111/j.1464-5491.2008.02620.x

Maine, A., Dickson, A., Truesdale, M., & Brown, M. (2017). An application of Bandura's 'four sources of self-efficacy' to the self-management of type 2 diabetes in people with intellectual disability: An inductive and deductive thematic analysis. *Research in Developmental Disabilities, 70*, 75–84. https://doi.org/10.1016/j.ridd.2017.09.004

MacRae, S., Brown, M., Karatzias, T., Taggart, L., Truesdale-Kennedy, M., Walley, R., … Davies, M. (2015). Diabetes in people with intellectual disabilities: A systematic review of the literature. *Research in Developmental Disabilities, 47*, 352–274. https://doi.org/10.1016/j.ridd.2015.10.003

Melville, C. A., Cooper, S.-A., Morrison, J., Allan, L., Smiley, E., & Williamson, A. (2008). The prevalence and determinants of obesity in adults with intellectual disabilities. *Journal of Applied Research in Intellectual Disabilities, 21*, 425–437. https://doi.org/10.1111/j.1468-3148.2007.00412.x

Merrick, J., Davidson, P. W., Morad, M., Janicki, M. P., Wexler, O., & Henderson, C. M. (2004). Older adults with intellectual disability in residential care centers in Israel: Health status and service utilization. *American Journal on Mental Retardation, 109*(5), 413–420.

New Zealand Guidelines Group. (2011). *Guidance on the management of type 2 diabetes*. Wellington: Ministry of Health.

NICE. (2015). Type 2 diabetes in adults: Management. Accessed from http://www.nice.org.uk/guidance/ng28, 7 Jan 2016.

Peyrot, M., Burns, K. K., Davies, M., Forbes, A., Hermanns, N., Holt, R., … Skovlund, D. E. (2013). Diabetes attitudes wishes and needs 2 (DAWN2): A multinational, multi-stakeholder study of psychosocial issues in diabetes and person-centred diabetes care. *Diabetes Research and Clinical Practice, 99*(2), 174–184. https://doi.org/10.1016/j/j.diabres.2012.11.016

Reddy, M., Rilstone, S., Cooper, O., & Oliver, N. S. (2016). Type 1 diabetes in adults: Supporting self-management. *British Medical Journal*, (352), 10. https://doi.org/10.1136/bmj.i998

Rey-Conde, T., Lennox, N., & McPhee, J. (2005). *Diabetes and intellectual disability—Perceptions from people with disability and their supporters*. Retrieved from http://espace.library.uq.edu.au/view/UQ:9458.

Rimmer, J. H., & Yamaki, K. (2006). Obesity and intellectual disability. *Mental Retardation and Development Disabilities Research Reviews, 12*, 22–27.

Rouse, L., & Finlay, W. M. L. (2016). Repertoires of responsibility for diabetes management by adults with intellectual disability and those who support them. *Sociology of Health & Illness, 38*(8), 1243–1257. https://doi.org/10.1111/1467-9566.12454

Samuel, V. T., & Shulman, G. I. (2012). Mechanisms for insulin resistance: Common threads and missing links. *Cell, 148*(5), 852–871. https://doi.org/10.1016/j.cell.2012.02.017

Shireman, T. I., Reichard, A., Nazir, N., Backes, J. M., & Greiner, K. A. (2010). Quality of diabetes care for adults with developmental disabilities. *Disability and Health Journal, 3*, 179–185.

Sohler, N., Lubetkin, E., Levy, J., Soghomonian, C., & Rimmerman, A. (2009). Factors associated with obesity and coronary heart disease in people with intellectual disabilities. *Social Work in Health Care, 48*(1), 76–89. https://doi.org/10.1080/00981380802451160

Skinner, T. C., Carey, M. E., Cradock, S., Daly, H., Davies, M. J., Doherty, Y., … DESMOND Collaborative. (2006). Diabetes education and self-management for ongoing and newly diagnosed (DESMOND): Process modelling of pilot study. *Patient Education and Counseling, 64*(1–3), 369–377.

Stedman, K. V., & Leland, L. S., Jr. (2010). Obesity and intellectual disability in New Zealand. *Journal of Intellectual & Developmental Disability, 35*(2), 112–115. https://doi.org/10.3109/13668251003717928

Taggart, L., Brown, M., & Karatzias, T. (2014). Diabetes. In L. Taggart & W. Cousins (Eds.), *Health promotion for people with intellectual disabilities* (pp. 69–76). Maidenhead: Open University Press/McGraw-Hill Publishers.

Taggart, L., Coates, V., & Truesdale-Kennedy, M. (2012). Management and quality indicators of diabetes mellitus in people with intellectual disabilities. *Journal of Intellectual Disability Research, 57*(12), 1152–1163. https://doi.org/10.1111/j.1365-2788.2012.01633.x

Taggart, L., Coates, V., Clarke, M., Bunting, B., Davies, M., Carey, M., … Karatzias, T. (2015). A study protocol for a pilot randomised trial of a structured education programme for the self-management of type 2 diabetes for adults with intellectual disabilities. *Trials, 16*, 148–156. https://doi.org/10.1186/s13063-015-0644-y

Taggart, K., Truesdale, M., Carey, M. E., Martin-Stacey, L., Scott, J., Bunting, B., … Clarke, J. M. (2018). Pilot feasibility study examining a structured self-management diabetes education programme, DESMOND-ID, targeting HbA1c in adults with intellectual disabilities. *Diabetic Medicine, 35*, 137–146. https://doi.org/10.1111/dme.13539

The Society for Endocrinology, Metabolism and Diabetes of South Africa Type 2 Diabetes Guidelines Expert Committee. (2017). The 2017 SEMDSA guideline for the management of type 2 diabetes guideline committee. *Journal of Endocrinology, Metabolism and Diabetes of South Africa, 21*(Suppl/ 1), S1–S196.

Trip, H., Conder, J., Hale, L., & Whitehead, L. (2016). The role of key workers in supporting people with intellectual disability in the self-management of their diabetes: A qualitative New Zealand study. *Health and Social Care in the Community, 24*(6), 789–798. https://doi.org/10.1111/hsc.12262

United Nations (2006). *Convention on the rights of persons with disabilities. Optional protocol.* Retrieved from http://www.un.org/disabilities/documents/convention/convoptprot-e.pdf.

Whitehead, L. C., Trip, H. T., Hale, L. A., & Conder, J. (2016). Negotiated autonomy in diabetes self-management: the experiences of adults with intellectual disability and their support workers. *Journal of Intellectual Disability Research, 60*(4), 389–397. https://doi.org/10.1111/jir.12257

Whitehead, L., Crowe, M., Carter, J., Maskill, V., Carlyle, D., Bugge, C., & Frampton, C. (2017). A nurse-led interdisciplinary approach to promote self-management of type 2 diabetes: A process evaluation of post-intervention experiences. *Journal of Evaluation in Clinical Practice*. https://doi.org/10.1111/jep.12594

Wilkinson, A., Whitehead, L., & Ritchie, L. (2011). Factors influencing the ability to self-manage diabetes for adults living with type 1 or 2 diabetes. *International Journal of Nursing Studies, 51*(1), 111–122.

World Health Organization (2016). *Global report on diabetes*. Retrieved from http://apps.who.int/iris/bitstream/han-le/10665/204871/9789241565257_eng.pdf;jsessionid=F3F53A9E5D0E3A50BA8556F78667C596?sequence=1.

World Health Organization (2017) *Diabetes Fact Sheet*. Retrieved May 30, 2018, from http://www.who.int/en/news-room/fact-sheets/detail/diabetes.

Yamaki, K. (2005). Body weight status among adults with intellectual disability in the community. *Mental Retardation, 43*(1), 1–10.

Factors in Weight Management: A Research Story

56

Richard R. Saunders, Muriel D. Saunders,
Joseph E. Donnelly, and Lauren T. Ptomey

Prologue

Concurrent with the movement towards community integration, personal freedom and choice have become the hallmarks of community-based services for adults with intellectual and developmental disabilities (IDD). The more these adults have exercised their rights to freedom and choice, the more they are making the same lifestyle choices as those without IDD and, as such, have become overweight or obese. Designing ways to address issues of weight with these individuals is neither less nor more complex than similar issues with individuals without IDD. With both populations, understanding the causal factors and, therefore, understanding how to intervene is like peeling an onion: the removal of each layer leads to another. Moreover, every person and every onion is different.

As an example, let us begin with a success story from a year-long pilot project that will be described below in detail. We recruited about 35 participants from a day-service/employment facility and introduced them to a reduced-calorie diet. As might be expected, the participants showed varying degrees of weight loss over the 6-month intervention and, some, over the 6-month maintenance phase. Among these, a subset of 10 individuals showed more consistent weight loss than many others. Near the end of the 12-months we noticed cheering, high-fiving, and other forms of satisfaction outside the meeting room as individual weights dropped below 200 lbs. At that point, we learned that the local equestrian program for adults with IDD had a 200 lb. maximum weight for mounting a horse, and the facility was about to open for the spring. Clearly, choosing or not choosing to follow the diet had explicit, tangible consequences.

Briefly, this pilot encouraged a reduced calorie diet with small cash incentives for tracking foods and beverages consumed, included small cash incentives for walking and tracking steps taken, and monthly meetings with project staff to discuss problems and successes. Our experiences with these and additional participants revealed that there were just about as many motivating factors as there were participants. Some worked hard at weight loss motivated by a partner, relative, or friend. Some wanted to alleviate pain in limbs and joints that they attributed to excess weight. Some had been diagnosed as prediabetic or with gout. Some were motivated by seeing friends lose weight. Some enjoyed the small cash

R. R. Saunders (✉) · M. D. Saunders (✉)
Schiefelbush Institute for Life Span Studies,
University of Kansas, Lawrence, KS, USA
e-mail: msaundrs@ku.edu

J. E. Donnelly · L. T. Ptomey
Center for Physical Activity and Weight
Management, University of Kansas,
Lawrence, KS, USA
e-mail: jdonnelly@ku.edu; Lptomey@ku.edu

J. L. Matson (ed.), *Handbook of Intellectual Disabilities*, Autism and Child Psychopathology
Series, https://doi.org/10.1007/978-3-030-20843-1_56

incentives, although these were provided for reporting their food and beverage intake and not specifically for weight loss. It is perhaps noteworthy that with many participants, having cash in hand was a relatively rare event. Most received payment from their employers as direct deposit to their checking accounts or to a guardian of their finances. Others seemed to be motivated by the praise they received from the research staff. Some saw their role in our pilot as a "job" of sorts, as if they were working for the University, and took pride in it. Thus, as these individuals were similar to persons without disabilities in their problems managing weight, they were also similar in what motivated them to change.

In addition to motivational differences, we also learned of the many differences in their living environments, including their residences, social situations and the broader environments around their residences. Among the most influential of these was the rate of staff turnover in their homes. Only a few lived entirely independently and we observed the support staff for those not living independently often changed once or more across the research period. Turnover was potentially significant when staff attitudes about weight management or weight loss changed dramatically: some staff supported the participants efforts because they too were dealing with similar issues, but others did not, as they did not see the need. Others brought attitudes about the IDD population that conflicted with the goals of weight loss. A not uncommon refrain was, "You know, eating is one of the few things these people can enjoy." The potential for participants to move from a diagnosis of prediabetic to a Type 2 diabetic seemed to have little impact on this point of view—this despite the fact that diabetes would significantly alter the participants food and beverage choices, and perhaps a host of other medical, residential, and monetary issues as well.

Both staff and relatives often brought concerns about the components of the diet. Some felt that the sugar substitutes in diet beverages would likely be far more harmful than excess weight. Similar misgivings were voiced regarding the sodium content of many of the prepackaged entrees that formed a core diet component. Staff also voiced concerns about the difficulties in shopping for the entire home, when the needs of the participant differed from others. Purchasing entrees conflicted with the food budgeting process in the homes. How does one purchase bulk meats, for example, for 2 people and entrees for one person. This produced a real and complex cost dilemma. Similarly, staff felt uncomfortable serving different foods to different residents. Many individuals we encountered enjoyed raw vegetables served with generous portions of ranch dressing—a real "no no" on a weight loss diet. Many staff felt uncomfortable serving generous portions (including second helpings) to some residents, but not the participant. Desserts posed a similar problem.

