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Handbook of Dual Diagnosis

Assessment and Treatment in Persons with Intellectual Disorders

Springer

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Contents

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History of Dual Diagnosis

1

Johnny L. Matson and W. Jason Peters

Introduction

For most of the history of modern research on intellectual disabilities (ID), mental health was not addressed. In fact, to the extent that these topics were addressed, they were considered separate. Also, until the 1960s and 1970s, psychoanalytic interventions dominated. The general thinking was that people with ID did not have sufficient ego strength to develop mental health problems. With the development of biobehavioral treatments such as applied behavior analysis and psychotropic drugs, the thinking regarding dual diagnosis changed rapidly. The focus of this volume will be on the emerging assessment and intervention strategies that are available.

Nature and Incidence

One of the pioneering researchers in the field of dual diagnosis was Frank Menolascino. In one of the earliest studies on the topic, 616 children who were under the age of 8 were studied (Menolascino, 1965). All of the participants were suspected of displaying ID. He noted that 31% or 191 children had "emotional disturbances." Of this group, 24.5% had ID, while 6.5% had primary emotional disturbance only. These mental health problems were characterized as prominent. Menolascino stressed the need to routinely assess for mental health problems in children with ID, a position that gradually gained momentum.

Eaton and Menolascino (1982) further underscore the importance of the findings of the 1965 paper. They countered the suggestion that persons with ID do not have mental health concerns. Rather, because of central nervous system impairments, these authors argued that the rates of mental health problems occur at higher levels among persons with ID compared to the overall population. This was an early acknowledgement of the serious nature of dual diagnosis. Over time, these higher incidence rates were reported in numerous studies (Matson & Barrett, 1982; Matson, Barrett, & Helsel, 1988; Prout & Schaefer, 1985; Reed & Clements, 1989).

There are a number of factors that appear to contribute to these high rates of psychopathology. Eaton and Menolascino (1982) were among the first researchers to address this issue. They noted that persons with ID have high incidence of central nervous impairments. They further stress that these persons have poor social and interpersonal coping skills which can result in more severe expressions of psychopathology.

Ruedrich and Menolascino (1984) also noted that service delivery models needed to change

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with the move toward deinstitutionalization. Where services are delivered, the need for more community-based mental health programs became critical issues. Additionally, they noted that many behaviors, which are normalized in institutional settings, will not be tolerated in the community. Unfortunately, while these insights were published over three decades ago, the same concerns persist today. This issue is particularly troubling since researchers and clinicians have long recognized that persons with ID experience the full range of mental health disorders (Reiss, 1988).

Rates of psychopathology are also higher in persons with ID relative to the overall population. For example, Emerson and Hatton (2007) looked at rates of psychopathology among 64 children and adolescents with ID, and 17,774 children and adolescents without ID. These authors found that 36% of their sample with ID, and 8% of their sample without ID, evinced mental health problems. The authors report that these rates are consistent with others who have looked at rates of psychopathology in the ID population (Dykens, 2000; Einfeld & Tonge, 1996; Emerson, 2003).

Crews, Bonaventura, and Rowe (1994) found a 15.55% point prevalence of mental health problems in over 1000 people with ID. These authors reported higher rates of psychopathology in persons with mild levels of ID. Whether these rates are disproportionate, or whether it is more difficult to detect mental health issues in more severely intellectually impaired persons, is open to question. Borthwick-Duffy (1994) addressed this point and stressed the difficulties with definitions, symptom presentation, and identification of various forms of psychopathology. Also in question are causes and risk factors for these various problems (Matson & Sevin, 1994). The importance of these factors is further underscored by the recognition that mental health concerns are a major factor in the failure to adjust to living in the community.

Much has yet to be done to better understand psychopathology among persons with ID. One of the more recent developments involves evaluating specific types of behavior and mental health concerns. For example, Glenn, Bihm, and Lammers (2003) assessed anxiety and depression in persons with ID. Their sample consisted of 46 people from borderline to moderate cognitive functioning. They noted high correlations on self-report for depression and anxiety disorders. Depressive episodes in adults with ID have also been studied by Cooper and Collacott (1996). Their paper was a review, and the main conclusion was that the research is limited and much more needs to be done. The same conclusion applies today since the pace of innovation has been very slow in recent decades.

In another of these studies, Folch et al. (2018) look at the relationship between self-injurious behavior (SIB) and other forms of psychopathology among persons with ID. The sample of 833 adults with ID presented with 16.2% who also demonstrated SIB. Risk factors included comorbid psychopathology and the use of psychotropic medications. Thus, research in the field of dual diagnosis is going from the very general to the more specific. However, as noted, the rate of new knowledge development has slowed and a great deal has yet to be learned. A major reason for this switch appears to be the dramatic increase in research on autism. Many researchers and clinicians who previously studied the broader field of dual diagnosis now appear to have narrowed their focus to autism.

Rationale and Early Treatment Studies

The early efforts to treat persons with ID began in the 1960s. As the new technology based on operant and classical conditioning emerged, it became known as behavior modification and was then applied to a number of independent living skills. Whitman and Scibak (1981) noted a gradual increase in these studies between 1962 and 1979. The number of studies by topic from highest to lowest was self-help, social behavior, language, and behavior problems such as self-stimulation, SIB, aggression, and tantrums. Thus, the roots of dual diagnosis research began with the identification and treatment of challenging behaviors.

The complexity of these treatments has increased with time. Ellis (1963), in an early toilet training study, relied heavily on daytime schedules and reinforcement in a stimulus-response paradigm. Perhaps even more important than early successes like this one was the notion that persons with ID could learn and that the behavior modification template was an effective means of accomplishing this goal.

As the field of behavior modification evolved, distinctions between variations of this treatment model emerged. The term behavior modification has faded, while terms such as applied behavior analysis have become popular replacements. Other distinctions were made by Gardner and Cole (1987). They make a distinction between behavior management and behavior controls, which are largely external applications of contingencies to active behavior change. The latter intervention is deemed more important, and it is argued that it should be applied more frequently since the focus is on teaching coping skills that are controlled by the client.

Another big focus, and a set of interventions that emphasizes coping skills, involves social skills training. The importance of treatment in this area is further underscored by the fact that social skills deficits are a defining characteristic of ID and have been for decades (Doll, 1941; Heber, 1959). These social excesses and deficits are linked to overall adjustment in mental health. This point has been underscored for many years (Phillips & Zigler, 1961).

Early attempts to teach social skills to persons with ID go back for decades. Knapczyk and Yoppi (1975), for example, used a token system to assist five children with ID in the acquisition of appropriate play skills. Also, training packages that included instructions, modeling, and reinforcement have been employed to train hand waving (Stokes, Baer, & Jackson, 1974) and asking questions (Twardosz & Baer, 1973), among other skills. More recently, peer-delivered social skills training has been described as an effective intervention for adults with Down syndrome and autism spectrum disorder (Davis, Spriggs, Rodgers, & Campbell, 2018).

Social skills training has a role to play in most instances of dual diagnosis. Another treatment that has received a great deal of attention is the use of psychotropic medication with this population. The widespread use of these medications among persons with ID goes back for decades. For example, Burk and Menolascino (1968) describe the use of Haldol to treat "emotionally disturbed" children with ID.

From these early beginnings, the use of psychotropic drugs has become very popular in the treatment of persons with mental health issues. Often, and unfortunately, behavior problems such as conduct disorders are treated with psychotropic drugs, usually antipsychotics (Santosh & Baird, 1999). These rates continue to trend upward. For example, in a study done in New York state, Tsiouris, Kim, Brown, Pettinger, and Cohen (2013) studied 4069 adults with ID. They found that 58% of their sample was on one or more psychotropic drugs. Thus, even with the move to deinstitutionalization and the association that persons in the community receive fewer psychotropic drugs than persons living in institutions and other congregate settings, rates continue to rise. These authors state the obvious; psychotropic drugs are overused with this population.

One of the major reasons for the overuse of these medications is the continuing use for off-label problems with little or no empirical support. The biggest problem in this regard continues to involve challenging behaviors (Molyneux, Emerson, & Caine, 1999; Tsiouris, 2010). In a British study, 32,306 adults with ID were assessed. They found that 49% of the sample received psychotropics with a higher percent of persons with challenging behaviors than persons without challenging behaviors receiving these drugs (R. Sheehan et al., 2015).

As noted, increased numbers of people are receiving psychotropic drugs for challenging behaviors displayed by persons with ID, and at increasingly higher doses (Matson & Neal, 2009). A number of uncontrolled studies report on the effectiveness of this treatment model. However, the most well-controlled study on the topic showed no difference between placebo,

risperidone, or Haldol for the treatment of challenging behaviors in adults with ID (Tyrer et al., 2008).

The issues with psychotropic medications are further exacerbated by the increasing use of multiple medications. A paper by Stolker, Heerdink, Leufkens, Clerkx, and Nolen (2001) highlights this point. Their paper was based on a Dutch sample of 105 individuals with ID who were over 16 years of age. They reported that for persons who received medication, 48% were prescribed multiple psychotropic drugs. Factors most commonly associated with the use of two or more psychotropic medications included longer psychiatric inpatient stays, psychosis, aggression, and attention-seeking behavior.

All of these historical trends in medication use for persons with ID targeting mental health issues and challenging behaviors must be viewed in the light of the potential drug side effects that can result. Risk factors include longer durations of drug use, use of multiple drugs, and higher dose levels. Tardive dyskinesia, akathisia, sedation, and withdrawal effects are among the behaviors noted as concerns (Matson & Mahan, 2010). These concerns have been known and discussed since at least the 1950s (Ey, Faure, & Rappard, 1956). Unfortunately, few methods have been developed to assess drug side effects in persons with ID, and research on the topic has been sporadic at best.

Defining and Assessing Dual Diagnosis

As noted, the general consensus is that children and adults with ID display significantly greater rates of psychopathology than the general population (Emerson, Einfeld, & Stancliffe, 2010). On the assessment front, much work is needed. However, some scales with good psychometrics have been reported. Three of these measures will be discussed, and their historical significance will be noted.