Differences in various skills among the participants posed additional challenges. Some demonstrated good money skills and others struggled, in the grocery store, for example. Consider the complexities of shopping for a week on a limited budget. For all of us, it can be a difficult task to fill a grocery basket up to but not beyond our budget limit. As with us without IDD, it can be embarrassing to get nearly done with check out and realize we must return one or more items to the shelves; it can be particularly so for the person with IDD. Cooking and food preparation skills also came into play. Some of our participants who did cook often worked from a few basic recipes. To be successful at weight management, some of these had to be changed. One of the most essential changes was the need to reduce how much one cooked or prepared at each meal. We also observed that when participants opened a can or box of food, they often consumed its entire contents immediately, rather than reshelving the remainder for a later meal. The difficulties in changing food preparation and cooking were not limited to the participants. In many residences, the approaches and recipes derived from the staff's personal histories. Given any particular meal menu, provided by supervisory staff or dietary consultants, there is room for much individualization and improvisation and many such improvisations were counter to weight loss.

The foregoing suggests why this chapter will not reveal a single formula for successful weight management. It will, however, describe an approach which was successful with many participants despite the factors just described.

Literature Review of Research Findings

Intellectual and developmental disabilities (IDD), are characterized by significant limitations in both intellectual functioning (IQ <75) and in adaptive behavior including conceptual skills (e.g., language and literacy, self-direction), social skills (e.g., interpersonal skills, ability to follow rules) and practical skills (e.g., personal care, use of money/phone) which originate before the age of 18 (American Association on Intellectual and Developmental Disabilities, 2012). These limitations are likely to impact food choice, calorie counting, and so forth. Lack of regular physical activity, increased sedentary behavior (Bartlo & Klein, 2011; Sundahl, Zetterberg, Wester, Rehn, & Blomqvist, 2015), and unhealthy dietary choices (Adolfsson, Sydner, Fjellstrom, Lewin, & Andersson, 2008; Draheim, Stanish, Williams, & McCubbin, 2007; Robertson, Emerson, Baines, & Hatton, 2014; Stancliffe et al., 2011) are associated with weight gain in the general population, as well as in adults with IDD. In addition, genetic predisposition (Farooqi & O'Rahilly, 2005), the use of weight-inducing medications (De Winter, Bastiaanse, Hilgenkamp, Evenhuis, & Echteld, 2012; Hsieh, Rimmer, & Heller, 2013; Stancliffe et al., 2011), and living arrangement (e.g., institution, group home, family home) (de Winter et al., 2012; Melville, Hamiltom, Hankey, Miller, & Boyle, 2007; Stancliffe et al., 2011) all contribute to weight gain in this group. Individuals with IDD represent ~1% to 3% of the US population (American Association on Intellectual and Developmental Disabilities, 2012) with a prevalence of obesity (BMI ≥ 30 kg/m^2) equal to (Stancliffe et al., 2011) or greater than the general population (Hsieh et al., 2013; Melville et al., 2007; Robertson et al., 2014). Adults with IDD have a higher prevalence of obesity-associated health conditions including cardiovascular disease, risk factors for cardiovascular disease, cardiovascular disease mortality (Draheim, 2006; Lauer & McCallion, 2015; Reichard, Stolzle, & Fox, 2011), type 2 diabetes (Balogh, Lake, Lin, Wilton, & Lunsky, 2015; Shireman, Reichard, Nazir, Backes, & Greiner, 2010) and rates of hospitalization related to type 2 diabetes (Balogh et al., 2015) compared with their non-IDD peers.

Several reports from government agencies (2005) and professional organizations (Ptomey & Wittenbrook, 2015) have recommended efforts to reduce the prevalence of overweight and obesity in adults with IDD. However, the lack of evidence regarding the effectiveness of weight management, particularly evidence relative to weight maintenance following weight loss, limits the ability to make sound weight management recommendations for this group. Spanos, Melville, and Hankey (2013), in the most recent systematic review, identified a limited number of trials (n = 22) on weight management in adults with IDD. These trials were conducted over relatively short time frames (8–12 weeks) using nonrandomized designs, in small (n < 25), nonpowered, heterogeneous samples. The majority of trials (21/22, 95%) were not conducted in accordance with current weight management guidelines which recommend "≥ 6 months in a comprehensive lifestyle program that assists participants in adhering to a lower-calorie diet and in increasing physical activity through the use of behavioral strategies" (Jensen et al., 2014). Weight loss observed in these trials was minimal (<3%). In contrast, clinically significant weight loss (≥3%) was observed in 24 and 16 week nonrandomized, multicomponent trials that included energy reduced diets, increased physical activity, and behavioral interventions (e.g., Melville et al., 2011). None of the trials described in the Spanos review included a long-term (≥1 year) comprehensive weight maintenance program as recommended by current clinical guidelines (Jensen et al., 2014). Publications subsequent to the Spanos review (Spanos et al., 2013) have demonstrated the effectiveness of a 1-year weight maintenance intervention in a small sample (n = 28) of adults with IDD who achieved ≥3% weight loss over 16 weeks (Spanos et al., 2013) and clinically significant weight loss (~8 kg) in a sample of 33 adults with IDD attending a community occupational therapy day center, in response to a 17-week intervention consisting of reduced energy intake, physical activity and a token economy motivational system (Martinez-Zaragoza, Campillo-Martinez, & Ato-Garcia, 2015). Pett et al. (2013)

reported modest weight loss (~3 kg) in a small sample (*n* = 30) of young adults (~25 years) in response to a 12-week recreation center-based healthy lifestyle intervention that focused on nutrition and physical activity. However, Bergstrom, Hagstromer, Hagberg, and Elinder (2013) reported no change in BMI or waist circumference in 129 adults with IDD in community residences who completed a 12–16-month intervention designed to improve both diet and physical activity. In summary, prior research on weight management with individuals with IDD has not produced particularly remarkable results.

The majority of weight management trials in adults with IDD have focused on compliance with healthy eating recommendations or energy reductions using a conventional meal plan diet (Bergstrom et al., 2013; Martinez-Zaragoza et al., 2015; Pett et al., 2013; Spanos et al., 2013). Conventional diets (CD) are effective in the general population (Jensen et al., 2014); however, understanding, planning and complying with a CD may be difficult in adults with IDD, and may, at least in part, be associated with the general lack of success of weight management interventions in this group. The Stop Light Diet (SLD) (Epstein & Squires, 1988) which categorizes foods by energy content: green (low energy, consume freely), yellow (moderate energy; consume in moderation), and red (high energy, consume sparingly) has been shown to provide effective weight management for children (Collins, Warren, Neve, McCoy, & Stokes, 2006) and for adults with physical disabilities (Damschroder et al., 2014; Reichard et al., 2015), and may be appropriate for use in adults with IDD.

Our Research Story in Three Phases

Phase 1

In preliminary discussions among a number of researchers in the Life Span Institute at the University of Kansas, the problem of weight management in adults with IDD became a shared topic of interest between a team of behavior analysts with extensive experience in settings where these adults are supported and a team of experienced diet and exercise researchers. In thinking about research to address the problem, this group recognized that as a team, we knew relatively little about the dietary habits of these individuals. Thus, we set about to learn more and several phases of research unfolded.

First, we enrolled residents of several group homes of 3–5 individuals in an observation and data collection project. They consented to our recording their daily dietary intake with digital photo "plate-waste" methods (e.g., Ptomey et al., 2015). We began visiting each home prior to the serving of each breakfast and evening meal. We recorded the sizes and capacities of their meal utensils: plates, bowls, glasses, cups, and so forth. When the meal was served, we photographed each place setting before they ate. When finished, we photographed each place setting again. Notes were taken to clarify any possible confusion as to the food and beverage items, such as the types of bread, margarine, salad dressings, and so forth. We also photographed the contents of their lunch boxes (taken to work, for example) and photographed the contents when they returned in the evening. We also were permitted to follow them to those away settings and to record any additions or changes that occurred. For example, some made purchases from agency vending machines and others swapped food items with friends.

The photos and notes were analyzed by staff trained in evaluating the foods according to nutritional content and kilo-calories (kcal). The average daily consumption exceeded 2200 kcal and often ranged between 2500 and 3000 (typical needs range from 1800 to 2000). These levels were clearly not consistent with weight reduction.

Our time in these homes revealed some other circumstances with potential impact both on why intake was relatively high and how one might begin an intervention. For examples, the plates and bowls were often filled to capacity, second servings were offered when there were leftovers, and typically everyone received the same portions regardless of their height and weight. We also observed that meals followed diet plans (from supervisors and consulting professionals) that sometimes led to unintended consequences. For example, where one glass of orange juice was a recommended breakfast component, the only glasses in the home were actually tumblers with 12–16 oz. capacities. Staff served the glasses full or nearly so. Another observation was that our post-meal photos were superfluous as participants usually had empty plates and lunchboxes. (In our subsequent research, we observed that these circumstances were not unique to the particular homes in that first project; they were seen in many other settings as well.)

Seeking to design a weight loss intervention for evaluation with IDD individuals, the team shared an approach that was having some success in persons in the general population. That approach was referred to as the "packaged meal" approach. Specifically, individuals were encouraged to eat prepackaged entrees of 300 kcal or less at lunch and dinner, to eat five servings of fruits and vegetables daily, and to supplement those with low calorie shakes. The fruits and vegetables could be fresh, frozen or canned. The entrees could be those ordered online from weight management companies or could be frozen entrees from the local grocer. Shake mixes that could be "expanded" by adding water, fruit, and ice in a blender to make filling meals, were the supplements of choice. Such shake mixes are also available online and in local stores. The goal of the packaged meal approach was to limit intake to about 1200–1500 kcal per day, while providing feelings of "fullness." Moderate to vigorous exercise was also encouraged, but not scheduled or supervised by the team. This approach had several characteristics that seemed promising for individuals with IDD—simplified food purchasing, no calorie counting, and no extreme constraints on what foods to eat. Incorporation of pictorial charts of foods organized according to the Stop Light Diet, to allow more choices in meals, appeared useful, as well.

To have some confidence in the diet we were about to share with individuals with IDD, the behavior analysts (who could benefit from some weight loss themselves) began following the diet. With relatively close compliance for several months, the behavior analysts lost 8% and 12% of their baseline weights and suffered no ill affects. In short, the diet seemed safe to share.

Phase II[1]

We enrolled 73 participants, with mild or moderate intellectual disabilities (BMI = 37.0 ± 8.4, age = 31.6 ± 9.6 years, 56% female) (Saunders et al., 2011). Approximately 30% of participants lived at home, while others were in supported living consisting of 1–4 residents. We provided them with shake mixes that were donated by HMR Weight Management Resources, Corp. There are other makers of such mixes, and a few chose to purchase those. Most of our participants purchased their frozen entrees (e.g., Smart Ones, Lean Cuisine, Lean Gourmet, Healthy Choice) in local grocery stores and a few ordered entrees from HMR, WeightWatchers, NutriSystem, Jenny Craig and so forth. There are many, many entrees to choose from and frequently one brand or another is on sale at very affordable prices. Our participants were also encouraged to drink lots of water and to look for other beverages, such as diet sodas, with low or 0 calories. For those who wanted snacks, we encouraged lower-calorie snacks or snacks in smaller amounts. The 100-calorie packages of popular cookies and crackers became a common purchase for many participants. Others, with help from caregivers, purchased large volume packages and repackaged the contents into servings of approximately 100 calories. Of course, this tactic failed without close supervision.

This diet is also referred to as one based on "volumetrics." In such a diet, one tries to eat lower-calorie foods in high volume and eat higher-calorie foods in low volume. In keeping with that principle, we encouraged participants to eat extra servings of fruits or vegetables for a

---

[1] The weight charts, stop light guides, intake self-recording forms and so forth, described in Phases II and III may be found at, and downloaded from www.diet.ku.edu.

snack, rather than cookies, crackers, and candy. For example, we shared that it takes a lot of carrot sticks to get the same amount of calories as a very small serving of cookies; thus, the carrots would more likely provide the feeling of fullness with fewer total calories.

We recommended to our participants that they bring an "advocate" (parent, caregiver, case manager) to our initial teaching sessions and to monthly follow-up visits. We felt it was a good idea to have someone who could discuss our recommendations and help remember the dieting steps to follow.

An advocate also can act as a cheerleader. He or she can also help solve problems, like making sure that other people in the dieter's life know that the individual is trying to lose weight and what their diet consists of. An advocate might also help the dieter with using the materials we distributed. Among these materials was a food-tracking chart that the team designed. The chart could be used to keep a record of what foods and beverages were consumed each day. The chart was intended as a way to see how closely the dieter was following the diet.

We also conducted a food preference assessment that identified what foods and beverages were preferred by the dieter. The preference assessment formed the basis for individualizing the pictorial stoplight (SLD) materials. We taught our participants to eat and drink a lot of green light items, to use moderation with yellow light items, and avoid red light items unless they were part of a low-calorie entrée. We also suggested that our participants weigh frequently and to record their weight on a personal weight chart. We provided a weight-progress chart that became a graph when the participant circled his or her weight for each day or each time the participant weighed. We color coded weight ranges on the chart such that each color band encompassed the individual's weights by BMI. When our participants met their first weight-loss goal, as indicated by weights circled in the next lower BMI band, we offered a certificate of achievement. Some participants enjoyed adding additional Jayhawk (University of Kansas) stickers to their certificates as they met additional goals. We purchased the stickers at campus stores, but stickers of another kind could work as well.

In addition to the certificates and stickers, we provided bragging cards at our monthly meetings. Participants often asked for something in writing to show a parent, friend, caregiver or advocate who could not attend the meeting. We entered their current progress on the cards for them to take with them. We also offered the participants small amounts of cash or other prizes as incentives for keeping records of their diet and of their progress, but not specifically for weight loss. That is, record keeping was the focus of the cash rewards and not weight loss, per se.

Because exercise can be an important contribution to efforts to be healthy, we encouraged our participants to continue whatever form of regular exercise that they were already engaged in. We also offered a walking game board. If they wanted to try and increase their activity levels by walking more, the game board gave them a way to track how many steps they were walking. We provided a step counter (inexpensive pedometer) to each participant who wanted to use the game board.

As a partial guide when dining out, we also provided our participants with lists of menu items from area restaurants that were approximately 500 calories or less, as provided by the restaurants' websites.

At these meetings (held monthly) we learned many important facts about what the individuals and their caregivers knew and did not know about weight management. We also recorded many personal characteristics and lifestyle circumstances that became important in supporting them in their weight loss efforts and in informing us how to expand and alter how and what information to share with future participants.

We learned for example, that prior to our meetings, many participants and caregivers would not have been able to correctly label a number of food and beverage items as green, yellow or red. Orange juice (and other fruit juices) was a common example. Having been taught that orange juice was healthier, compared to sodas for example, its high calorie content was unknown or ignored. Bananas, potatoes, bread, cheese, and beans are other examples that participants did not identify has having higher calorie contents.

This phase covered 12 months. Results for the 66 participants who completed the study (10% loss to follow-up) demonstrated weight loss of 6.4%, and 8.7% from baseline at 6 and 12 months, respectively. These levels of weight loss are similar to the results from the successful Diabetes Prevention Program (Knowler et al., 2002) and exceed the 3% minimum weight loss suggested as clinically relevant in the American College of Sports Medicine Position Stand "Appropriate physical activity intervention strategies for weight loss and prevention of weight regain for adults," (Donnelly et al., 2009). These encouraging results led to the development of the third phase of our research—a trial designed to compare both weight loss and weight maintenance between the eSLD and a recommended care or conventional diet (CD) meal plan diet in adults with IDD.

Phase III

Briefly, 150 overweight/obese adults with mild to moderate IDD, living either in the community with parents/guardians, or in a supported living environment with a paid caregiver, who served as a "study partner," were randomized to either an eSLD or a conventional diet (CD) as part of a multicomponent weight management intervention (6 months weight loss, 12 months maintenance) (Ptomey et al., 2018). In addition to the energy reduced diet, participants were asked to increase physical activity (150 min/wk), self-monitor diet and physical activity, and attend behavioral strategy/educational sessions conducted by a health educator during monthly home visits.

Participants. IDD diagnoses were verified by the Community Service Provider operating in Kansas under the auspices of a Community Developmental Disability Organization (CDDO). Additionally, participants were required to be able to walk, and to communicate preferences (e.g., foods liked/disliked), wants (e.g., more to eat/drink), and needs (e.g., assistance with food preparation) through spoken or sign language, or via alterative communication systems such as voice output communication aids. To enhance the generalizability of our findings, individuals who used medications for prevalent conditions associated with obesity (e.g., hypertension, lipid abnormalities, type 2 diabetes) or other medications commonly prescribed for individuals with IDD, which may impact body weight, including medications to treat depression or other psychological/behavioral problems, were allowed to participate. Clearance from their primary care physician was required for all participants.

Informational brochures describing the project and containing study contact information were mailed/e-mailed to case managers, service providers and CDDOs in the recruitment area of 50 miles. Study staff also made in-person visits to these organizations to describe the trial and to solicit participation. A member of the investigative team, who was familiar with the sensitive issues regarding recruitment of individuals with IDD, contacted potential participants and/or caregivers to describe the trial in detail and determine initial eligibility. Written informed consent, approved by the Human Subjects Committee was obtained from either the participant (self as guardian) or a legal guardian, as well as the study partner. Participants were computer randomized, stratified by the number of adults with IDD in a residence (alone, 1–2, or 3–5), with equal allocation to the eSLD and CD groups. Multiple adults in a single residence volunteering to participate were assigned to the same intervention group. There were 24 residences with 2 participants and 9 residences with 3 participants.

Caregiver role. We recruited a parent/guardian, residential support staff, or an individual who assists with food shopping/meal preparation to serve as a study partner for each participant. Study partners were asked to attend all meetings between study staff and participants and to support, encourage, and assist participants in complying with the study protocol, including selection and preparation of foods consistent with the assigned diet, providing opportunities for increased physical activity, and assisting with self-monitoring of diet and physical activity.

Study partners received $50 gift cards at 6, 12 and 18-months as compensation for the additional burden associated with participation in this trial. Study partners who were unable to complete the trial were replaced.

Weight Loss: eSLD Components

- 2 portion-controlled entrées/day
- 2 portion-controlled shakes/day
- 5 one-cup servings of fruits and vegetables/day
- Ad libitum non-caloric beverages.
- Additional meals and snacks selected using the Stop Light diet guide.

A chart which included lists and pictures of common food items, color coded to the SLD system, was provided to assist with meal planning, food shopping and selection of appropriate snacks. As in Phase II (Saunders et al., 2011), the SLD was enhanced (eSLD) by encouraging consumption of high volume, low energy, portion-controlled meals (PCMs; entrées/shakes) and fruits and vegetables (Academy of Nutrition and Dietetics, 2011). PCMs are effective in controlling portion size and reducing both energy and fat intake and simplify meal planning, food shopping and meal preparation for individuals with IDD and their study partners. PCMs include nutritional information on the label (calories, fat, protein, etc.) making it easier to adhere to specific energy and nutrient recommendations. The simplicity of using PCMs may be especially relevant as caregivers (study partners) frequently have limited formal education specific to nutrition, and the turnover rate among paid caregivers is high (Humphries, Traci, & Seekins, 2008). Participants and study partners were provided with a list of entrées available from several manufacturers (Healthy Choice™, Smart Ones ™, Michelina's ™, etc.) which met our caloric, fat, and sodium requirements. They were asked to purchase entrées of their choice which are available at most grocery stores at a cost of $2.00–$4.00 each. Two low-energy, high-volume shakes/day (HMR Weight Management Services Corp) were provided at no cost to participants in the eSLD group during the 6-month weight loss phase. These shakes could be consumed as a snack or as a drink with a portion-controlled entrée. Participants desiring foods in addition to these recommendations were encouraged to select additional low energy (green) foods from the SLD chart.

Weight Loss: CD Components

- 500–700 kcal/day energy deficit
- Recommended servings of fruits, vegetables, grains, dairy, and protein.
- Instructions regarding serving sizes and food measurement.

Participants randomized to the CD group were taught to consume a nutritionally balanced, low-calorie, high-volume, lower fat (20–30% energy intake) diet following the MyPlate approach as recommended by the US Department of Agriculture (2013). Participants and study partners were taught to select, purchase and prepare appropriate foods, as well as to serve a portion of appropriate size to achieve the desired reduction in energy intake. During weight loss energy intake was reduced to ~500 to 700 kcal below daily estimated resting energy expenditure as estimated using the equation of Mifflin-St Jeor (Mifflin et al., 1990) multiplied by 1.4–1.6 to account for energy expenditure associated with physical activity. As this equation was not developed with individuals with IDD, the estimated energy intake was used as a starting point, which was adjusted in response to weight loss or gain as assessed during monthly home visits. Participants and study partners were provided with examples of meal plans, which suggested the number of servings of grains, proteins, fruits and vegetables, dairy, and fats to achieve the desire energy intake for weight loss, and were counseled on appropriate portion sizes using three-dimensional food models and taught to measure foods to facilitate compliance with serving size recommendations. Participants in the CD group received the monetary equivalent of the cost of the shakes provided to the eSLD group ($30.00/mo.) for the purchase of foods appropriate for CD only during the active weight loss intervention (0–6 months).

Weight Loss: Physical Activity

Participants in both groups were encouraged to accumulate a minimum of 30 min./day of moderate intensity physical activity 5 days/week (150 min/week). We are aware that this level of physical activity is less than recommended by current guidelines for weight management (Donnelly et al., 2009; Jensen et al., 2014). However, based on experience from our pilot trial in adults with IDD, and other reports in the literature demonstrating the low levels of physical activity in this group (Barnes et al., 2013), accumulating 150 min/week of moderate intensity physical activity represents a reasonable recommendation. Brisk walking was recommended as it is inexpensive, safe, fits easily into the daily routine, and can be performed alone or with others. Pedometers (Omron HJ-320, Lake Forest, IL) were provided to all participants as both a motivational tool and to self-monitor physical activity.

Education/Behavioral Sessions

All education/behavioral sessions were conducted during home visits with both participants and their study partner. The frequency and duration of these sessions was equal for both the eSLD and CD groups across the intervention. All sessions were led by health educators trained to deliver the intervention by shadowing a co-investigator experienced with weight management for adults with IDD. This co-investigator also supervised health educators during their initial session with participants and study partners, and attended a minimum of 4 monthly sessions, at random, to insure the intervention was delivered as intended using a checklist. Health educators were assigned to participant/study partners at baseline and delivered both the weight loss (0–6 months) and maintenance interventions (7–18 months) to those individuals.

Intervention

Orientation. Prior to initiating the intervention, participants and study partners completed a 60–90 min. Orientation session with their health educator designed to teach the basic principles of the diet and physical activity intervention, as well as describe the procedures and requirements for self-monitoring. A shorter follow-up home visit (~30 min) was completed ~2 weeks following the initial orientation to answer questions and address any issues that may have arisen as participants. During the home visit, medication information (name, frequency, and amount) was obtained and classified (yes/no) based on the association with weight gain (Domecq et al., 2015).

Diet. Both the eSLD and CD groups were taught basic principles regarding meal planning, food shopping, meal preparation and how to incorporate high volume/low energy foods in their diets, which have been shown to improve weight loss and maintenance (Rolls, 2005). Participants/study partners in the eSLD group were provided with a list of PCMs appropriate for this diet, a month's supply of low calorie shakes, and a color-coded chart (green, yellow, red) which categorized foods based on the Stop Light diet. Participants/study partners in the eSLD group were trained to use the chart to assist in meal planning, grocery shopping, decisions regarding snack foods, and compliance with the diet in special situations such as eating away from home (restaurants, parties, etc.). Participants/study partners in the CD group were taught how to select, prepare, and portion foods to achieve the required level of energy reduction.

Physical activity. The physical activity recommendations were explained to both intervention groups and instructions on how to wear and use the pedometer were provided. Participants were taught how to accumulate physical activity by incorporating activity into their daily routine (work breaks, lunchtime, etc.) and were provided with basic information regarding exercising in hot or cold weather, local sites for indoor walking, appropriate footwear, etc.

Self-monitoring. Participants, with assistance from their study partner, were trained to self-monitor their diet and physical activity across the intervention (0–18 months) using colorful pictorial 7-day tracking sheets specific to their group assignment (eSLD or CD). Participants in the eSLD group were asked to check the pictorial representations of the number of PCMs, shakes, fruits and vegetables, and other green, yellow or red foods consumed each day. Similarly, participants in the CD group were asked to check the number of daily servings of vegetables, fruits, dairy, meat and grains. The tracking sheets for both groups contained space for recording daily pedometer steps and minutes of non-walking physical activity. Participants in both groups were also provided with a pictorial game board to track their progress towards a goal of accumulating 100,000 steps that was associated with a monetary incentive ($5.00). Self-monitoring tracking sheets were collected and summarized by the health educator during monthly meetings and used to provide feedback to participants and study partners regarding compliance with the intervention.