The first scale developed to address mental health concerns of people with ID was the Psychopathology Instrument for Mentally Retarded Adults (PIMRA; Matson, Kazdin, & Senatore, 1984). At this writing, it is the most heavily researched scale and has well-established reliability and validity. An informant report and a self-report were developed. The scale was designed to measure seven forms of psychopathology: somatoform disorders, personality problems, schizophrenia, psychosexual disorders, depression, anxiety, and adjustment disorders.

A number of the subscales have been validated. Swiezy, Matson, Kirkpatrick-Sanchez, and Williams (1995) published a paper in which they addressed the schizophrenia and depression subscales. Sixty-five adults with mild or moderate ID who lived in both the community and in institutions were studied. Participants lived in the southern USA. Subscale scores were validated against DSM-III-R criteria.

Another study addressing schizophrenia and the PIMRA was published comparing a sample of adults with mild or moderate ID (n = 48) to a sample of persons without ID (n = 38; Linaker & Helle, 1994). These authors observed that persons with ID had fewer delusions, they were more incoherent, and they displayed higher levels of flat affect than the sample with typical intellectual functioning.

A Swedish adaptation of the PIMRA was published by Gustafsson and Sonnander (2005). They tested 66 staff informants in group homes and 71 residential staff. Higher PIMRA scores were associated with DSM-III-R diagnoses made by a psychiatrist.

In another study on this topic, in this case by Dutch investigators, 89 adults with mild ID were evaluated (van Minnen, Savelsberg, & Hoogduin, 1994). They found good reliability for the informant and self-rating forms. Interrater reliability was low to modest with good criterion validity. Also, correlations between the two forms were modest, but high PIMRA scores were related to behavior problems. The point of these studies is that researchers internationally recognized in short order the need for measures to aid in the diagnosis of mental health issues in persons with ID. Also, some interesting patterns emerged. For example, meeting criteria for multiple disorders was common, and at least for persons in the mild

to moderate range of ID, there were no differences in the ratio of diagnosis when compared to the general population (Linaker & Nitter, 1990).

The move to recognize dual diagnosis and to develop scales to address this issue was further underscored with the introduction of the Reiss Screen for Maladaptive Behavior. This scale, which was developed to assess various forms of psychopathology in persons with ID, has proven to have good reliability and validity (Sturmey, Burcham, & Perkins, 1995). The measure consists of eight scales: psychosis, autism, depression (physical signs), avoidant behavior, aggression, depression (behavioral signs), dependent personality disorder, and paranoia (Havercamp & Reiss, 1997). In the Havercamp and Reiss (1997) study, the authors used factor analysis to establish that a goodness of fit was found with the factors of the scale. The analysis was carried out with 448 people with mild to profound ID. These results were largely confirmed by others (Sturmey & Bertman, 1994). However, in instances where confirmation was not supported, Reiss (1997) stated that significant statistical errors were present. Also, the Reiss Screen has had other uses, such as establishing prevalence rates of psychopathology among persons with ID (Reiss, 1990).

The final scale briefly reviewed as an important advance in differential diagnosis of persons with ID is the Diagnostic Assessment for the Severely Handicapped (DASH and DASH-II; Matson, Gardner, Coe, & Sovner, 1991). The first scales used in differential diagnosis focused on mild and moderate ID primarily. However, as the evidence accumulated, it became evident that symptom expression for the most cognitively impaired was considerably different than what was noted for persons in the mild to moderate range of ID – then came the DASH-II, which focused on a scale devoted to persons with severe and profound ID. In the initial paper by Matson and associates (1991), 506 persons with severe and profound ID were assessed on the 83-item scale. The DASH-II has 13 subscales. They include anxiety, mood disorders, impulse control, mania, organic problems, pervasive developmental disorders, elimination disorders, and sleep problems. In this inaugural study, relatively good reliability was reported.

A number of studies have addressed the psychometrics of the DASH-II. Typical of this research was a paper by Matson and Smiroldo (1997) where they validated the mania scale of the DASH-II. In this paper, 22 adult inpatients with severe or profound ID were studied. All participants had previously been diagnosed with bipolar disorder or received no Axis I diagnosis on DSM-IV. The DASH-II mania subscale proved to be an accurate method of diagnosing mania/bipolar disorder.

Hill and Furniss (2006), in a study conducted in the UK, evaluated patterns of dual diagnosis and challenging behaviors. Their sample consisted of 82 children and young adults who lived in one of four residential facilities. Participants with elevated DASH-II scores on the pervasive developmental disorders/autism scale were more likely to also have elevated scores on the organic disorders, anxiety, and mania scales as well as more stereotypies than persons without elevated pervasive developmental disorders/autism scores.

Another study looked at the relationship between dual diagnosis and adaptive skills as measured by the Vineland Adaptive Behavior Scales (Matson, Rivet, Fodstad, Dempsey, & Boisjoli, 2009). These authors studied 377 people between 18 and 88 years of age. Three hundred twelve people were diagnosed with profound ID, 40 had severe ID, and 21 evinced moderate ID, while 4 participants were classified with mild ID. Pervasive developmental disorders/autism accounted for 90 people. Three groups were then established, ID, ID plus PDD/autism, and ID plus PDD/autism, and another elevated disorder on the DASH-II. The more handicapping conditions that were present, the greater the adaptive skills deficits.

The sequence of developments in dual diagnosis assessment has revolved around norm-based checklists that cover a variety of forms of psychopathology. The earliest scales were developed in the USA and focused on mild to profound ID, or for people with higher levels of cognitive functioning. Scales were then developed for severe-profound levels of ID specifically. A number of

other countries in short order adapted existing scales or developed new measures for their respective populations. The greatest activity occurred between the 1980s and early 2000s. There is a need for a great deal of more research on the topic. However, as previously noted, what seems to have occurred in large part is a shift to the area of autism. Diagnostic replacement has occurred in many instances with fewer people receiving a primary diagnosis of ID and more people receiving an autism diagnosis. It is recognized that an increase in research on autism, which had been neglected for some time, is a good thing. However, other behavioral, developmental, and mental health issues persist and warrant continued study.

New Innovations and Models of Care

In the last few decades, the model of service delivery for persons with dual diagnosis has changed considerably. The focus has been on normalization: the move from institutions to the community. As a result, assessment and intervention has shifted to a large degree from inpatient to outpatient services. One justification for this major shift in care models was that institutional care created and/or exacerbated mental health issues. Also, it was argued that community mental health would result in as good or better mental healthcare. However, the data do not support these claims. Nøttestad, Strømgren, and Linaker (2000) found that psychopathology among persons with ID remained at high rates and that little or no change in rates and severity of dual diagnosis changed with deinstitutionalization. These authors also noted that access to qualified psychologists and psychiatrists was low, and there was a decrease in the use of psychologists. This pattern, based on our informal observations, appears to exist in many countries. As a result, the challenge is to increase the number of qualified professionals and to develop more robust models of care delivery.

Gardiner, Iarocci, and Moretti (2017) underscore the challenges associated with the treatment of dual diagnosis. They note how complex these problems can be to diagnose and treat given the interface between mental health issues and ID. They rightly point out that well-validated and innovative treatments are needed. However, relative to other populations with mental health needs, there is a dearth of available methods and procedures. Additionally, few researchers are actively working on the problem presently.

One option is to adapt treatments for mental health problems in the general population. In practice, this is certainly the most common approach presently. Torrey (1993), for example, notes that while not trained to treat those with dual diagnosis, psychiatrists in the community, in general, have the basic skills to deal with mental health concerns such as depression, anxiety, and psychosis. This author points out that, with some modifications that are specific to the ID population, they should be able to provide appropriate treatments to these persons. Again, however, little has been done to develop formal models to address this issue.

Another topic that is of considerable interest, particularly with children and adolescents, is parental involvement. In one such study, Sheehan and Guerin (2018) describe a treatment model to deal with parental loss and grief, particularly during the first few months of their child's diagnosis. They describe a "Dual Process Model" based off of timing of information sharing and how it was done.

Being able to predict particularly problematic behaviors in persons with mental health concerns has also been a topic of study. Skeem, Mulvey, Lidz, Gardner, and Schubert (2002) describe an outpatient treatment model for persons who have chronic violent outbursts. The context of the service model involved changes in living arrangements and their social relationships with others. This goal was accomplished with a prescreen via medical chart reviews. Issues such as number of violent acts and ongoing psychopathology, such as delusions and hallucinations, were plotted. Those with red flags (about 10%) were assessed with the Brief Symptom Inventory to determine the degree of

drug use, the number of violent incidences in the community over the previous 2 months, and scores on the hostility scale. This preventative model was aimed at establishing which individuals would be the best candidates for intensive treatment directed at decelerating aggression.

Conclusions

The field of dual diagnosis is relatively new as a major mental health discipline. Having said that, the overlap in psychopathology and ID is now recognized. Treatment developments such as psychotropic medications along with advances in behavior therapy and applied behavior analysis have played major roles. Also, beginning in the 1980s, a number of standardized tests were developed. Initially, these methods were focused primarily on adults with mild or moderate ID. With time, more tests were designed for children and persons with severe or profound ID.

The last two decades, however, has seen a marked drop-off in research interest. This trend is unfortunate because the number of afflicted persons continues to be high. Also, much more work is needed regarding models for treatment delivery and for specific mental health problems that are displayed. This later point has led to the need to adapt treatments for persons with typical IQ to persons with ID. The current volume is focused on a review of the current status of assessment and treatment in dual diagnosis. Specific strengths in the literature and the need for future developments are also covered.

References

Borthwick-Duffy, S. A. (1994). Epidemiology and prevalence of psychopathology in people with mental retardation. *Journal of Consulting and Clinical Psychology, 62*, 17.