Monthly meetings. Health educators conducted monthly 45–60 min. Home visit meetings with participants and their study partner across the 18-month trial. Participant's body weight was assessed wearing a t-shirt, shorts and without shoes, and weekly self-monitoring tracking sheets were reviewed to provide feedback regarding progress with weight loss and compliance with the study protocol. Health educators answered questions and taught problem solving/relapse prevention strategies to assist participants in maintaining compliance with the intervention. Twenty-four hour dietary recalls were also conducted by the health educator and used to provide feedback relative to compliance with the dietary recommendations. The 24-hour recalls followed the USA Department of Agriculture multiple pass protocol which includes five steps: (*1*) the quick list, where participants provide an uninterrupted listing of foods and beverages consumed; (*2*) the forgotten foods list, where participants are queried on categories of foods that have been documented to be frequently forgotten; (*3*) a time and occasion at which foods were consumed; (*4*) the detail cycle, which elicits descriptions of the specific foods and portions consumed aided by items designed to provide reference and improve recall accuracy such as three-dimensional food models, standard size glasses, mugs, bowls, chip bags, drink bottles, thickness sticks, measuring cups and spoons, and standardized geometric shapes (Wright et al., 2007), and (5) a final probe review (Blanton, Moshfegh, Baer, & Kretsch, 2006, Conway, Ingwersen, & Moshfegh, 2004, Conway et al., 2003). In addition, participants were provided with a simple chart for recording their weight on a frequent basis.

Incentives. In addition to the small monetary incentive for accumulating steps previously described, we gave participants small monetary incentives for recording consumption of foods consistent with the study diets (≤ $16.00/mo.), and for reducing their BMI by 1 kg/m^2 ($10.00). We gave incentives for steps walked, dietary compliance, and decreased BMI were provided during weight loss (0–6 months) but not during weight maintenance (7–18 months). However, during weight maintenance we provided $20.00 was for attendance at monthly health educator meetings. Participants had the potential to receive ~$560.00 in incentives across the 18-month intervention.

Weight maintenance. During weight maintenance (7–18 months) participants in the eSLD group were counseled to continue consumption of PCMs (2 entrées/shakes/day) and increase consumption of foods in the green and yellow categories of the Stop Light diet. Participants in the CD group were provided with energy intake recommendations for weight maintenance based on estimated resting energy expenditure (Mifflin et al., 1990) multiplied by 1.6–1.8 to account for physical activity, and provided with examples of meal plans consisting of suggested servings of grains, proteins, fruits and vegetables, dairy, and fats based on their maintenance energy requirements. Participants in both groups were asked to continue the 150 min/wk recommendation for physical activity. Participants who gained weight, based on weight measured during monthly home visits, were provided with suggestions for improving compliance with both dietary and physical activity recommendations for weight maintenance.

Results

Outcomes Analysis. This phase was powered to detect a difference of 2.6 kg in mean weight loss between the eSDL and CD groups at 6 months (a difference based on prior published data on results with the eSLD and CD diets). Height, weight and waist circumference were measured using generally used techniques in weight loss research. Dietary intake was measured with repeated administration of 3-day food records and analyses. Physical activity was measured not only from the records of steps walked produced by the participants, but also through 7-day accelerometer data. The analysis plan for data may be found in (Ptomey et al., 2018).

Participants. One hundred fifty adults with IDD were randomized to the eSLD (n = 78) or CD groups (n = 72). Body weight assessments were obtained on 67 (87%) and 57 (76%) participants following weight loss (6 months) and 54 (70%) and 47 (65%) participants following weight maintenance (18 months) for the eSLD and CD groups, respectively.

Baseline participant characteristics are presented. Participants were obese (BMI ~37 kg/m^2), middle-age (~36 years) adults with IDD; ~17% were diagnosed with Down Syndrome, while 57% were female. Approximately 16% of the sample was non-Caucasian, and ~82% had at least a high school education (as provided in Kansas special education programs). The majority of participants (~67%) lived in a group situation and received support from paid staff (~74%). Demographic characteristics and baseline measures were similar between the treatment groups.

Body Weight/BMI/Waist Circumference

Weight loss (0–6 months). Weight loss at 6 months was significantly greater in the eSLD (−6.8 ± 5.5 kg; −7.0%) compared with the CD group (−3.6 ± 5.3 kg; −3.8%). The proportion of participants achieving weight loss of >5% among participants completing the 6-month weight loss intervention was greater in the eSLD (62.3%) compared with the CD group (40.4%). None of the covariates of interest (sex, use of obesogenic medications, Down Syndrome, number of study partners, or their relationship with the participant) impacted change in weight at 6 months. Similar to the results for body weight, the decreases in both BMI and waist circumference were significantly greater in the eSLD compared with the CD group.

Weight maintenance (7–18 months). Weight increased from 7–18 months in the eSLD group (+0.3 ± 5.1 kg; 0.3%) and decreased in the CD group (−2.5 ± 8.4 kg; 2.2%); however, the between group difference was not statistically significant ($p = 0.052$).

Weight loss + maintenance (0–18 months). Weight loss at 18 months did not differ significantly between the eSLD (−6.4 ± 7.9 kg; 6.7%) and CD groups (−6.6 ± 9.2 kg; 6.4%, $p = 0.94$). The proportion of participants achieving ≥5% weight loss among participants completing the 18-month intervention was 57.4% and 49.0% in the eSLD and CD groups, respectively. None of the covariates of interest (sex, use of weight-inducing medications, Down Syndrome, number of study partners, or their relationship with the participant) impacted change in weight at 18 months. Similar to the results for body weight, there were no significant differences between groups for change in BMI ($p = 0.98$) or waist circumference ($p = 0.64$) across 18 months.

Intervention Compliance

Dietary recommendations. Participants randomized to the eSLD group were asked to consume 2 portion controlled entrées, 2 low-calorie shakes and 5 servings of fruits and vegetables each day. Self-reported compliance with these recommendations was poor. For example, 9% and 10% of participants complied with the entrée, and 17% and 0% complied with the shake recommendations at 6 and 18 months, respectively. Self-reported compliance with the fruit and vegetable recommendation was observed in 28% and 31% participants at 6 and 18 months. Participants randomized to the CD group were asked to consume a lower fat (<30% total energy intake), reduced energy (~500 kcal/day) diet which included 5 servings of fruits and vegetables per day. Self-reports indicated approximately 29% and 38% of participants in the CD group complied with the recommendation for lower fat intake at 6 and 18 months. Energy intake was reduced by 500 or more calories per day in approximately 29% and 26% of participants in the CD group at 6 and 18 months, as assessed by self-report. Self-reported compliance with the fruit and vegetable recommendation was achieved by 40% and 35% of participants at 6 and 18 months. There were no differences between the groups for changes in energy intake, % fat at intake, or servings of fruits or vegetable at 6 or 18 months.

Self-monitoring. Across the 18-month trial randomized participants completed 54% and 61% of requested monthly self-monitoring reports in the eSLD and CD groups, respectively. Completion of monthly reports was significantly higher in the CD (83%) compared with the eSLD group (70%, $p = 0.01$) among those actively participating in the trial.

Physical activity. Approximately 66%, 62% and 36% of the study sample provided valid accelerometer data at baseline, 6 months, and 18 months, respectively. Moderate-to-vigorous physical activity (MVPA) was generally low, ranging from ~ 15 min/day at baseline, to ~12 min/day at 18 months. There were no significant differences in MVPA between the eSLD and CD groups at baseline, 6 or 18 months. The proportion of participants meeting the MVPA goal of ≥150 min/wk across the 18-month trial was low (~8%) and did not differ significantly between the eSLD and CD groups at baseline, 6, or 18 months.

Discussion

Our results show that adults with IDD can lose significant amounts of weight and maintain that reduction over 18 months. Among the interesting aspects of our findings are that self-reported compliance with the diet and self-recorded data on physical activity did not predict the weight loss and maintenance observed outcomes. Apparently, our participants complied with sufficient components of the intervention to be successful dieters, but were poor at documentation. Another interesting result was that, while the eSLD diet produced the most weight loss initially, those on the CD plan eventually showed similar weight loss, with both diets leading to positive maintenance results. Overall, the results are encouraging for further efforts to design weight loss interventions that can be effective, and perhaps more effective, with adults with IDD than those without IDD.

Our participants received their diet and exercise training, and their monthly follow up meeting with the health educator, in their own homes or in their places of employment/day service. These visits were costly to the project, particularly when participants lived or worked 40–50 miles from the research offices. Many will question whether “taking the intervention to the participant” across the study duration was a major contributing factor in participant success. Certainly, the technical and practical details of each diet can be conveyed by phone, or possibly with some, by email.

Two practical considerations in choosing some form of remote communication and data collection may not be immediately obvious to the researcher. First, almost none of our participants across the pilot and the controlled clinical trial owned a scale. Prior to our work with them, they did not weigh at home, only at the office of a physician or other health care provider. Thus, reporting their monthly weight by telephone will require the provision of a scale. Second, many adults with IDD, and particularly those living alone, do not answer telephone calls from persons not on their cell-phone approved list. Many parents and staff provide an app that blocks non-identified callers. Initial monthly communications will in many cases need to be routed through the advocate.

There is more to this issue than the logistics of communication. Much of our planning for the single-arm pilot and the controlled clinical trial were based on direct observations of participants and their advocates and of their living and working conditions. While data can be acquired remotely, much may be missed that would be just as important in designing the next phase, and the one after that, and so forth. Health educators who visit the participants are able to see the particulars of the conditions in which each participant lives and works, which include the characteristics of their support staff, their home furnishings, their food storage capacities, and their recreation interests (e.g., TV, video games, sports). A specific and relevant example is observation of whether the participant owns a food blender—a device essential for making smoothies.

Another very important example is oral hygiene. Individuals who are endentulous or who are missing numerous teeth or who have pain-producing conditions, will likely have food texture preferences that will impact compliance with aspects of the diet. Certainly, in a practical application of our methods, without the restrictions on participant-to-participant consistency imposed by a research protocol, results of these observations would be important in gradually tailoring the intervention to individual differences.

Related questions arise from the differences in data collection in an in-home versus remote comparison. When our health educators met with participants, they collected the intake tracking forms, reviewed the data recorded by the participant with the participant and their advocate, and distributed new blank forms. With remote administration, it is unclear how the forms would be collected and replaced and it is unclear what would be lost in not reviewing the self-recorded data with the participant and advocate. Those reviews, in many cases, served to improve the understanding by the participant and advocate of what was to be recorded and how to record it. An example is that our tracking forms displayed pictures of carrots to represent "any vegetable" and pictures of apples to represent "any fruit." We learned early on that some participants were not markings these icons because they ate no carrots or apples. Self-recording of steps on the pedometers sometimes suffered in the following way: if the participant recorded 6321 steps yesterday, but forgot to reset the counter to zero, they simply recorded a new total the next day that reflected 6321 plus whatever was accumulated in the ensuing 24-hours. A more significant example of a discussion at monthly meetings, was that participants frequently needed reminders (despite our pictorial handouts) of what foods were red, yellow or green—particularly red items that they were hoping were green!

Other benefits of in-home or in-workplace meetings are that the health educator is able to get some sense of how the participant handles money. Some pocketed the money, some gave it to the advocate for safekeeping, and others seemed to "surrender" the money, less for safekeeping, than more as if it was required. Comments by participants regarding how they were going to spend their money were revealing as well. Some shared very realistic savings goals, such as a new purse, new hat, or new DVD. Others had surprisingly long-term goals, but goals confirmed by advocates, such a taking a cruise or other vacation. The in-person monthly interactions also provide insight on whether the small monetary incentives played much of a motivating role at all. The discovery of the rules at the riding stable is an example. One other observation regarding money deserves mention. Some of our participants lost 50–60 lbs., or upwards of 25% of their body weight. Consequently, their clothes no longer fit. Participants with families with the wherewithal to respond were delighted to purchase new clothes. Those in poverty needed and will need special attention.

When a health educator is visiting in the home, he or she is more likely to hear or be part of conversations about current and upcoming events in the home or at the workplace. Adults with IDD are like everyone else in their desire to have interesting, entertaining, and fun things to do. For many of our participants, Friday was bowling night, Saturday was dinner-out night, Sunday was lunch with mom and dad, Wednesday evening was the social at their church, and anyone's birthday, anniversary, or award was reason to party. In almost every case we encountered (not unexpectedly), these events involve food—usually, high-calorie food—pizza; cookies, cake, and ice cream; and all-you-can-eat buffets. It is a tough sell on the part of the health educator to discourage the all-you-can-eat pizza buffet for $3.99 per person or to ask the advocate to assist the participant in declining the snacks at the church social.

The health educator visiting the home also becomes aware of the effects of poverty on adults with IDD. As with nondisabled adults, the monthly food budget can be exhausted before the month ends. Typical of persons in this predicament is to survive off the most filling, but least costly foods and beverages available (e.g., sugary sodas) and, in response to receipt of the next government check, overeat. In any practical application of the diet methods we described, consideration of how to assist the participants to avoid this type of nutritional rollercoaster will be important.