Burk, H. W., & Menolascino, F. J. (1968). Haloperidol in emotionally disturbed mentally retarded individuals. *American Journal of Psychiatry, 124*, 1589–1591.

Cooper, S.-A., & Collacott, R. A. (1996). Depressive episodes in adults with learning disabilities. *Irish Journal of Psychological Medicine, 13*, 105–113.

Crews, W. D., Bonaventura, S., & Rowe, F. (1994). Dual diagnosis: Prevalence of psychiatric disorders in a large state residential facility for individuals with mental retardation. *American Journal on Mental Retardation, 98*, 724–731.

Davis, M. A. C., Spriggs, A., Rodgers, A., & Campbell, J. (2018). The effects of a peer-delivered social skills intervention for adults with comorbid Down syndrome and autism Spectrum disorder. *Journal of Autism and Developmental Disorders, 48*, 1869–1885.

Doll, E. A. (1941). The essentials of an inclusive concept of mental deficiency. *American Journal of Mental Deficiency, 46*, 214–219.

Dykens, E. M. (2000). Annotation: Psychopathology in children with intellectual disability. *The Journal of Child Psychology and Psychiatry and Allied Disciplines, 41*, 407–417.

Eaton, L. F., & Menolascino, F. J. (1982). Psychiatric disorders in the mentally retarded: Types, problems, and challenges. *The American Journal of Psychiatry, 139*, 1297–1303.

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*, 99–109.

Ellis, N. R. (1963). Toilet training the severely defective patient: An SR reinforcement analysis. *American Journal of Mental Deficiency, 68*, 99–103.

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

Emerson, E., Einfeld, S., & Stancliffe, R. J. (2010). The mental health of young children with intellectual disabilities or borderline intellectual functioning. *Social Psychiatry and Psychiatric Epidemiology, 45*, 579–587.

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.

Ey, I., Faure, H., & Rappard, P. (1956). Les reactionis d'intolérance vis-à-vis de la chlorpromazine. *Encephale, 45*, 790–796.

Folch, A., Cortés, M. J., Salvador-Carulla, L., Vicens, P., Irazábal, M., Muñoz, S., … Vilella, E. (2018). Risk factors and topographies for self-injurious behaviour in a sample of adults with intellectual developmental disorders. *Journal of Intellectual Disability Research, 62*(12), 1018–1029.

Gardiner, E., Iarocci, G., & Moretti, M. (2017). Integrative care for adolescents with dual diagnosis: Considering trauma and attachment within an innovative model for clinical practice. *Journal of Mental Health Research in Intellectual Disabilities, 10*, 321–344.

Gardner, W. I., & Cole, C. L. (1987). Behavior treatment, behavior management, and behavior control: Needed distinctions. *Behavioral Interventions, 2*(1), 37–53.

Glenn, E., Bihm, E. M., & Lammers, W. J. (2003). Depression, anxiety, and relevant cognitions in persons with mental retardation. *Journal of Autism and Developmental Disorders, 33*, 69–76.

Gustafsson, C., & Sonnander, K. (2005). A psychometric evaluation of a Swedish version of the psychopathology inventory for mentally retarded adults (PIMRA). *Research in Developmental Disabilities, 26*, 183–201.

Havercamp, S. M., & Reiss, S. (1997). The Reiss screen for maladaptive behavior: Confirmatory factor analysis. *Behaviour Research and Therapy, 35*, 967–971.

Heber, R. (1959). A manual on terminology and classification in mental retardation. *American Journal of Mental Deficiency, 64*, 1–17.

Hill, J., & Furniss, F. (2006). Patterns of emotional and behavioural disturbance associated with autistic traits in young people with severe intellectual disabilities and challenging behaviours. *Research in Developmental Disabilities, 27*, 517–528.

Knapczyk, D. R., & Yoppi, J. O. (1975). Development of cooperative and competitive play responses in developmentally disabled children. *American Journal of Mental Deficiency, 80*, 245–255.

Linaker, O. M., & Helle, J. (1994). Validity of the schizophrenia diagnosis of the Psychopathology Instrument for Mentally Retarded Adults (PIMRA): A comparison of schizophrenic patients with and without mental retardation. *Research in Developmental Disabilities, 15*, 473–486.

Linaker, O. M., & Nitter, R. (1990). Psychopathology in institutionalised mentally retarded adults. *The British Journal of Psychiatry, 156*(4), 522–525.

Matson, J. L., & Barrett, R. P. (1982). *Psychopathology in the mentally retarded*. New York, NY: Grune & Stratton.

Matson, J. L., Barrett, R. P., & Helsel, W. J. (1988). Depression in mentally retarded children. *Research in Developmental Disabilities, 9*, 39–46.

Matson, J. L., Gardner, W. I., 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. *The British Journal of Psychiatry, 159*, 404–409.

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., & Neal, D. (2009). Psychotropic medication use for challenging behaviors in persons with intellectual disabilities: An overview. *Research in Developmental Disabilities, 30*, 572–586.

Matson, J. L., Rivet, T. T., Fodstad, J. C., Dempsey, T., & Boisjoli, J. A. (2009). Examination of adaptive behavior differences in adults with autism spectrum disorders and intellectual disability. *Research in Developmental Disabilities, 30*, 1317–1325.

Matson, J. L., & Sevin, J. A. (1994). Theories of dual diagnosis in mental retardation. *Journal of Consulting and Clinical Psychology, 62*, 6.

Matson, J. L., & Smiroldo, B. B. (1997). Validity of the mania subscale of the Diagnostic Assessment for the Severely Handicapped-II (DASH-II). *Research in Developmental Disabilities, 18*, 221–225.

Menolascino, F. J. (1965). Emotional disturbance and mental retardation. *American Journal of Mental Deficiency, 70*, 248–256.

Molyneux, P., Emerson, E., & Caine, A. (1999). Prescription of psychotropic medication to people with intellectual disabilities in primary health-care settings. *Journal of Applied Research in Intellectual Disabilities, 12*, 46–57.

Nøttestad, J. A., Strømgren, B., & Linaker, O. M. (2000). Psychiatric and behavioral disturbances in elderly mentally retarded before and after deinstitutionalization. *Nordic Journal of Psychiatry, 54*, 281–286.

Phillips, L., & Zigler, E. (1961). Social competence: The action-thought parameter and vicariousness in normal and pathological behavior. *The Journal of Abnormal and Social Psychology, 63*, 137.

Prout, H. T., & Schaefer, B. M. (1985). Self-reports of depression by community-based mildly mentally retarded adults. *American Journal of Mental Deficiency, 90*, 220–222.

Reed, J., & Clements, J. (1989). Assessing the understanding of emotional states in a population of adolescents and young adults with mental handicaps. *Journal of Intellectual Disability Research, 33*, 229–233.

Reiss, S. (1988). Dual diagnosis in the United States. *Australia and New Zealand Journal of Developmental Disabilities, 14*, 43–48.

Reiss, S. (1990). Prevalence of dual diagnosis in community-based day programs in the Chicago metropolitan area. *American Journal on Mental Retardation, 94*, 578–585.

Reiss, S. (1997). Comments on the Reiss screen for maladaptive behaviour and its factor structure. *Journal of Intellectual Disability Research, 41*, 346–354.

Ruedrich, S., & Menolascino, F. J. (1984). Dual diagnosis of mental retardation and mental illness. In *Handbook of mental illness in the mentally retarded* (pp. 45–81). Boston, MA: Springer.

Santosh, P. J., & Baird, G. (1999). Psychopharmacotherapy in children and adults with intellectual disability. *The Lancet, 354*, 233–242.

Sheehan, P., & Guerin, S. (2018). Exploring the range of emotional response experienced when parenting a child with an intellectual disability: The role of dual process. *British Journal of Learning Disabilities, 46*, 109–117.

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.

Skeem, J. L., Mulvey, E. P., Lidz, C., Gardner, W., & Schubert, C. (2002). Identifying psychiatric patients at risk for repeated involvement in violence: The next step toward intensive community treatment programs. *International Journal of Forensic Mental Health, 1*, 155–170.

Stokes, T. F., Baer, D. M., & Jackson, R. L. (1974). Programming the generalization of a greeting response in four retarded children. *Journal of Applied Behavior Analysis, 7*, 599–610.

Stolker, J. J., Heerdink, E. R., Leufkens, H. G., Clerkx, M. G., & Nolen, W. A. (2001). Determinants of multiple psychotropic drug use in patients with mild intellectual disabilities or borderline intellectual functioning and psychiatric or behavioral disorders. *General Hospital Psychiatry, 23*, 345–349.

Sturmey, P., & Bertman, L. J. (1994). Validity of the Reiss screen for maladaptive behavior. *American Journal of Mental Retardation, 99*, 201–206.

Sturmey, P., Burcham, K. J., & Perkins, T. S. (1995). The Reiss screen for maladaptive behaviour: Its reliability and internal consistencies. *Journal of Intellectual Disability Research, 39*, 191–195.

Swiezy, N. B., Matson, J. L., Kirkpatrick-Sanchez, S., & Williams, D. E. (1995). A criterion validity study of the schizophrenia subscale of the Psychopathology Instrument for Mentally Retarded Adults (PIMRA). *Research in Developmental Disabilities, 16*, 75–80.

Torrey, W. C. (1993). Psychiatric care of adults with developmental disabilities and mental illness in the community. *Community Mental Health Journal, 29*, 461–476.

Tsiouris, J. A. (2010). Pharmacotherapy for aggressive behaviours in persons with intellectual disabilities: Treatment or mistreatment? *Journal of Intellectual Disability Research, 54*, 1–16.

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*, 719–731.

Twardosz, S., & Baer, D. M. (1973). Training two severely retarded adolescents to ask questions. *Journal of Applied Behavior Analysis, 6*, 655–661.