Conclusion

When our team undertook to write this chapter, we assumed that some who read it, will do so to inform or guide further research. Others, we assumed, would do so to learn of practical supports for individuals with IDD needing to deal with weight problems. Our research projects were guided not just by scientific hypotheses, but also by the observations that preceded the pilot research and occurred in the pilot. Thus, we elected to share our overall experiences as a story, thru which we could share both data and anecdotes, and hopefully have satisfied both audiences.

References

Academy of Nutrition and Dietetics (2011) *How effective (in terms of client adherence and weight loss and maintenance) are meal replacements (liquid meals, meal bars, frozen prepackaged meals)*? Retrieved from http://www.adaevidencelibrary.com/evidence.cfm?evidence_summary_id=250141

Adolfsson, P., Sydner, Y. M., Fjellstrom, C., Lewin, B., & Andersson, A. (2008). Observed dietary intake in adults with intellectual disability living in the community. *Food & Nutrition Research, 52*. https://doi.org/10.3402/fnr.v52i0.1857

American Association on Intellectual and Developmental Disabilities (2012). *Definition of intellectual disabilities*. Retrieved from http://www.aaidd.org/content_100.cfm?navID=21.

Balogh, R. S., Lake, J. K., Lin, E., Wilton, A., & Lunsky, Y. (2015). Disparities in diabetes prevalence and preventable hospitalizations in people with intellectual and developmental disability: A population-based study. *Diabetic Medicine, 32*, 235–242. https://doi.org/10.1111/dme.12573

Bartlo, P., & Klein, P. J. (2011). Physical activity benefits and needs in adults with intellectual disabilities: Systematic review of the literature. *American Journal of Intellectual and Developemental Disabilities, 116*, 220–232. https://doi.org/10.1352/1944-7558-116.3.220

Bergstrom, H., Hagstromer, M., Hagberg, J., & Elinder, L. S. (2013). A multi-component universal intervention to improve diet and physical activity among adults with intellectual disabilities in community residences: A cluster randomised controlled trial. *Research in Developmental Disabilities, 34*, 3847–3857. https://doi.org/10.1016/j.ridd.2013.07.019

Blanton, C. A., Moshfegh, A. J., Baer, D. J., & Kretsch, M. J. (2006). The USDA automated multiple-pass method accurately estimates group total energy and nutrient intake. *Journal of Nutrition, 136*, 2594–2599.

Collins, C. E., Warren, J., Neve, M., McCoy, P., & Stokes, B. J. (2006). Measuring effectiveness of dietetic interventions in child obesity: A systematic review of randomized trials. *Archives of Pediatrics & Adolescent Medicine, 160*, 906–922. https://doi.org/10.1001/archpedi.160.9.906

Conway, J. M., Ingwersen, L. A., & Moshfegh, A. J. (2004). Accuracy of dietary recall using the USDA five-step multiple-pass method in men: An observational validation study. *Journal of the American Dietetic Association, 104*, 595–603. https://doi.org/10.1016/j.jada.2004.01.007

Damschroder, L. J., Lutes, L. D., Kirsh, S., Kim, H. M., Gillon, L., Holleman, R. G., … Richardson, C. R. (2014). Small-changes obesity treatment among veterans: 12-month outcomes. *American Journal of Preventive Medicine, 47*, 541–553. https://doi.org/10.1016/j.amepre.2014.06.016

De Winter, C. F., Bastiaanse, L. P., Hilgenkamp, T. I., Evenhuis, H. M., & Echteld, M. A. (2012). Overweight and obesity in older people with intellectual disability. *Research in Devevelopmental Disabilities, 33*, 398–405. https://doi.org/10.1016/j.ridd.2011.09.022

Domecq, J. P., Prutsky, G., Leppin, A., Sonbol, M. B., Altayar, O., Undavalli, C., … Murad, M. H. (2015). Clinical review: Drugs commonly associated with weight change: A systematic review and meta-analysis. *Journal of Clinical Endocrinology and Metabolism, 100*, 363–370. https://doi.org/10.1210/jc.2014-3421

Donnelly, J. E., Blair, S. N., Jakicic, J. M., Manore, M. M., Rankin, J. W., & Smith, B. K. (2009). Appropriate physical activity intervention strategies for weight loss and prevention of weight regain for adults. *Medicine & Science in Sports & Exercise, 41*, 459–471.

Draheim, C. C. (2006). Cardiovascular disease prevalence and risk factors of persons with mental retardation. *Mental Retardation and Developmental Disabilities Research Reviews, 12*, 3–12. https://doi.org/10.1002/mrdd.20095

Draheim, C. C., Stanish, H. I., Williams, D. P., & McCubbin, J. A. (2007). Dietary intake of adults with mental retardation who reside in community settings. *American Journal of Mental Retardation, 112*, 392–400. https://doi.org/10.1352/0895-8017(2007)112[0392:dioawm]2.0.co;2

Epstein, L., & Squires, S. (1988). *The stoplight diet for children: An eight-week program for parents and children*. Boston, MA: Little Brown & Company.

Farooqi, I. S., & O'Rahilly, S. (2005). New advances in the genetics of early onset obesity. *International Journal of Obesity (Lond), 29*, 1149–1152. https://doi.org/10.1038/sj.ijo.0803056

Hsieh, K., Rimmer, J. H., & Heller, T. (2013). Obesity and associated factors in adults with intellectual disability. *Journal of Intellectual Disabilities Research*. https://doi.org/10.1111/jir.12100

Humphries, K., Traci, M. A., & Seekins, T. (2008). Food on file: Pilot test of an innovative method for recording food intake of adults with intellectual disabilities living in the community. *Journal of Applied Research in Intellectual Disabilities, 21*, 126–173.

Jensen, M. D., Ryan, D. H., Apovian, C. M., Ard, J. D., Comuzzie, A. G., Donato, K. A., … Yanovski, S. Z. (2014). 2013 AHA/ACC/TOS guideline for the management of overweight and obesity in adults: a report of the American College of Cardiology/American Heart Association Task Force on Practice Guidelines and The Obesity Society. *Journal of the American College of Cardiology, 63*, 2985–3023. https://doi.org/10.1016/j.jacc.2013.11.004

Knowler, W. C., Barrett-Connor, E., Fowler, S. E., Hamman, R. F., Lachin, J. M., Walker, E. A., & Nathan, D. M. (2002). Reduction in the incidence of type 2 diabetes with lifestyle intervention or metformin. *The New England Journal of Medicine, 346*, 393–403.

Lauer, E., & McCallion, P. (2015). Mortality of people with intellectual and developmental disabilities from select US State disability service systems and medical claims data. *Journal of Applied Research in Intellectual Disabilities*. https://doi.org/10.1111/jar.12191

Martinez-Zaragoza, F., Campillo-Martinez, J. M., & Ato-Garcia, M. (2015). Effects on physical health of a multicomponent programme for overweight and obesity for adults with intellectual disabilities. *Journal of Applied Research in Intellectual Disabilities*. https://doi.org/10.1111/jar.12177

Melville, C. A., Boyle, S., Miller, S., Macmillan, S., Penpraze, V., Pert, C., … Hankey, C. R. (2011). An open study of the effectiveness of a multi-component weight-loss intervention for adults with intellectual disabilities and obesity. *British Journal of Nutrition, 105*, 1553–1562. https://doi.org/10.1017/s0007114510005362

Melville, C. A., Hamiltom, S., Hankey, c. R., Miller, S. L., & Boyle, S. (2007). The prevalence and determinants of obesity in adults with intellectual disabilities. *Obesity Reviews, 8*, 223–230.

Mifflin, M. D., St Jeor, S. T., Hill, L. A., Scott, B. J., Daugherty, S. A., & Koh, Y. O. (1990). A new predictive equation for resting energy expenditure in healthy individuals. *American Journal of Clinical Nutrition, 51*, 241–247.

Pett, M., Clark, L., Eldredge, A., Cardell, B., Jordan, K., Chambless, C., & Burley, J. (2013). Effecting healthy lifestyle changes in overweight and obese young adults with intellectual disability. *American Journal of Intellectual and Developmental Disabilities, 118*, 224–243. https://doi.org/10.1352/1944-7558-118.3.224

Ptomey, L. T., Saunders, R. R., Saunders, M., Washburn, R. A., Mayo, M. S., Sullivan, D. K., … Donnelly, J. E. (2018). Weight management in adults with intellectual and developmental disabilities: A randomized controlled trial of two dietary approaches. *Journal of Applied Research in Intellectual Disabilities, 31*(Suppl 1), 82–96. https://doi.org/10.1111/jar.12348. Epub 2017 Mar 23.

Ptomey, L. T., Sullivan, D. K., Lee, J., Goetz, J. R., Gibson, C., & Donnelly, J. E. (2015). The use of technology for delivering a weight loss program for adolescents with intellectual and developmental disabilities. *Journal of the Academy of Nutrition and Dietetics, 115*, 112–118. https://doi.org/10.1016/j.jand.2014.08.031

Ptomey, L. T., & Wittenbrook, W. (2015). Position of the academy of nutrition and dietetics: nutrition services for individuals with intellectual and developmental disabilities and special health care needs. *Journal of the Academy of Nutrition and Dietetics, 115*, 593–608. https://doi.org/10.1016/j.jand.2015.02.002

Reichard, A., Saunders, M. D., Saunders, R. R., Donnelly, J. E., Lauer, E., Sullivan, D. K., & Ptomey, L. (2015). A comparison of two weight management programs for adults with mobility impairments. *Disability and Health Journal, 8*, 61–69. https://doi.org/10.1016/j.dhjo.2014.08.002

Reichard, A., Stolzle, H., & Fox, M. H. (2011). Health disparities among adults with physical disabilities or cognitive limitations compared to individuals with no disabilities in the United States. *Disability and Health Journal, 4*, 59–67. https://doi.org/10.1016/j.dhjo.2010.05.003

Robertson, J., Emerson, E., Baines, S., & Hatton, C. (2014). Obesity and health behaviours of British adults with self-reported intellectual impairments: Cross sectional survey. *BMC Public Health, 14*, 219. https://doi.org/10.1186/1471-2458-14-219

Rolls, B. J. (2005). *The volumetrics eating plan*. New York: Harper Collins Publishers.

Saunders, R. R., Saunders, M. D., Donnelly, J. E., Smith, B. K., Sullivan, D. K., Guilford, B., & Rondon, M. F. (2011). Evaluation of an approach to weight loss in adults with intellectual or developmental disabilities. *American Journal of Intellectual and Developmental Disabilities, 49*, 103–112.

Shireman, T. I., Reichard, A., Nazir, N., Backes, J. M., & Greiner, K. A. (2010). Quality of diabetes care for adults with developmental disabilities. *Disability and Health Journal, 3*, 179–185. https://doi.org/10.1016/j.dhjo.2009.10.004

Spanos, D., Melville, C. A., & Hankey, C. R. (2013). Weight management interventions in adults with intellectual disabilities and obesity: A systematic review of the evidence. *Nutrition Journal, 12*, 132. https://doi.org/10.1186/1475-2891-12-132

Stancliffe, R. J., Lakin, K. C., Larson, S., Engler, J., Bershadsky, J., Taub, S., … Ticha, R. (2011). Overweight and obesity among adults with intellectual disabilities who use intellectual disability/developmental disability services in 20 U.S. States. *American Journal oj Intellectual and Developmental Disabilities, 116*, 401–418. https://doi.org/10.1352/1944-7558-116.6.401

Sundahl, L., Zetterberg, M., Wester, A., Rehn, B., & Blomqvist, S. (2015). Physical activity levels among adolescent and young adult women and men with and without intellectual disability. *Journal of Applied Research in Intellectual Disabilities*. https://doi.org/10.1111/jar.12170

US Department of Agriculture. (2013). *Choose MyPlate*. Washington, DC: USDA.

Wright, J. D., Borrud, L. G., McDowell, M. A., Wang, C. Y., Radimer, K., & Johnson, C. L. (2007). Nutrition assessment in the National Health and Nutrition Examination Survey 1999–2002. *Journal of the American Dietetic Association, 107*, 822–829. https://doi.org/10.1016/j.jada.2007.02.017

57

Incontinence

Johnny L. Matson and Abigail Issarraras

Introduction

Incontinence is a debilitating and common problem among persons with intellectual disability (ID). Typically, this larger problem is broken into four subtypes: diurnal and nocturnal enuresis, and diurnal and nocturnal encopresis. Not only are those problems frequent, but they are more common among persons with ID than they are in the general population (von Gontard, 2013). If not treated, these problems may persist into adulthood. They restrict access to activities, increase the likelihood of infections, and are associated with comorbid psychological problems such as attention deficit hyperactivity disorder (ADHD), anxiety, and depression (von Gontard, 2013).

Several factors put the person at greater risk for problems of incontinence. Hellström, Hanson, Hansson, Hjälmås, and Jodal (1990), for example, studied the toileting habits of 3556 7-year-old children in Sweden. They found that daytime incontinence was more common in girls while nocturnal incontinence was more common in boys. Other factors that appear to be established risk factors include parental history of toileting problems, recent emotional stress, urinary infections, and a history of bedwetting among male siblings (Sureshkumar, Craig, Roy, & Knight, 2000).

The notion that establishing risk factors is important is not new, although early studies tended to be case studies versus more methodologically controlled reports. A case in point is a paper by Bert Beverly published in 1933. He reports on approximately 250 cases of mostly nocturnal enuresis. It is noted that 64% of the cases were boys. He posits that mental health concerns may contribute to incontinence. For example, Beverly (1933) observed that the children in his study were often nervous, fearful, experiencing "infantile habits" such as thumbsucking, nail biting, and frequent crying. He also speculated that jealousy of a new sibling may also contribute to incontinence. The author also states that incontinence can stop as soon as incentives are introduced. Many of these assumptions have proven to be incorrect over time, but not all of them. Certainly, incentives (read reinforcement) have proven effective, but more structure and various learning theory based strategies above and beyond reinforcement are also required in most cases.

The intensity of the intervention in general, increases as the level of cognitive disability increases. As Böhmer, Taminiau, Klinkenberg-Knol, and Meuwissen (2001) point out, for a sample of 215 people with ID who were hospitalized,

---

J. L. Matson · A. Issarraras (✉)
Department of Psychology, Louisiana State University, Baton Rouge, LA, USA
e-mail: aissar1@lsu.edu

J. L. Matson (ed.), *Handbook of Intellectual Disabilities*, Autism and Child Psychopathology Series, https://doi.org/10.1007/978-3-030-20843-1_57

laxatives for constipation were frequently prescribed with poor results. Developing effective psychological interventions for constipation is quite important, as a result, since they found that 69% of their sample evinced this problem.

Interventions based on operant conditioning dates to at least the early 1960s. Ellis (1963) employed a stimulus-response model. Bladder extension and approach to a toilet were reinforced in a person with ID living in a developmental center in Louisiana (USA). The shaping of this response eventually led to reinforcement of voiding in the toilet. Ellis (1963) also attempted to establish times when the child was likely to void. He prompted toileting at these times. This approach has been referred to as a "timing method."

Dayan (1964) who was the Director of Psychology at the Sunland Training Center in Mariana, Florida, described the implementation of Ellis' toilet training model at the Florida center. The residents treated were 6–12 years of age with IQs of 30 and below. He identified 25 children as "soilers" or "wetters" or both. Children were placed on the toilet every 2 h. If the child voided while on the toilet they were reinforced, but they were ignored if they did not void. Dayan (1964) reported success as measured by a marked decrease in clothing that needed to be washed.

Several early treatment studies of incontinence for people with ID used Ellis' model. One of these studies was published by Waye and Melnyr (1973). They treated a 15-year-old male who was blind and who had profound ID. According to the authors, he only had one or two words. He was placed on the toilet for 5 min from every 15 min while he was actively mobile and walking around the institutional ward. Time was taken off his training for meals. Reinforcement for voiding included rubbing his head and ears and saying the child's name. Marked improvements were noted as measured by fewer dirtied clothes and less time sitting on the stool before voiding. Improvements were still evident at a 1-year follow-up. These early successes spurred additional study and development.

Assessment

Assessment generally falls into two broad categories. One involves frequency counts of voiding, time on the toilet before voiding (i.e., less time is better), and counting soiled clothes. Fluid intake and volume of voiding have also been used as outcome measures (Hoebeke, Bower, Combs, De Jong, & Yang, 2010). A second type of assessment used less frequently is norm-based scales. These measures are used to survey populations or individuals to determine the particular toileting problems that they experience. An example of this type of measure is the Profile of Toileting Issues (POTI; Matson, Neal, Hess, & Kozlowski, 2011). The scale was specifically developed to assess/screen for toileting problems in persons with ID. Their sample consisted of 108 people with ID who were 16–89 years of age. Participants ranged from mild to profound ID. The scale is a paper and pencil informant test with 56 items. Items are scored as either No Problem or Problem Present or Does Not Apply. Typical items include lost toileting skills, has daytime toileting accidents, hides soiled clothes, has blood in stool, and has hard stools. The test has good reliability and validity.

In another study using the POTI, Matson, Horovitz, and Sipes (2011) found that there were several very frequently endorsed items. These problems included has toileting accidents during the day, has toileting accidents at night, has had wet underwear in the past month, and is on a medication with a known side effect of constipation. One major advantage of a scale like this is to educate staff on a standardized set of problems that need to be addressed. Also, a screener of this sort helps identify problems of incontinence that require treatment.

General Treatment Issues

One of the first issues that need to be addressed is medical concerns. In some cases, particularly where severe physical and/or cognitive disabilities are present, voiding may be difficult to train (e.g., spina bifida). Also, cognitive and/or sensory limitations may make it difficult to teach the requisite skills. Secondary to these concerns are medical problems caused by the toileting problems. Impactions can cause distress or severe damage to the bowel and may necessitate the use of medications that may have side effects and which can become habit forming. Urinary tract infections also occur in approximately 10% of children with urinary incontinence (Loening-Baucke, 1997). These problems are more common in girls than boys.

Psychological Treatment

A number of psychological based issues have been noted as well. Effective treatments typically involve operant and classical conditioning. As a result, these methods are often labor-intensive. Levine and Elliot (1970) address the issue of toilet training persons with profound ID, where limited staff are available. They worked with 103 people ranging in age from 4–48 years old. Their focus was on providing general guidance on the principles of applied behavioral analysis (ABA) during a 10-hour workshop. Based on this introduction to the principles of learning, the authors reported a marked decrease in accidents and in the amount of soiled linen.

Baumeister and Klosowski (1965) took a somewhat different approach. They focused on a group approach to toilet training for persons with severe ID who lived in a developmental center. The individuals treated could not feed themselves, could not speak, and in general, were very limited with respect to their adaptive behavior. The clients who were treated were 10–25 years of age, and all of them could walk. The methods of treatment described previously were used and again proved to be effective.

Equipment

The most famous and enduring technical advance for incontinence is the bell and pad program devised by Mowrer and Mowrer (1938). Sensors are connected when the person wets the bed, causing a buzzer to sound. Over time the person is conditioned to awake before voiding. This method has been used extensively, primarily for persons with typical cognitive development. However, Azrin, Bugle, and O'Brien (1971) developed a daytime variant for persons with ID. The device was named the "pants alarm." These authors trained four persons with profound ID. Two snaps are attached to the crotch of the underwear and to a circuit box that is worn at the waist on a belt. A toilet with a sensor affixed to it that sounded when urinating in the toilet was also used. Drinks were provided every 30 min, and each child was placed on the toilet at 30 min intervals. Voiding in the toilet resulted in praise, hugs, and candy. Urinating in the underpants resulted in one spank to the buttocks, followed by a 10 min time-out. The authors report success with the method.

Chang, Lee, Chou, Chen, and Chen (2011) also describe a mobile system to detect voiding. In their paper, a 9-year-old boy with multiple disabilities was trained. They also used a wetness sensor. This technology was paired with reinforcement for appropriate voiding in the toilet versus his pants.

Client Characteristics

A series of studies have also been published looking at factors associated with client risk-factors. Smith, Britton, Johnson, and Thomas (1975) specifically address the issue of adults with profound ID. As they rightly point out, most research and clinical focus is and has been on children, a factor which is still largely existent in current practice. They suggest that these problems with incontinence may also be very common in adults, particularly among persons who have severe deficits in cognitive functioning. Ambulation can also be an issue, especially for persons diagnosed with severe or profound ID. Additionally, these persons may have difficulty following instructions. Therefore, considerable structure and routine are necessary.

Lancioni (1980) also underscores that point in toilet training persons with profound ID. He notes that physical handicaps are also common in this population, and the focus of treatment therefore is often on prevention rather than the promotion of independent toileting. In his study, nine children and adolescents between 8–16 years of age who had profound ID, and were deaf and blind were trained. They used potty-alert and pants alarm devices to help staff detect accidents. He defined toileting behaviors as independent toileting, partially independent toileting, incomplete toileting, attempted voiding, assisted voiding, and accidents. Water and soft drinks were provided frequently to increase voiding trials. Participants were seated close to the bathroom door so they could touch it. Urination attempts were prompted every 20 min. Edible reinforcers were provided for self-initiations. Over time and with increased success, the chairs participants sat in were moved further from the bathroom. Accidents were followed by cleaning up the chair and floor right around it. The author reported good success using this method. The take away with respect to persons with ID is that modifications to assessment and treatment are likely, and the therapist must be prepared to work with adults as well as children. This point is underscored by Smith and Smith (1977). They trained 13 institutionalized persons with ID over a range of ages. They concluded that children in their sample could be trained much faster than adults over the age of 25 years. Smith and Smith (1977) note that younger children had an 80% decrease in toileting accidents in 5–6 weeks while older more cognitively impaired persons required 8–9 weeks of training to acquire similar improvements. Along these same lines, Dalrymple and Ruble (1992) noted that lower cognitive and by extension verbal skills correlated with the achievement of continence at later ages. These factors point to Bettison's (1986) hypotheses that incontinence is largely due to difficulty learning.

Training Issues

The bulk of toilet training with persons with ID has involved professional staff, primarily in the field of psychology. However, for efficiency and to promote generalization, parents have also been included as trainers in some cases. Ozcan and Cavkaytar (2009) describe one such attempt. They note that the parent may play several roles including a parenting role, but also they can serve as an advocate and as a teacher. They also point out that parents may be particularly helpful in eliminating incontinence since they spend more time with their children than any other adult. These authors also suggest that including parents in training can be very valuable in promoting generalization, and ensuring the lasting effects of treatment.

Feldman, Ducharme, and Case (1999) also note that for families who have children with ID are more likely to be neglected. These authors state that researchers show that parents are particularly responsive to training packages. Feldman et al. (1999) report that common characteristics of these training models include reinforcement, feedback, modeling, pictures used as prompts, and standardized instructions. In their study, a picture-based training manual was used to train basic skills in changing diapers, treating diaper rash, how to give their child a bath, and personal safety tips. They note that skills were largely maintained at a 3-year follow-up.

Treatment for Enuresis

Parent involvement in training has also been emphasized by Rinald and Mirenda (2012). In their paper they describe a training workshop for six parents of developmentally disabled children. A PowerPoint presentation was followed by role play practice with a mannequin and supervised by the workshop leader. Parents then implemented the behaviorally based treatment at home for 5–8 days with phone support from professionals. Reinforcement of appropriate voiding, increased fluid intake, scheduled voiding attempts, redirecting the child when accidents occurred, and specific strategies to increase generalization and maintenance proved to be effective.

Dalrymple and Ruble (1992) also addressed parents in their survey of toileting habits of children with autism. As the reader is likely aware, upwards of 70% of persons with autism also experience ID, thus making this population in large part, compatible with a general ID population with respect to issues in toilet training. Delays in training were commonly reported. For example, 20% of parents needed more than 3 months to establish successful training, despite the fact that training was started later than is the case for typically developing children. The parents also reported a regression in skills for 30% of the 100 children who were trained. Most parents used positive reinforcement while about 1/3 resorted to punishments such as scolding. Risk factors for establishing appropriate toileting included ID and lack of verbal skills. Sensory impairments are also a major risk factor. Luiselli (1994) notes that few studies have focused on ID and sensory impairments. In his paper using a toileting schedule, reinforcement and prompting were employed with children in a residential setting who had these deficits.

Mahoney, Wagenen, and Meyerson (1971) in another early toilet training study taught three typically developing children and five children with ID. Children in this experiment had to be ambulatory, transportation was required, and the families had to agree to participate for 8 weeks. Only one of the children with ID had speech despite the fact that they ranged in age from 4 to 9 years old. These authors used a pants alarm which buzzed when a toileting accident occurred. Parents were asked to not feed or provide drink for an hour before training since food and praise were primary reinforcers in the treatment. Using verbal and physical prompts plus reinforcement, the children were taught to walk to the toilet when the pants alarm went off. Other target behaviors in the sequence included lowering their pants, sitting (or in the case of boys, standing) near the toilet, voiding, and then replacing clothing. Gradually fading out prompts including the pants alarm occurred as success in toileting skills increased. These methods proved to be successful.