Tyrer, P., Oliver-Africano, P. C., Ahmed, Z., Bouras, N., Cooray, S., Deb, S., … Crawford, M. (2008). Risperidone, haloperidol, and placebo in the treatment of aggressive challenging behaviour in patients with intellectual disability: A randomised controlled trial. *The Lancet, 371*, 57–63.

van Minnen, A., Savelsberg, P. M., & Hoogduin, K. A. L. (1994). A Dutch version of the Psychopathology Inventory for Mentally Retarded Adults (PIMRA). *Research in Developmental Disabilities, 15*, 269–278.

Whitman, T. L., & Scibak, J. W. (1981). Behavior modification research with the mentally retarded. In J. L. Matson & J. R. McCartney (Eds.), *Handbook of behavior modification with the mentally retarded* (pp. 1–28). Boston, MA: Springer US.

2 The Classification of Mental Disorders: Dual Diagnosis in Persons with Intellectual Disabilities

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Introduction

Names matter. Nosology matters. *What is defined as a mental disorder* and *how* we define mental disorders matter. Neuroscientists in the laboratory and in clinical settings have a professional obligation to constantly challenge the state of knowledge in order to classify mental disorders accurately and fairly. This responsibility is rooted in our identity as scientists and in our professional ethics of beneficence, autonomy, nonmaleficence, and justice. In developing classification systems *beneficence*, advocating for good, requires that the system be directed by science to the service of individuals and society. Balancing the needs of the individual and society in the classification of mental disorders is complex as demonstrated by comparing the American Psychiatric Association's *Diagnostic and Statistical Manual* (DSM) and the World Health Organization's *International Classification of Diseases* (ICD); the ICD intentionally targets more global conceptualizations of the *individual* and *society* (Luckasson & Schalock, 2013). By avoiding unnecessarily medicalization, classification systems support *autonomy*, or self-directed decision-making, and respect for the dignity of the individual while avoiding the *maleficence*, or harm, by promoting stigma. Classification systems must respect *justice*, or basic fairness, to fairly inform the allocation of scarce treatment, educational and support resources, and how courts define culpability. This chapter explores the evolution of the major diagnostic systems for mental disorders – the APA *DSM*, the WHO *ICD*, and the NIMH *Research Domain Criteria* (RDoC), the National Association for the Dually Diagnosed *Diagnostic Manual-Intellectual Disability* (DM-ID), and the Royal College of Psychiatrists *Diagnostic Criteria for Psychiatric Disorders for Use with Adults with Learning Disabilities/Mental Retardation* (DC-LD). In addition, the American Association on Intellectual and Developmental Disabilities *Intellectual Disability: Definition, Classification, and Systems of Supports* manual (Schalock et al., 2010) and legal definitions of ID will be explored. Additional diagnostic concepts and systems will be introduced including precision medicine (Desmond-Hellman & Sawyers, 2011), the hierarchical taxonomy of psychopathology (HiTOP) (Kotov et al., 2017), the p factor (Caspi & Moffitt, 2018; Lahey et al., 2012), and lastly the International Classification of Sleep Disorders (ICSD) (International Classification of Sleep Disorders, 2014).

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A Brief History of Classification Systems

Classification is the organization of knowledge for a specific purpose. While philosophers have pondered the mind for centuries, and lectures on psychology date to the sixteenth century, modern psychology dates to Wilhelm Wundt and William James in the nineteenth century (Lapointe, 1972). "Psychiatry" was first proposed as a medical specialty by Johann Christian Reil in 1808 (Marneros, 2008). While a clinical taxonomy was necessary to catalogue mental diseases, early systems of classification were limited by the technology of the day leading to nosologies that were not based on the etiology of a disease. Over 100 years later this fundamental problem remains. The classifications of mental disorders may be categorical or dimensional, but the biomarkers for mental illness await discovery.

Mental disorders may be classified for clinical, research, epidemiological, allocation of supports, or legal purposes. The process of diagnosis is a skill informed by knowledge, honed by experience, and procured by necessity. The result of the diagnostic process is communicated among clinicians through the language of a diagnostic or classification system. An ideal system for classifying medical disorders, including mental illness, does not exist (Kendler, 2009). Such an ideal system would require a measure of reliability and validity that remains elusive; however, each generation of classification brings new richness to the language of diagnosing mental disorders (Ashley, 2015). Modern diagnostic/classification systems are based on the science and technology of a point in time to guide decision-making. Science and technology are refined by epistemology, the study of knowledge. The beauty of science is that it evolves through observation, research, hypothesis, experimentation, analysis, and replication. Science allows for technological innovations which in turn prompt scientific breakthroughs, made dually from failures and successes.

The early modern classification systems of mental disorders were developed from skilled observations of a constellation of symptoms (Cuthbert, 2014; Kendler, 2016). In the nineteenth century, diagnostic possibilities were limited. The 1880 census identified seven categories of mental illness – mania, melancholia, monomania, paresis, dementia, dipsomania, and epilepsy. At the turn of the twentieth century, clinicians recognized the limitations of a nosology based solely on symptom description (Grob, 1985), a dilemma which continues today despite technological advances, especially in genetics and neuroimaging, which have refined our understanding of mental illness. Kraepelin's nosology, based in part on the work of Kahlbaum and Hecker, expanded on the observation of symptoms to include the progression of mental disorders over time (Kendler, 2016). Kraepelin published what is widely believed to be the first diagnostic system in psychiatry in 1899 in his sixth edition of *Compendium der Psychiatrie*, basing his system on both generational family histories and longitudinal research (Palm & Möller, 2011). Kraepelin's system included the demarcation between exogenous and endogenous diseases, basing the categorization of mental disorders largely on this criterion (Halter, Rolin-Kenny, & Grund, 2013). Decades later, Landis and Page (1938) proposed a "biosocial approach" to classifying mental disorders, a concept expanded by George Engel (Engel, 1977) to the biopsychosocial approach as prevailing diagnostic systems moved classification to a medical model.

While patient care and research are obvious functions diagnostic systems, early modern systems focused on epidemiology and statistical counts. In addition to research and clinical care, modern diagnostic systems inform educational programs, governmental functions, insurance practices, and legal processes. Diagnostic systems are influenced to some degree by history, theoretical approaches, culture, politics, and purpose. Telles-Correia (2018) draws on the work of Spitzer, Klein, Feighner, and others to identify the purpose of identifying mental disorders:

1. To know which diagnoses should or not be included in the classifications,
2. To separate the area of responsibility in medical systems from other societal systems,

3. To avoid dangerous medicalization of social problems,
4. To distinguish between pathological and normal,
5. To identify conditions that implicitly have a call to action to psychiatrists,
6. To identify the cases that justify societal recognition of the sick role,
7. To understand situations that may prevent legal imputability,
8. To avoid false positives, over-medicalization, unnecessary labeling, and/or wasting of resources, and
9. To define psychiatry's position as a special medical discipline (p1)

The most commonly used diagnostic systems, the DSM and ICD, are categorical. In other words, disorders are defined by symptom lists with the symptoms and requirements for rendering a specific diagnosis reached by expert consensus. This expert consensus is informed by research and clinical practice. The DSM and ICD have undergone multiple revisions and informed practice for over half a century. In the United States, the DSM-5 stipulates specific criteria for mental disorders for clinical practice and research, while the ICD-10 provides codes that are used for billing purposes. Companion diagnostic systems have been developed to facilitate the use of the DSM and ICD systems to diagnose mental disorders in persons with ID. The DM-ID serves as a companion to the DSM. The DC-LD parallels the ICD. The RDoC was proposed in 2008 as a research framework. Unlike the DSM and ICD, the RDoC is not a diagnostic system but is designed to promote the study of "mental disorders based on dimensions of observable behavior and neurobiological measures" (National Institutes of Mental Health). This transdiagnostic focus on brain/behavior relationships is meant to inform the DSM/ICD descriptive nosologies (Cuthbert, 2014). Other transdiagnostic approaches include HiTOP (Kotov et al., 2017) and the p factor (Caspi & Moffitt, 2018; Lahey et al., 2012).

The Diagnostic and Statistical Manual

The DSM is currently the foremost resource used by clinicians for diagnosis and research. Currently in its fifth edition, the DSM has evolved over time to fit the needs of clinicians, researchers, and other users, and with each revision, it has undergone considerable changes. From the psychoanalytic bent of DSM-I to the multiaxial system of DSM-III to the current categorical but multidimensionally informed approach used in DSM-5, each revision has resulted in expansion, revision, and refinement (Clark, Cuthbert, Lewis-Fernández, Narrow, & Reed, 2017; Halter et al., 2013).

The rudimentary beginnings of a diagnostic system in the United States may be traced to the 1840s when the United States Census included questions about mental illness for the first time. The 1840 Census asked a lone question about intellectual disability and mental illness, allowing classification for "idiocy or insanity" (Clark et al., 2017). This first attempt to classify mental illness proved controversial due to a greater number of African American citizens in northern states being labeled as mentally ill (Litwack, 1958). This "data" was used to justify the practice of slavery as beneficial (Haller, 1972). Over the next 40 years, the statistical classification of mental illness among institutionalized persons progressed to include mania, monomania, dipsomania, melancholia, paresis, dementia, and epilepsy without providing diagnostic criteria (Halter et al., 2013). In 1908, the US Census Bureau called for the creation of a nomenclature for mental diseases. The American Medico-Psychological Association (now called the American Psychiatric Association) and the National Committee for Mental Hygiene answered the call to classify mental illness for statistical purposes with the 1918 publication of the *Statistical Manual for the Use of Institutions for the Insane* (Shorter, 1997). Still, the United States lacked a medical nosology for mental illness.

In 1927, the New York Academy of Medicine began a movement aiming to develop and classify diseases. The National Conference on

Nomenclature of Disease met in 1928 with the APA contributing to the effort to develop a nosology. In 1933 the *Standard Classified Nomenclature of Diseases* was published with a three-page section on mental diseases. The last revision in 1942 included the *Statistical Manual for the Use of Hospitals for Mental Diseases* (APA, 1952). In parallel, the US Army, under the leadership of William Menninger, developed the War Department Technical Bulletin titled *Medical 203* which included 52 disorders grouped in five categories (Houts, 2000). Both *Medical 203* and the *Statistical Manual for the Use of Hospitals for Mental Diseases* played a prominent role in the development of DSM, with its first publication in 1952 by the American Psychiatric Association (Clark et al., 2017). The following sections will describe each version of the DSM in detail.