Many of these studies aimed at curbing incontinence in persons with ID use a similar set of psychologically based principles. Sells-Love, Rinaldi, and McLaughlin (2002) describe a treatment study with a 19-year-old female carried out in a special education classroom. The program was teacher implemented and resulted in a decrease in the frequency of wet pants. Hyams, McCoull, Smith, and Tyrer (1992) did a 10 year follow-up of 14 persons with severe or profound ID who were trained using operant methods. They note that complete independent training was not maintained. However, they report that maintenance was better for persons who received intensive individual training versus group training.

Taylor, Cipani, and Clardy (1994) describe the training of an incontinent 10-year-old with autism and severe ID. In their study a modified version of the very popular Azrin and Foxx rapid toileting procedure was used (Azrin & Foxx, 1971). The method involved massed practice using operant conditioning and modeling. A time schedule for prompting attempts to void in the stool was followed by edible reinforcers and praised for successful voiding. In their initial study, nine adult males with profound ID were trained. In addition to the methods of applied behavior analysis first described, a pants alarm was employed. When accidents occurred, the clients were to have the alarm deactivated by the trainer. Next, the resident walked to the laundry room to get clean clothes. Then they changed their clothes. A one-hour time out then followed. No fluids were ingested within 30 min, and the toilet schedule of sitting on the stool every 30 min continued.

For the present study, the primary modification to the Azrin and Foxx (1971) method was the removal of the child's pants 5 min after being given liquids. Pants remained off until a correct voiding episode occurred. Each successful elimination resulted in the addition of 1 min to the pants-on period. A modified Azrin and Foxx treatment was also described by Chung (2007). This author treated a 12-year-old male with ID. The main changes included shortened training from eight to three hours daily. Secondly, the overcorrection procedure where the child was required to clean up after accidents was dropped.

Didden, Sikkema, Bosman, Duker, and Curfs (2001) also used a modified Azrin and Foxx procedure. They trained six children with a specific genetic form of ID, Angelman syndrome. Not only did they obtain good effects, the effects also were largely still maintained 2½ years later.

Continuing the theme of successfully using operant methods to train continence in children with ID, Smith, Smith, and Kwok Yi Lee (2000) describe three cases where training including shaping by getting closer and closer to the toilet and reinforcing appropriate responses. The kids had ID and were between the ages of 8–12 years old. Two children had mild ID (one person had fragile X syndrome) and one child had moderate ID. These children were described as having been treatment resistant. To reach the criteria for continence, the time required was 51 days, 54 weeks, and 130 weeks respectively; this is much longer than what has been reported in most toilet training studies employing persons with ID.

Luiselli (1996) used a stimulus control procedure to teach toileting skills to a 7-year-old girl with pervasive developmental disorder-not otherwise specified (PDD-NOS). She was prompted to sit on the toilet and was reinforced. The program took about 2 weeks and proved to be successful.

A somewhat different approach was described by Hagopian, Fisher, Piazza, and Wierzbicki (1993). They treated a 9-year-old boy with profound ID. Major components of treatment included social praise and edibles for appropriate voiding and limited attention while changing clothes after an accident. The latter procedure includes no verbal interaction and minimal physical interaction. An addition twist to the intervention was described as water prompting. This method involved gradually pouring about five ounces of lukewarm water over the child's private areas while seated on the toilet. Also, the child engaged in self-injurious behavior (SIB) during the day long training (most likely with an escape function). Blocking SIB attempts and reinforcing intervals where SIB was absent was the treatment for this challenging behavior. This treatment proved to be effective.

Kroeger and Sorensen-Burnworth (2009) underscore the popularity of the Azrin and Foxx (1971) toileting method. In this review, the authors note that this approach is the most commonly studied procedure among persons with ID, autism, and other developmental disabilities.

Bettison, Davidson, Taylor, and Fox (1976) describe a procedure involving changing and mild punishment to produce an effective intervention for daytime enuresis training. Training components such as praise for voiding in the toilet are described for persons with ID who ranged in age from 7 to 50 years of age. Before the advent of the Azrin and Foxx (1971) approach, behavioral methods used fewer components and more punishment. Even within the original Azrin and Foxx (1971) method, the mild punishment in the form of overcorrection was dropped in later studies. Also, as the field has advanced, guidelines regarding what procedures are best at various stages of development have been addressed.

Some aspects of early training methods have continued to be popular. For example, auditory signals as a discriminative stimulus, practicing steps of the voiding process, prompting to the toilet, and developing skills regarding pants down and pants up (Van Wagenen, Meyerson, Kerr, & Mahoney, 1969). Another of these skills which has been commonly stressed for decades is the focus on adequate fluid intake, which of course is linked to successful trials of voiding (Van Laecke, Raes, Vande Walle, & Hoebeke, 2009).

More recent efforts at toilet training have focused on modeling, and video modeling in particular (Keen, Brannigan, & Cuskelly, 2007). In their study, five boys with autism were trained on daytime urinating. Video modeling was paired with shaping and reinforcement strategies. Children who received modeling had more significant gains versus those children who did not.

Nocturnal enuresis has also been a source of attention in the experimental literature. Smith (1981) describes the training of five adolescents and young adults between 15–21 years of age. All of the participants were diagnosed with severe or profound ID. Three of these people had extreme hyperactivity, verbal and physical aggression, and temper tantrums, which of course complicated the learning process. Trainers were two nurses who worked the night shift. After a 2 week baseline, bedding was checked hourly to determine if it was wet. If wet, the sleeper was awakened and clothing was changed before going back to sleep. During the intervention and alarm system was used to alert the client and staff to a voiding episode. When the alarm sounded, the nurse would wake up the trainee, if the alarm had not been sufficient. The nurse then encourages the client to sit on the toilet by the bed and try to urinate. The trainee was asked to feel the dry sheets as a means of differentiating wet from dry. The wet items were then placed in the dirty laundry. Being dry for the entire evening was followed by a range of reinforcers including extended bedtime, tea, biscuits, access to a juke box, tokens, and trendy clothes. These rewards were not available if the trainee had wet the bed. Length of training ranged from 20 to 93 weeks before continence was achieved. Gains were maintained at a 2-year follow-up.

Medical Treatments for Encopresis

Encopresis has also been treated, in this instance with a general population of children (over 100) at the University of Oklahoma School of Medicine (Wright & Walker, 1978). The main point here is the psychologically based treatments for this problem have been available for some time. Also, many of the methods and procedures are similar to what has been described for the treatment of enuresis. The steps of their procedure began with a physical exam. We would note that this step is certainly warranted given the physical issues such as impactions, that can results and which can cause damage to the intestinal tract. However, for enuresis medical complications are far less likely. Thus, this step may not be necessary unless infection, pain in urinating, or other complications are present.

The second step of the procedure described by Wright and Walker (1978) involved an interview with the parents. The main goal here was to rule out potential psychological disturbance, ID, or behavior problems that may supersede toileting issues. Also, these issues would of course complicate the training process. Step three involved explaining the training process to the parents, and, where applicable, to the child. Step four involved emptying the colon, usually with an enema the night before the program was initiated. Of course, for enuresis this would not be done. The child was then to go to the restroom first thing in the morning. The child sits on the stool, and if they are able to void feces, social and tangible reinforcers are given.

If voiding does not occur, the parent inserts a glycerin suppository and then dresses the child. Here we would note that current thinking would most likely delete enemas and suppositories unless an impaction was present or the child had not passed feces for two or three days with the other training components in place. Additionally, close supervision by the pediatrician would be in order.

These authors note that by the completion of breakfast the suppository should have taken effect. At that point, the child could be taken back to the bathroom for a second voiding attempt. Successful voiding would be followed by verbal and tangible reinforcers. However, the quantity would be less than in instances where successful voiding occurred on the first attempt. If a third attempt was needed before school, a second enema would be used to prompt toileting. As a personal observation, if a procedure like this were to be used, it would be advisable to start the treatment on a Saturday when practical, so that the two days of training would be possible without school issues.

The authors suggest that successful voiding should be possible with only occasional enemas after the first few days. After school the child is checked to determine if any soiling had occurred. When the child was clean, they earned a reward. If the child was soiled, a mild punisher was used. Prompting to the toilet and the use of reinforcement and punishment are of course hallmarks for the bulk of the enuresis programs. Common reinforcers these authors endorse include money, candy, periods of attention from the parents, small toys, and extra privileges. Commonly used punishers they endorse are restricted TV viewing (or possible phone/computer use), monetary fines, extra chores around the house, and loss of privileges.

Lowery, Srour, Whitehead, and Schuster (1985) in another larger study of encopresis treated 58 children for encopresis. After meals, children were prompted to void in the toilet. If defecation did not occur during a 2-day period, enemas were used to prompt defecation. This intervention obviously had a more medical focus than many of the treatments described previously. However, the authors did note considerable gains for most children with respect to continence.

Psychological Treatments for Encopresis

A brief review of psychologically based treatment for encopresis follows next. Most of these interventions pair applied behavior analysis with timed/routine scheduled toileting attempts. In one study that diverged from this model reported by Carpenter (1989), a 22-year-old man with Prader–Willi syndrome was treated. In this case study, the author reported using group therapy, play therapy, family therapy, and medication. This approach cannot be endorsed here due to the lack of controlled data. However, it is worth mentioning to demonstrate the therapeutic approach to encopresis is not a monolith.

A more commonly used approach incorporating applied behavior analysis is described by Doleys and Arnold (1975). They treated an 8-year-old boy using the full cleanliness training model, where the child cleaned up all aspects of the soiling accident. This procedure was paired with social and tangible reinforcement for appropriate toileting. The authors report good success, although inconsistencies by the parents resulted in some regression at the follow-up.

As noted, most of the treatments that have been described in the literature on encopresis have focused on various aspects of learning theory. Piazza, Fisher, Chinn, and Bowman (1991), for example, describe a two-step procedure consisting of increasing bowel movements and discrimination training. This study used observations of possible signs of defecation and pant checks. If bowel movements occurred in the stool and when the person was clean at pant checks, social praise and preferred objects were used as reinforcers. The two trainees were a 5-year-old girl and 15-year-old male, both of whom were diagnosed with profound ID. They were taken to the toilet at regular intervals and prompted to void. After 7 weeks, 100% continence had been achieved for both clients.

Another learning based treatment involved prompting to sit on the stool after meals, which was described by Smith, Franchetti, McCoull, Pattison, and Pickstock (1994). The four participants diagnosed with severe ID trained were also given an increase of fiber in their diet when treatment began. These authors report that the trainees all closely approach the goal of normal toileting.

Encopresis is a problem of considerable magnitude in the ID population. Huntley and Smith (1999) make this point and go on to describe a study they did on the topic. They describe follow-up on 9 people with severe ID who had received behavioral treatment, similar to what has already been described for encopresis. These authors report marked improvement in 8 of the 9 cases with 6 people being relatively accident free.

Conclusions

After more than five decades of development, both clinicians and researchers have together arrived at several fundamentals regarding treatment of incontinence among persons with ID. First, the problem is common in this population and may extend into adulthood if not properly addressed through intensive treatment. Second, this set of problems can produce a great deal of stress and can be a major impediment to independent living (Matson & LoVullo, 2009). Third, comorbid behavior problems such as noncompliance, aggression, and temper tantrums are common. Problems of this type result in the need to modify how toileting issues are assessed, and inevitably impact the treatment planning to address such toileting issues. Fourth, people who have the most serious cognitive deficits are likely to require more intensive interventions over larger periods of time to produce meaningful effects.

The fifth point, and perhaps the most important, is that treatments are generally effective for both enuresis (diurnal and nocturnal) and encopresis (Lancioni, O'Reilly, & Basili, 2001). A host of different treatment elements have been described in the literature including psychotherapy. Having said that, most studies employ several common elements, including whether the enuresis or encopresis occurs during the day or at night. Scheduled times for toileting attempts are common and are often paired with prompts to the toilet by a caregiver be it a professional staff member or a family member. Next, prompts are also used to get the persons to undress and sit on the toilet. When voiding occurs, social and tangible reinforcers are given, at least during initial successful attempts. Also, what we will call dry/clean awareness is a frequent aspect of the treatment. This has been accomplished with a bell and pad alarm at night or pants alarm during the daytime. Another method, which avoids these devices, is to simply have the trainee feel clothing or sheets to determine whether or not they are clean and dry. Modeling is another component which has been incorporated in this model. This can be done with a doll or via videotape. Other components frequently used include gradually fading (i.e., decreasing) the number of prompts, scheduled visits to the toilet, and reinforcement as the trainee becomes more proficient in independent toileting. Early studies used some mild forms of punishment; punishment procedures have included the trainee being involved in cleaning up when accidents occur, a brief (e.g., 5 min) loss of attention, or a designated period without receiving preferred tangible reinforcers. More recently, most researchers have dropped aversive aspects of treatment. Without these punishment procedures, treatments still appear to be very effective.

Moving forward, an important aspect of treatment should be doing as much as possible to avoid “reinventing the flat tire.” Cost-effective, positively reviewed interventions that can be implemented in days or a few weeks are available. However, with each new generation, the potential for at least some of this information being lost is always possible. One method to counter this has been the large number of “How-To” books that have been written and which have largely targeted parents. Incorporating this information into college and high school classes on psychology and education would seem to be another potential means of disseminating this information. No doubt, technology for treating incontinence in persons with ID has been one of the bigger success stories in the field over the last century.

References

Azrin, N. H., Bugle, C., & O’Brien, F. (1971). Behavioral engineering: Two apparatuses for toilet training retarded children. *Journal of Applied Behavior Analysis, 4*(3), 249–253.

Azrin, N. H., & Foxx, R. M. (1971). A rapid method of toilet training the institutionalized retarded. *Journal of Applied Behavior Analysis, 4*(2), 89–99.

Baumeister, A., & Klosowski, R. (1965). An attempt to group toilet train severely retarded patients. *Mental Retardation, 3*(6), 24–26.

Bettison, S. (1986). Behavioral approaches to toilet training for retarded persons. *International Review of Research in Mental Retardation, 14*, 319–350.

Bettison, S., Davidson, D., Taylor, P., & Fox, B. (1976). The long-term effects of a toilet training programme for the retarded: A pilot study. *Australian Journal of Mental Retardation, 4*(4), 28–35.

Beverly, B. I. (1933). *Incontinence in children* (pp. 718–725). Chicago Pediatric Society.

Böhmer, C. J., Taminiau, J. A., Klinkenberg-Knol, E. C., & Meuwissen, S. G. (2001). The prevalence of constipation in institutionalized people with intellectual disability. *Journal of Intellectual Disability Research, 45*, 212–218.

Carpenter, S. P. (1989). Development of a young man with Prader-Willi syndrome and secondary functional encopresis. *Canadian Journal of Psychiatry, 34*(2), 123–127.

Chang, Y.-J., Lee, M.-Y., Chou, L.-D., Chen, S.-F., & Chen, Y.-C. (2011). A mobile wetness detection system enabling teachers to toilet train children with intellectual disabilities in a public school setting. *Journal of Developmental and Physical Disabilities, 23*(6), 527–533.

Chung, K.-M. (2007). Modified version of Azrin and Foxx’s rapid toilet training. *Journal of Developmental and Physical Disabilities, 19*(5), 449–455.

Dalrymple, N. J., & Ruble, L. A. (1992). Toilet training and behaviors of people with autism: parent views. *Journal of Autism and Developmental Disorders, 22*(2), 265–275.

Dayan, M. (1964). Toilet Training retarded children in a state residential institution. *Mental Retardation, 2*(2), 116–117.

Didden, R., Sikkema, S. P. E., Bosman, I. T. M., Duker, P. C., & Curfs, L. M. G. (2001). Use of modified Azrin-Foxx toilet training procedure with individuals with

Angelman syndrome. *Journal of Applied Research in Intellectual Disabilities, 14*(1), 64–70.

Doleys, D. M., & Arnold, S. (1975). Treatment of childhood encopresis: Full cleanliness training. *Mental Retardation, 13*(6), 14–16.

Ellis, N. R. (1963). Toilet training the severely defective patient: An S-R reinforcement analysis. *American Journal of Mental Deficiency, 68*(1), 98–103.

Feldman, M. A., Ducharme, J. M., & Case, L. (1999). Using self-instructional pictorial manuals to teach child-care skills to mothers with intellectual disabilities. *Behavior Modification, 23*(3), 480–497.

Hagopian, L. P., Fisher, W., Piazza, C. C., & Wierzbicki, J. J. (1993). A water-prompting procedure for the treatment of urinary incontinence. *Journal of Applied Behavior Analysis, 26*(4), 473–474.

Hellström, A.-L., Hanson, E., Hansson, S., Hjälmås, K., & Jodal, U. (1990). Micturition habits and incontinence in 7-year-old Swedish school entrants. *European Journal of Pediatrics, 149*(6), 434–437.

Hoebeke, P., Bower, W., Combs, A., De Jong, T., & Yang, S. (2010). Diagnostic evaluation of children with daytime incontinence. *The Journal of Urology, 183*(2), 699–703.

Huntley, E., & Smith, L. (1999). Long-term follow-up of behavioural treatment for primary encopresis in people with intellectual disability in the community. *Journal of Intellectual Disability Research, 43*(6), 484–488.

Hyams, G., McCoull, K., Smith, P. S., & Tyrer, S. P. (1992). Behavioural continence training in mental handicap: A 10-year follow-up study. *Journal of Intellectual Disability Research, 36*(6), 551–558.

Keen, D., Brannigan, K. L., & Cuskelly, M. (2007). Toilet training for children with autism: The effects of video modeling. *Journal of Developmental and Physical Disabilities, 19*(4), 291–303.

Kroeger, K. A., & Sorensen-Burnworth, R. (2009). Toilet training individuals with autism and other developmental disabilities: A critical review. *Research in Autism Spectrum Disorders, 3*(3), 607–618. https://doi.org/10.1016/j.rasd.2009.01.005

Lancioni, G. E., O'Reilly, M. F., & Basili, G. (2001). Treating encopresis in people with intellectual disabilities: A literature review. *Journal of Applied Research in Intellectual Disabilities, 14*(1), 47–63.

Lancioni, G. E. (1980). Teaching independent toileting to profoundly retarded deaf-blind children. *Behavior Therapy, 11*(2), 234–244. https://doi.org/10.1016/S0005-7894(80)80023-2

Levine, M. N., & Elliot, C. B. (1970). Toilet training for profoundly retarded with a limited staff. *Mental Retardation, 8*(3), 48–50.

Loening-Baucke, V. (1997). Urinary incontinence and urinary tract infection and their resolution with treatment of chronic constipation of childhood. *Pediatrics, 100*(2), 228–232.

Lowery, S. P., Srour, J. W., Whitehead, W. E., & Schuster, M. M. (1985). Habit training as treatment of encopresis secondary to chronic constipation. *Journal of Pediatric Gastroenterology and Nutrition, 4*(3), 397–401.

Luiselli, J. K. (1994). Toilet training children with sensory impairments in a residential school setting. *Behavioral Interventions, 9*(2), 105–114.

Luiselli, J. K. (1996). A case study evaluation of a transfer-of-stimulus control toilet training procedure for a child with pervasive developmental disorder. *Focus on Autism and Other Developmental Disabilities, 11*(3), 158–162.

Mahoney, K., Wagenen, R. K. V., & Meyerson, L. (1971). Toilet training of normal and retarded children 1. *Journal of Applied Behavior Analysis, 4*(3), 173–181. https://doi.org/10.1901/jaba.1971.4-173

Matson, J. L., Horovitz, M., & Sipes, M. (2011). Characteristics of individuals with toileting problems and intellectual disability using the Profile of Toileting Issues (POTI). *Journal of Mental Health Research in Intellectual Disabilities, 4*, 53–63.

Matson, J. L., & LoVullo, S. V. (2009). Encopresis, soiling and constipation in children and adults with developmental disability. *Research in Developmental Disabilities, 30*(4), 799–807.

Matson, J. L., Neal, D., Hess, J. A., & Kozlowski, A. K. (2011). Assessment of toileting difficulties in adults with intellectual disabilities: An examination using the profile of toileting issues (POTI). *Research in Developmental Disabilities, 32*(1), 176–179.

Mowrer, O. H., & Mowrer, W. M. (1938). Enuresis—A method for its study and treatment. *American Journal of Orthopsychiatry, 8*(3), 436–459. https://doi.org/10.1111/j.1939-0025.1938.tb06395.x

Ozcan, N., & Cavkaytar, A. (2009). Parents as teachers: Teaching parents how to teach toilet skills to their children with autism and mental retardation. *Education and Training in Developmental Disabilities, 44*(2), 237–243.

Piazza, C. C., Fisher, W., Chinn, S., & Bowman, L. (1991). Reinforcement of incontinent stools in the treatment of encopresis. *Clinical Pediatrics, 30*(1), 28–32.

Rinald, K., & Mirenda, P. (2012). Effectiveness of a modified rapid toilet training workshop for parents of children with developmental disabilities. *Research in Developmental Disabilities, 33*(3), 933–943.

Sells-Love, D., Rinaldi, L. M., & McLaughlin, T. F. (2002). Toilet training an adolescent with severe mental retardation in the classroom: A case study. *Journal of Developmental and Physical Disabilities, 14*(2), 111–118.

Smith, L., Smith, P., & Kwok Yi Lee, S. (2000). Behavioural treatment of urinary incontinence and encopresis in children with learning disabilities: Transfer of stimulus control. *Developmental Medicine & Child Neurology, 42*(4), 276–279.

Smith, L. J. (1981). Training severely and profoundly mentally handicapped nocturnal enuretics. *Behaviour Research and Therapy, 19*(1), 67–74.

Smith, L. J., Franchetti, B., McCoull, K., Pattison, D., & Pickstock, J. (1994). A behavioural approach to retraining bowel function after long-standing constipation and faecal impaction in people with learning disabilities. *Developmental Medicine & Child Neurology, 36*(1), 41–49.

Smith, P. S., & Smith, L. J. (1977). Chronological age and social age as factors in intensive daytime toilet training of institutionalized mentally retarded individuals. *Journal of Behavior Therapy and Experimental Psychiatry, 8*(3), 269–273.

Smith, P. S., Britton, P. G., Johnson, M., & Thomas, D. A. (1975). Problems involved in toilet-training profoundly mentally handicapped adults. *Behaviour Research and Therapy, 13*(4), 301–307. https://doi.org/10.1016/0005-7967(75)90035-2

Sureshkumar, P., Craig, J. C., Roy, L. P., & Knight, J. F. (2000). Daytime urinary incontinence in primary school children: A population-based survey. *Journal of Pediatrics, 137*(6), 814–818.

Taylor, S., Cipani, E., & Clardy, A. (1994). A stimulus control technique for improving the efficacy of an established toilet training program. *Journal of Behavior Therapy and Experimental Psychiatry, 25*(2), 155–160. https://doi.org/10.1016/0005-7916(94)90010-8

Van Laecke, E., Raes, A., Vande Walle, J., & Hoebeke, P. (2009). Adequate fluid intake, urinary incontinence, and physical and/or intellectual disability. *The Journal of Urology, 182*(4), 2079–2084.

Van Wagenen, R. K., Meyerson, L., Kerr, N. J., & Mahoney, K. (1969). Field trials of a new procedure for toilet training. *Journal of Experimental Child Psychology, 8*(1), 147–159.

von Gontard, A. (2013). Urinary incontinence in children with special needs. *Nature Reviews Urology, 10*, 667–674.

Waye, M. F., & Melnyr, W. T. (1973). Toilet training of a blind retarded boy by operant conditioning. *Journal of Behavior Therapy and Experimental Psychiatry, 4*(3), 267–268. https://doi.org/10.1016/0005-7916(73)90085-2

Wright, L., & Walker, C. E. (1978). A simple behavioral treatment program for psychogenic encopresis. *Behaviour Research and Therapy, 16*(3), 209–212.

Correction to: Early Intervention

Michael J. Guralnick and Mary Beth Bruder

Correction to:
Chapter 39 in: J. L. Matson (ed.), *Handbook of Intellectual Disabilities*, Autism and Child Psychopathology Series, https://doi.org/10.1007/978-3-030-20843-1_39

The original version of Chapter 39 was inadvertently published with the following article note. The article note has been removed from the chapter in the book.

The updated online version of this book can be found at
https://doi.org/10.1007/978-3-030-20843-1_39

J. L. Matson (ed.), *Handbook of Intellectual Disabilities*, Autism and Child Psychopathology
Series, https://doi.org/10.1007/978-3-030-20843-1_58

Index

J. L. Matson (ed.), *Handbook of Intellectual Disabilities*, Autism and Child Psychopathology Series, https://doi.org/10.1007/978-3-030-20843-1