DSM

In 1948, the APA published the first *Diagnostic and Statistical Manual* targeting clinical diagnosis. With the publication of the DSM-I, the APA sought to integrate multiple classification systems in order to increase clinical utility and reliability (Houts, 2000). The DSM-I synthesized the *Medical 203,* ICD-6, and the *Statistical Manual for the Use of Hospitals for Mental Diseases*, including narrative descriptions of disorders with a strong psychoanalytical influence. For example, many disorders were classified as *reactions* and others as *psychoneurotic* disorders (Halter et al., 2013). The DSM-I is comprised of five sections including Diseases of the Psychobiologic Unit, Introduction to the Revised Nomenclature and Definition of Terms, Recording of Psychiatric Conditions, Statistical Reporting, and Statistical Classification of Mental Disorder. In addition, the manual has four appendices which include supplementary materials. In 130 pages, the DSM-I included over 100 disorders separated into organic brain syndromes and functional disorders (APA, 1952).

In 1959, Erwin Stengel submitted a report titled the *Classification of Mental Disorders* to the WHO. This review and critique of existing classification systems noted that there was no predominate international nosology for mental disorders or terminology detailing the national systems in use. He noted the lack of a standardized use of terms and suggested that classification systems include a glossary of terms that was neutral regarding theoretical orientation. Stengel noted that an insufficient understanding of the etiology of mental disorders would require an operational classification system. And finally, he called for a system with greater utility for outpatient treatment and child psychiatry (Stengel, 1959). A series of international meetings on the diagnosis of mental illness followed Stengel's report. In addition, questions regarding reliability of the DSM diagnoses were raised (Blashfield et al., 2014). In 1961, Thomas Szasz published *The Myth of Mental Illness*, an influential treatise against the existence of mental illness. The DSM-I established a categorical approach to the diagnosis of mental disorders, but by the mid-1960s, a revision was on the horizon.

DSM-II

DSM-II was released by the APA in 1968. The international meetings on nomenclature resulted in broad agreement between the DSM-II and ICD-8 but only minor revisions to the DSM-I (Halter et al., 2013; Millon, Krueger, & Simonsen, 2010). The second edition of the manual features an expanded focus on outpatient treatment and included nearly 200 disorders; however, the overall page number of the manual remained relatively the same, reflecting the brevity of the narrative descriptions of disorders. The use of *reaction* terminology was removed, reflecting the waning influence of psychoanalysis, although *neurosis* did remain (Blashfield et al., 2014). When the DSM-II was revised in 1974 the diagnosis of homosexuality was amended to ego-dystonic homosexuality, a change determined by a vote of the APA members attending the 1973 national conference (Halter et al., 2013).

The reliability of diagnoses was challenged by the studies of Overall and Woodard, Beck, Kendall, and Rosenhan, leading researchers at Washington University to establish the Feighner criteria (Blashfield et al., 2014). The Washington University team identified 15 categories of mental disorders that were supported by research and proposed specific diagnostic criteria for those disorders (Feighner et al., 1972). This line of research, called neo-Kraepelinian, provided the foundation for the major shift in classification found in the DSM-III.

DSM-III and DSM-III-R

The DSM-III (APA, 1980) marked the beginning of a new era in the classification of mental illness. The APA moved from diagnosis based on the psychoanalytic tradition to empirically derived categories to increase the validity and reliability of diagnosis. Building on the paradigm shift of the Feighner criteria, Spitzer, Endicott, and Robins (1978) proposed the Research Domain Criteria (RDC) to operationalize the diagnosis of mental disorders. In line with the move toward a more empirically based diagnostic system, the NIMH sponsored field trials over a 2-year period using the RCD which included over 12,000 patients and 500 clinicians (Halter et al., 2013; APA, 1980).

The organization of the DSM-III marked a revolutionary shift to a descriptive approach with specific diagnostic criteria for over 160 diagnoses, including the new diagnosis, posttraumatic stress disorder. In recognition of the continuous nature of symptoms from mental health to illness, diagnosis required distress or impairment to be present. A new method of recording diagnoses was introduced with five axes (I, mental disorder; II, personality disorders or mental retardation; III, medical disorders; IV, stressors; V, highest level of function in the past year) (APA, 1980). The radical diagnostic system of the DSM-III launched a new era of mental health research. The descriptive diagnoses with measurable symptoms led to the development of new research instruments.

DSM-III-R (1987) was released 7 years after DSM-III and included updates reflecting the growing knowledge of mental illness based on the DSM-III nosology. The DSM-III-R included a new section on sleep disorders, changes to the substance abuse category, and added or expanded diagnostic criteria for 174 diagnoses (American Psychiatric Association, 1987). In 1990, Spitzer, Endicott, and Robins published the Structured Clinical Interview for DSM-III-R (SCID), with the SCID updated with each subsequent DSM revision. Research indicated that the SCID improved diagnostic reliability. The categorical diagnoses of the DSM-III and III-R stimulated a new era in research (Blashfield et al., 2014; Kawa & Giordano, 2012) which led to next major revision of the DSM 7 years later.

DSM-IV and DSM-IV-TR

After a 6-year review process, the DSM-IV (APA, 1994) was released. Updates included revisions in the diagnostic criteria for pervasive developmental disorder, learning disorders, motor skills disorders, and communication disorders and also moved these diagnoses from Axis II to I. Additionally, DSM-IV's definition and criteria for mental retardation were revised to coincide with the American Association on Mental Retardation's (now called the American Association on Intellectual and Developmental Disabilities) definition. A new category was created called Schizophrenia and Other Psychotic Disorders which merged the categories of Schizophrenia, Delusional Disorder, and Psychotic Disorder Not Elsewhere Classified, for the purposes of clinical utility and convenience of use. In a similar vein, the new category Substance-Related Disorders merged the prior categories of Psychoactive Substance Use Disorders and Psychoactive Induced Organic Mental Disorders. Besides the introduction of these new sections into the DSM-IV, a number of new disorders, including Asperger's disorder, bipolar II disorder, and narcolepsy, were also introduced. Several disorders were eliminated from DSM-IV as well, including trans-

sexualism, which was replaced with gender identity disorder (APA, 1994). The DSM-IV represented major shifts in mental health professions and research, and more would come only 6 years later with DSM-IV-TR. A major criticism of DSM-IV and DSM-IV-TR was the conceptualization of children as, essentially, small adults, rather than viewing disorders from a developmental perspective. This problem was one of the main issues resolved by the developmental and lifespan approach of the DSM-5 (Halter et al., 2013).

DSM-5

The DSM-5 lifespan approach is reflected in the volume's basic organization, opening with childhood onset neurodevelopmental disorders and progressing to the neurocognitive disorders. Major revisions to the neurodevelopmental disorder section include the introduction of "intellectual disability (intellectual developmental disorder)" to replace "mental retardation" and the diagnostic transfiguration of Asperger's disorder, pervasive developmental disorder, and autism to the all-encompassing autism spectrum disorder (ASD). The ASD terminology reflects research supporting the continuous nature of symptoms associated with ASD but has encountered controversy due to concerns that diagnostic changes might limit service access (Burns & Matson, 2017). The DSM-5 introduces the use of a specifier to indicate intellectual or language impairment and one of three levels of support: "support, substantial support, or very substantial support" (p. 52, APA, 2013). The DSM-5 replaces *not otherwise specified categories* with *other specified or unspecified disorders* to address challenges around diagnostic specificity and abandons the axis system of documenting diagnoses. In a step toward increased agreement between the DSM and ICD, the DSM-5 adopts the WHO *Disability Assessment Schedule* (WHODAS) to replace the DSM-IV Global Assessment of Functioning (GAF) scale (APA, 2013).

The DSM-5 field trials were extensive, involving mental health professionals from diverse professions and patients from medical/academic and regular clinical settings. To explore the high comorbidity of DSM diagnoses, Cross-Cutting Symptom measures were used and are available at https://www.psychiatry.org/psychiatrists/practice/dsm/educational-resources/assessment-measures. A website was established to describe the revision process and accept comments from patients, advocacy groups, and professionals. Work groups were subject to external review. While the DSM-5 review process faced greater empirical scrutiny than ever before, the APA recognized that the:

> DSM is a medical classification of disorders and as such serves as a historically determined cognitive schema imposed on cognitive and scientific information to increase its comprehensibility and utility. (p. 10, APA, 2013)

In recognizing the limitations of the DSM-5 and opining that a transition to a dimensional approach was premature, the APA recognized the importance of the dimensional approach historically and for the future. In the DSM-5, the APA concludes that a dimensional approach "will likely supplement or supersede current categorical approaches in coming years" (p.13, APA, 2013).

The Diagnostic Manual-Intellectual Disability-2 and the Diagnostic Criteria for Psychiatric Disorders for Use with Adults with Learning Disabilities/Mental Retardation

The DM-ID was developed by the National Association for the Dually Diagnosed (NADD) to guide clinical understanding of mental illness in persons with intellectual disability (Fletcher, Barnhill, & McCarthy, 2016). As such, it is not meant to replace the DSM but to offer valuable clinical insight into the presentation of DSM diagnoses in persons with ID. The National Association for the Dually Diagnosed (NADD) first published the DM-ID in 2007 to guide the diagnosis of mental disorders in persons with intellectual disability (*Diagnostic Manual--Intellectual Disability: A Textbook of Diagnosis*

of Mental Disorders in Persons with Intellectual Disability, 2007). The DM-ID was produced with the cooperation of the American Psychiatric Association (APA) to enhance the utility of the DSM-IV-TR when treating persons with ID. The NADD also hoped to encourage research to further define mental disorders in persons with ID. The DM-ID was well-received as evidenced by a field trial in 2006 which the majority of clinicians rated the DM-ID as easy to use and helpful in making a diagnosis (Fletcher et al., 2009). After the 2013 publication of the DSM-5, a second edition of the DM-ID was published (DM-ID-2). The DM-ID-2 was developed by expert consensus with over 100 clinicians contributing (Fletcher, Barnhill, McCarthy, & Strydom, 2016).

By supporting the accurate diagnosis of mental disorders in person with ID, the DM-ID-2 promotes improved quality of life for dually diagnosed individuals. Persons with ID are at increased risk for mental disorders and may suffer greater impairment if not accurately diagnosed and treated (Buckles, Luckasson, & Keefe, 2013; Koslowski et al., 2016; Munir, 2016; Turygin, Matson, & Adams, 2014). A large British cohort study using the primary care database found that of the 33,016 persons identified with ID, 21% had a mental illness at the study's onset and 34% had a diagnosis recorded during the 14-year study. In addition, 49% were being treated with psychotropic medication at the studies onset, and nearly 2/3 were treated with psychotropic medication at some point during the study (Sheehan et al., 2015). The fact that nearly half of the persons treated with psychotropic medications did not have a documented mental disorder highlights the importance of accurately diagnosing mental illness in persons with ID. Prescribing medications without establishing a diagnosis may be due, in part, to the difficulties in diagnosing mental illness in persons with ID. Challenges can include factors related to the patient or the system of care. Patient-related factors may include the ability to understand questions, process language, and recall and express emotional states and limited ability to provide sufficient detail to queries, and challenging behaviors that interfere with the assessment or cloud the presentation of an illness. System factors may include clinician inexperience assessing person with ID, lack of assessment tools, diagnostic overshadowing, or policies that complicate access to mental health care for persons with ID (Fletcher et al., 2016; *Policy Recommendations: Addressing the Mental Health and Wellness of Individuals with Intellectual Disabilities*). Diagnostic overshadowing refers to the misattribution of symptoms to the primary disorder or a behavioral problem, leading to a failure to properly diagnose. Care must be taken to avoid conflating ID with symptoms of mental illness as this contributes to the underdiagnosis of mental illness in persons with ID.

The DM-ID-2 follows the organization of the DSM but focuses on the examiners' observation rather than patient self-report. The diagnostic chapters begin with a summary of pertinent research before offering a review of specific diagnostic criteria with a focus on considerations necessary for diagnosing mental illness in persons with ID. The literature for the specific diagnosis in persons with ID is summarized before highlighting the DM-ID criteria in a table comparing the DSM-5 criteria with the criteria for mild to moderate ID and severe to profound ID. In addition to the diagnostic category chapters, the DM-ID-2 includes chapters on *Assessment and Diagnostic Procedures* and *Behavioral Phenotypes of Neurodevelopmental Disorders* (Fletcher et al., 2016).

The DSM guides diagnosis in the United States, but in many countries, the ICD system is preferred. The Royal College of Psychiatrists published the *Diagnostic Criteria for Psychiatric Disorders for Use with Adults with Learning Disabilities/Mental Retardation* (DC-LD) in 2001 as a counterpart to the ICD-10 to assist in the diagnosis of mental disorders in persons with moderate, severe, or profound ID. ID is called "learning disability" in the United Kingdom. The DC-LD arranges diagnoses on three axes – severity, etiology of LD, and psychiatric disorder. Psychiatric disorder is further divided into developmental disorders, psychiatric illnesses, personality disorders, problem behaviors, and other disorders (Cooper, Melville, & Einfeld, 2003).

The International Classification of Diseases

The ICD is the international standard for the classification of diseases. The ICD is used by over 100 countries and has been translated into 43 languages. It is designed to *allow the systematic recording, analysis, interpretation, and comparison of mortality and morbidity data collected in different countries or areas and at different times*. The ICD is used to translate diagnoses of diseases and other health problems into an alphanumeric code, which allows storage, retrieval, and analysis of the data (Reference Guide, 2018). Additional information may be accessed at https://icd.who.int/browse11/content/refguide.ICD11_en/html/index.html.

The ICD system applies to all health conditions and since 2001 has included a Family of International Classifications including The International Classification of Functioning, Disability and Health (ICF) and the International Classification of Health Interventions (ICHI) which are openly available at the WHO website (http://www.who.int/classifications/en/). The ICD is organized with the goal of accessibility to primary care, particularly in low- and middle-income countries where there may be few mental health professionals. Compared to the DSM, the ICD supports greater freedom in making clinical diagnoses by including guidelines rather than diagnostic criteria. Requirements for symptom count and/or a duration of illness are only included if validated to differentiate a disorder (Tyrer et al., 2014).

The roots of the ICD system of classification date to 1853 when the first International Statistical Congress (later called the International Statistical Institute) invited William Farr and Marc d'Espine to develop a classification of the causes of death. After multiple revisions by Willian Farr, *The Bertillon Classification of Causes of Death* was adopted in 1893. At the urging of Florence Nightingale in her paper *Hospital Statistics* the classification of diseases was adopted. In 1900, the French government invited international delegates to the review the first *International Revision of the Bertillon* or *International List of Causes of Death*. Delegates adopted the classification and called for future revisions at 10-year intervals (Reference Guide, 2018).

The *Sixth Revision of the International Lists of Diseases and Causes of Death* in 1948 marked major changes for oversight of the *Lists* and for mental health. The *Sixth Revision* International Conference recommended that the World Health Assembly place responsibility for compiling statistics under the United Nations' World Health Organization. Later that year the World Health Assembly adopted the resolution and in 1949 released the *Manual of the International Statistical Classification of Diseases, Injuries, and Causes of Death* (ICD-6) (WHO, 1949). For the first time in ICD history, mental illness was recognized in a separate chapter. Chapter 5 was titled *Mental, Psychoneurotic, and Personality Disorders*. The ICD-6 aims of providing a psychiatric classification scheme to facilitate clinical, statistical, and research goals were met with criticisms including the failure to conduct a comprehensive survey to inform the diagnoses selected, the use of "mental" as synonymous with "psychosis," and that the 26 categories of the 10-page chapter, *Mental, Psychoneurotic, and Personality Disorders*, was "too complicated and unwieldy" (p. 607) (Stengel, 1959).

There were no substantial changes to the chapter in ICD-7; however, collaboration between the American Psychiatric Association and the WHO resulted in broad agreement between the ICD-8 (WHO, 1967) and the DSM-II (APA, 1968). "Mental retardation" was first included as a mental disorder in ICD-8 (Bertelli, Munir, Harris, & Salvador-Carulla, 2016). In 1974 the WHO published a *Glossary of Mental Disorders and Guide to their Classification* to address concerns documented by Stengel (1959) that terminology varied across countries. The major change in the ICD-9 was the incorporation of the glossary as narrative descriptions included with categories (WHO, 1975). The *International Classification of Diseases, Clinical Modification* (ICD-9-CM) was used for billing purposes in the United States until October 2015 when the United States shifted to the ICD-10-CM for federal billing purposes. ICD codes are commonly used for

health-care billing with 70% of expenditures based on ICD codes (Reference Guide, 2018).

The ICD-10 was first used in 1994 (Reference Guide, 2018). In recognition of the importance of screening for mental illness and developmental disorders, the ICD-10 introduced Z codes that allow for billing for screening encounters. Chapter 5 *Mental and Behavioural Disorders* (coded as F00 -F99) is published as two stand-alone versions – *The ICD-10 Classification of Mental and Behavioural Disorders: Clinical Descriptions and Diagnostic Guidelines* (The Blue Book) (WHO, 1992) and *The ICD-10 Classification of Mental and Behavioural Disorders: Diagnostic Criteria for Research* (The Green Book) (WHO, 1993).

The ICD-11 was approved by the World Health Assembly 194 member states in 2015. Chapter 6 of the ICD-11 addresses *Mental, Behavioural or Neurodevelopmental Disorders*. A major change for the ICD-11 is the renaming of "mental retardation" to "disorders of intellectual development" (WHO, 2018). While the DSM-5 diagnosis of intellectual disability/intellectual developmental disorder is equivalent to the ICD -11 diagnosis of disorders of intellectual development (DID), the ICD-11 diagnosis of DID and International Classification of Functioning, Disability and Health diagnosis of ID have different meanings and uses. The ICD system is used for reporting diseases and tracking a variety of epidemiological measures. The ICD diagnosis DID indicate a health condition defined as:

> a group of etiologically diverse conditions originating during the developmental period characterized by significantly below average intellectual functioning and adaptive behavior that are approximately two or more standard deviations below the mean, based on appropriately normed, individually administered standardized tests. (p. 2, WHO, 2018)

DID may be classified as mild, moderate, severe, profound, provisional, or unspecified with an additional code for a known etiology (WHO, 2018). The IFC *Disability and Health* guides the quantification of health and disability for persons or populations. It is important that the IFC perspective recognizes disability as the result of impaired health. As such, disability may be experienced by anyone with a health condition. In the language of the IFC, intellectual functioning is 1 of 11 mental functions which may be assessed for impairment or disability (WHO, 2017). The WHO Disability Assessment Schedule (WHODAS 2.0) is used in conjunction with the ICF to assess health and disability (Ustün et al., 2010). The WHO offers a range of additional treatment, research, and policy tools at http://www.who.int/mental_health/en/.

The Research Domain Criteria

The RDoC was proposed in the 2008 NIMH Strategic Plan as a transdiagnostic, research framework to inform rather than replace the DSM/ICD diagnostic systems (Insel et al., 2010). The RDoC framework has been designed with a focus on neuroscience and behavior rather than symptoms or diagnosis, with the goal of translating the science into new ways to identify and treat mental illness (Cuthbert & Insel, 2013). The RDoC is intended to refine diagnostic validity and reliability as well as improve treatment validity (Lilienfeld & Treadway, 2016). Technological advances in molecular biology led the National Research Council to propose this "new taxonomy of human disease based on molecular biology" (Desmond-Hellman & Sawyers, 2011). The RDoC provides the mental health response to the National Research Council's call for "precision medicine" (Cuthbert, 2014).

The NIMH RDoC project was tasked with creating a data-driven framework for the use of neuroscience to study the basic behavioral components of mental illness. A further goal was to approach these behavioral components as a spectrum. For example, the clinical symptom "sleep disturbance" is explored across the 24-hour sleep-wake continuum (NIMH RDoC matrix). (Fig. 2.1). In addition, the RDoC endeavors to define each behavior across its presentations (normal to abnormal) while accounting for neurodevelopment and environmental influences. The importance and necessity of developing new

Cognitive Systems

Construct/Subconstruct		Genes Notice	Molecules	Cells	Circuits	Physiology	Behavior	Self-Report	Paradigms
Attention			Elements	Elements	Elements	Elements	Elements		Elements
Perception	Visual Perception		Elements	Elements	Elements	Elements	Elements	Elements	Elements
	Auditory Perception		Elements	Elements	Elements	Elements	Elements	Elements	Elements
	Olfactory/Somatosensory/Multimodal/Perception								Elements
Declarative Memory			Elements	Elements	Elements	Elements	Elements	Elements	Elements
Language					Elements	Elements	Elements	Elements	Elements
Cognitive Control	Goal Selection; Updating, Representation, and Maintenance ⇒ Focus 1 of 2 ⇒ Goal Selection				Elements			Elements	Elements
	Goal Selection; Updating, Representation, and Maintenance ⇒ Focus 2 of 2 ⇒ Updating, Representation, and Maintenance		Elements	Elements	Elements	Elements	Elements	Elements	Elements
	Response Selection; Inhibition/Suppression ⇒ Focus 1 of 2 ⇒ Response Selection		Elements	Elements	Elements	Elements	Elements	Elements	Elements
	Response Selection; Inhibition/Suppression ⇒ Focus 2 of 2 ⇒ Inhibition/Suppression		Elements	Elements	Elements	Elements	Elements	Elements	Elements
Construct/Subconstruct		**Genes Notice**	**Molecules**	**Cells**	**Circuits**	**Physiology**	**Behavior**	**Self-Report**	**Paradigms**
	Performance Monitoring		Elements		Elements	Elements	Elements	Elements	Elements
Working Memory	Active Maintenance		Elements	Elements	Elements	Elements			Elements
	Flexible Updating		Elements	Elements	Elements	Elements			Elements
	Limited Capacity		Elements		Elements	Elements			Elements
	Interference Control		Elements	Elements	Elements	Elements			Elements

Fig. 2.1 The Research Domain Criteria (RDoC) matrix

ways to measure the components was also recognized (Cuthbert & Insel, 2013). The resulting RDoC framework is conceptualized as a matrix or table with rows detailing the five *domains* (negative valence systems, positive valence systems, cognitive systems, systems for social processes, and arousal/modulatory systems) and columns representing eight *units of analysis* (genes, molecules, cells, circuits, physiology, behavior, self-report, and paradigms). Domains are further divided into *constructs*. *Elements* which may be the focus of study are noted in the cells of the matrix. For example, the domain of cognitive systems lists constructs including working memory which is further divided into subconstructs. Research into a subconstruct may include an independent variable based on an element of a unit of analysis, for example, dopamine (element) under molecules (unit of analysis). A picture is worth an infinite number of words when describing the RDoC matrix. The reader is referred to the NIMH website to explore the interactive RDoC matrix.

As the NIMH RDoC was being mapped out in 2009, the NIH announced the Human Connectome Project. This groundbreaking project aims to "construct a map [called a connectome] of the complete structural and functional neural connections" ("Human Connectome Project: About,"). The original project has expanded to encompass mapping of the brain from formation in utero and across the lifespan. The Connectome Coordination Facility (CCF) coordinates the lifespan studies and maintains the connectome database ("Human Connectome Project: About,") providing important information for RDoC research.

Since the introduction of the RDoC, the NIMH has provided funding opportunities to support research using the framework. The most researched domains have been cognitive systems, negative valence systems, and positive valence systems (Carcone & Ruocco, 2017). Research networks have begun to redefine how psychotic symptoms are categorized. The Bipolar-Schizophrenia Network on Intermediate Phenotypes (B-SNIP) consortium has identified three neurobiologically distinct psychosis biotypes (Clementz et al., 2016). Standard psychometric instruments such as the Minnesota Multiphasic Personality Inventory-2-Restructured Form have incorporated RDoC physiology (units of analysis) into psychopathology research (McCord, Achee, Cannon, Harrop,

& Poynter, 2017). New methods are emerging to assimilate data across units of analysis. Psychoneurometrics provides a way to link neurobiology and psychological studies (Patrick et al., 2013; Patrick, Durbin, & Moser, 2012). Patrick et al. (2012) define psychoneurometrics as the "systematic development of neurobiologically based trait measures using psychological (i.e., traditional psychometric) phenotypes as referents" (p.1048). The RDoC has garnered international acceptance (Lupien et al., 2017; Weine, Langenecker, & Arenliu, 2018). The transdiagnostic approach was considered in the design of the Signature Bank, a mental health database, at the Institut Universitaire en Santé Mentale de Montréal (IUSMM). The Signature Bank allows international access to data collected from thousands of persons seeking mental health treatment (access at: http://www.iusmm.ca/research/signature-bank.html) (Lupien et al., 2017).

Alexopoulos and Arean (2014) have proposed a five-step model for streamlining psychotherapy using RDoC principles in the 9-week *Engage* therapy program. *Engage* targets constructs under the domains of positive valence systems, negative valence systems, arousal and regulatory system, and cognitive systems in this intervention for depression in the elderly. The 12-week *Training for Awareness, Resilience, and Action* (TARA) targets constructs under the domains negative valence systems, arousal and regulatory systems, positive valence systems, cognitive systems, and social processes to treat adolescent depression (Henje Blom et al., 2014). Fonagy and Luyten (2018) have formulated a RDoC approach for management of conduct problems in youth. The NIMH RDoC provides a framework for a new generation of basic science and mental health treatment research that will influence future diagnostic systems (Hershenberg & Goldfried, 2015).

The transition from the DSM/ICD categorical diagnostic system to the dimensional RDoC framework has met with some challenges and criticism. Educating scientists and clinicians and linking RDoC research to clinical practice are challenges (Kozak & Cuthbert, 2016). It has been argued that the RDoC focuses on brain circuits at the expense of the person (Parnas, 2014). Lilienfeld and Treadway (2016) have identified conceptual, methodological, and logistical issues faced by the RDoC, offering recommendations for addressing concerns. They highlight the importance of identifying the parameters for RDoC success and a mechanism for comparing RDoC and DSM/ICD performance while noting areas where measurement and expression of neurobiology must be refined. Concerns regarding RDoC use of endophenotypes have been raised. Endophenotypes are measurable biomarkers that lie between the genotype (genetic loci responsible for the illness) and the phenotype or exophenotype (behavioral expression of the illness) of a mental illness. It is hypothesized that both the illness and the biomarker are related to the same genetic loci. Many of the elements included under the units of analysis are biomarkers that may function as endotypes, particularly the elements under molecules and physiology (Glahn, Knowles, & Pearlson, 2016; Gottesman & Gould, 2003). It has long been a goal of mental health nosology to identify mental disorders by their biological etiology. To establish such an etiology, the link between endophenotypes and exophenotypes must be carefully considered as the RDoC moves forward (Jablensky & Waters, 2014; Kirmayer & Crafa, 2014; Lilienfeld & Treadway, 2016). The RDoC aims to define "fundamental components to improve understanding of what is typical versus pathological" (Cuthbert & Insel, 2013 p. 4) which authors acknowledge may require a *super-normal* (p. 5) control group. Where persons with ID would fit in this spectrum from normal to abnormal is unclear. Because the etiology of ID is diverse, the RDoC approach is likely to provide important information about specific etiologies. What this will mean for the study of ID as a mental disorder remains unclear.

The process behind the development of the RDoC has been carefully archived on the NIMH website. This is especially important as the RDoC is designed to be a dynamic nosology with the ability to incorporate new data rapidly (Cuthbert, 2014). Significant changes to the original matrix have already occurred including proposals to strengthen the neurodevelopmental trajectory of

mental illnesses (Mittal & Wakschlag, 2017), incorporate a motor system domain (Garvey and Cuthbert, 2017), add a gut microbiome signature (Kelly, JR et al., 2016), remove specific gene references (NIMH website), and reorganize the positive valence system domain (RDoC Changes to the Matrix May 17, 2018). The NIMH website provides the most up-to-date information regarding the RDoC and includes helpful webcasts, the RDoC database (RDoCdb), and information for incorporating RDoC into research (Sanislow, 2016).

The Hierarchical Taxonomy of Psychopathology

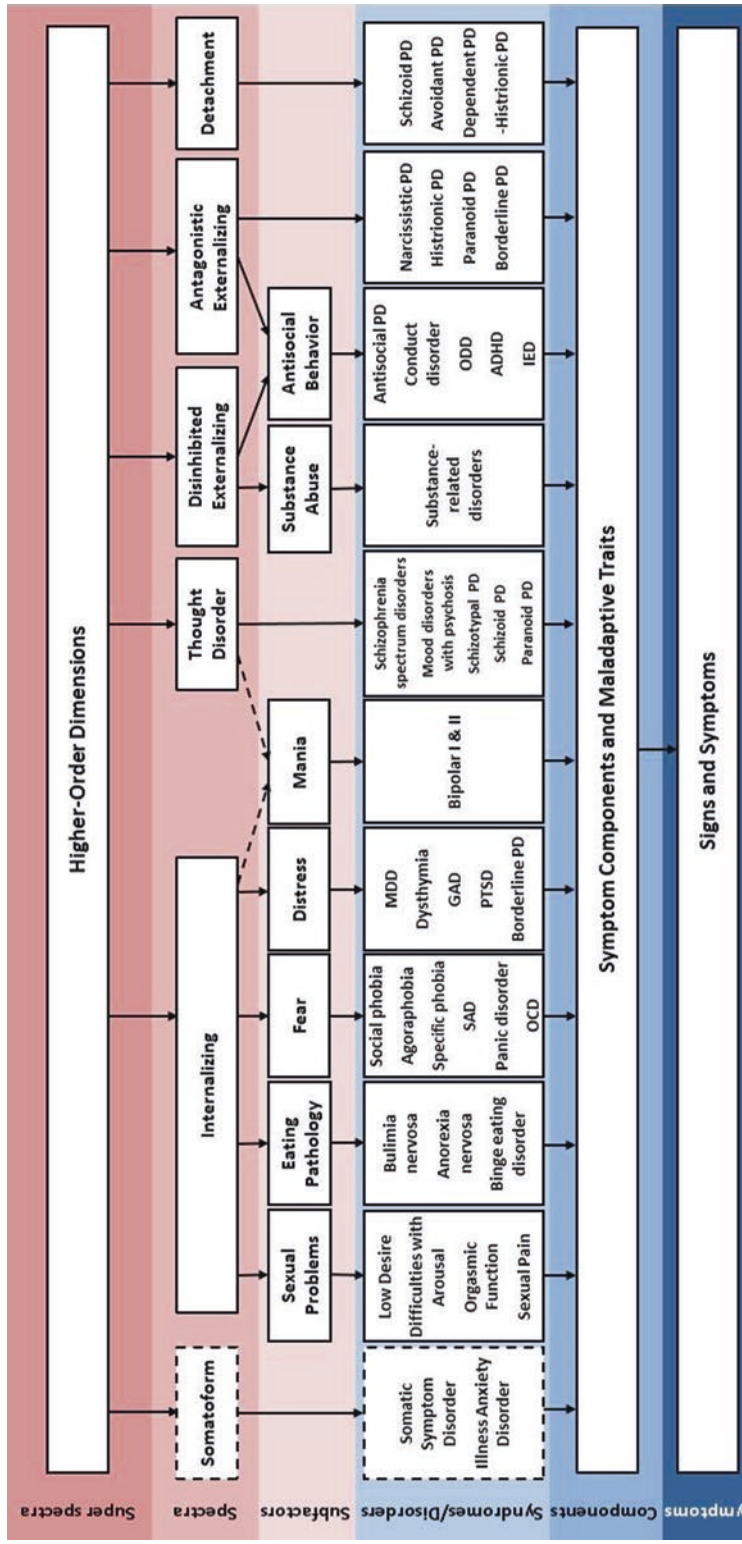
The HiTOP consortium (https://medicine.stonybrookmedicine.edu/HITOP) has proposed a dimensional, hierarchical taxonomy of psychopathology developed empirically. In this classification, behaviors are viewed as a continuum and can be assessed using quantitative measures (Kotov et al., 2017). Statistical analysis of the data allows the identification of dimensional syndromes. A foundation of this approach is the landmark research of Achenbach (1966) in classifying behaviors as "internalizing" and "externalizing," leading to the widely used Achenbach System of Empirically Based Assessment (ASEBA) which includes the Child Behavior Checklist (CBCL) (Achenbach, 2009). Additional foundations include dimensional approaches to the study of personality and thought disorders. Keyes et al. (2020) characterized a subdomain of the internalizing domain labeled the *thought disorder factor*. Aspects of personality have also been found to fit a dimensional model. The five-factor model (FFM) of personality (O'Connor, 2005) has been expanded to a FFM of personality disorder (FFM-PD) (Widiger, Lyman, Miller & Oltmans, 2012).

HiTOP identifies *Higher-Order Dimensions* or "super-spectra" which are divided into six *spectra*: internalizing, disinhibited externalizing, antagonistic externalizing, thought disorder, detachment, and, a proposed sixth category, somatoform. *Spectra* are divided into *subfactors* which are further divided into *syndromes* or disorders. The higher-order dimensions include the p factor (Lahey et al., 2012; Caspi 2014, Caspi & Moffitt, 2018). The p factor, named to mirror the general intelligence factor g, has been hypothesized to predict a general tendency to develop psychopathology. Confirmatory factor analysis of 1000 participants from the Dunedin Multidisciplinary Health and Development Study, a longitudinal cohort study, empirically validated factor p (Caspi et al., 2014). More recently, factor p has been linked to comorbidity, symptom severity, and the persistence of mental disorders (Caspi & Moffitt, 2018) (Fig. 2.2).

An important feature of HiTOP is this crossover with traditional DSM/ICD diagnostic categories. This fits with the goal that HiTOP function as a clinical and research taxonomy that might inform traditional nosologies as well as the RDoC (Kotov et al., 2017). HiTOP research has utilized over a dozen dimensional measures which can be accessed at (https://medicine.stonybrookmedicine.edu/system/files/HiTOP%20measures%20online.pdf). A single measure encompassing the model has not been developed. Additional research is needed on all levels of the hierarchy and across the lifespan. Notably, this model does not currently include the neurodevelopmental spectrum (Krueger et al., 2018).

Intellectual Disability: Definition, Classification, and Systems of Supports

The American Association of Intellectual and Developmental Disabilities (AAIDD) has been the leading organization supporting the rights of persons with intellectual and developmental disabilities for nearly 150 years. The AAIDD publishes the *Intellectual Disability: Definition, Classification, and Systems of Support* (The Manual) which identifies person with ID as having "significant limitations both in intellectual functioning and adaptive behavior as expressed in conceptual, social, and practical adaptive



https://renaissance.stonybrookmedicine.edu/HITOP/AboutHiTOP

Fig. 2.2 The Hierarchical Classification of Diseases (HiTOP)

Table 2.1 Comparison of the classification of intellectual disability by the American Psychiatric Association, the World Health Organization, and the American Association on Intellectual and Developmental Disabilities

Definitions of intellectual disability		
DSM-5 Intellectual disability (Intellectual developmental disorder)	ICD-11 Disorders of intellectual development	AAIDD Intellectual disability
A disorder characterized by deficits in both *intellectual functioning* and *adaptive functioning* with intellectual deficits confirmed by individually administered standardized testing and clinical assessment and adaptive functioning that does not meet developmental and sociocultural standards across multiple settings. *Disability begins during the developmental period.*	A group of etiologically diverse conditions originating during the developmental period characterized by significantly below average *intellectual functioning* and *adaptive behavior* that are approximately two or more standard deviations below the mean, based on appropriately normed, individually administered standardized tests. Where appropriately normed and standardized tests are not available, diagnosis of disorders of intellectual development requires greater reliance on clinical judgment based on appropriate assessment of comparable behavioral indicators.	A disability characterized by significant limitations both in *intellectual functioning* and in *adaptive behavior*, which covers many everyday social and practical skills. This disability originates *before the age of 18*. The AAIDD stresses that additional factors must be taken into account, such as the community environment typical of the individual's peers and culture. Professionals should also consider linguistic diversity and cultural differences in the way people communicate, move, and behave.
APA, 2013	WHO, 2018	AAIDD, 2010

skills" with onset before age 18 (Schalock et al., 2010). The AAIDD definition of intellectual disability has informed DSM and ICD intellectual disability diagnostic definitions (Tassé, Luckasson, & Nygren, 2013).

In 1910, the AAIDD Committee on Classification of the Feebleminded published the first definition and classification manual on ID. In addition to promoting policy and research, the AAIDD has developed instruments for assessing adaptive behavior (Tasse, et al., 2016) and person-centered planning (Thompson, Schalock, Tassé, et al., 2017). The 11th edition of The Manual was published in 2010, officially introducing "intellectual disability" to replace "mental retardation." Shortly after The Manual was published, Rosa's Law (Pub. L. 111-256) was enacted, replacing all references to mental retardation in Federal law with intellectual disability.

The AAIDD definition of ID is based on functionality in contrast with the DSM/ICD classifications of IDD and DID which take a medical approach. The lack of consensus in definition reflects the different missions of the AAIDD, APA, and WHO. The AAIDD shifts the focus to support which aligns with the promotion of human rights. The APA produces the DSM with the goal of facilitating diagnosis, treatment, and research (APA, 2013). While the WHO shares these APA goals, the ICD is designed to be used in countries with diverse cultures and economic resources. The ICD allows the diagnosis of DID on the basis of clinical judgment if testing is not readily accessible. (Table 2.1).

Additional Classification Resources

Diagnostic Classification of Mental Health and Developmental Disorders of Infancy and Early Childhood

In 1994, Zero to Three (formerly the National Center for Clinical Infant Programs) published the *Diagnostic Classification of Mental Health and Developmental Disorders of Infancy and Early Childhood* (DC:0-3) to support developmentally focused diagnosis and research. The third edition, the DC:0-5, expands the upper age to 5 years. The DC:0-5 was developed as a companion to the DSM/ICD classifications. The DC:0-5 was developed by experts representing

psychiatry, psychology, pediatrics, nursing, social work, and counseling (Zeanah et al., 2016). The DC:0-5 provides a crosswalk to the DSM and ICD.

The DC:0-5 includes a five axis classification system to document *clinical disorders* (I), *relational context* (II), *physical health conditions and considerations* (III), *psychosocial stressors* (IV), and *developmental competence* (V). Axes II, III, and IV include tables for recording *dimensions of caregiving,* the *infant/young child's contribution to the relationship*, *dimensions of the caregiving environment*, *stressors*, *and competencies*. There is an appendix with key milestones. Diagnostic algorithms, detailed narrative descriptions, and DSM/ICD crosswalk for diagnoses are featured for each disorder. The DC:0-5 addresses syndromes not captured by the DSM/ICD, including early atypical autism spectrum disorder and crying disorder. The expanded section on sensory processing disorders provides guidance for sensory issues that may be at the root of challenging child behaviors. The section on *Trauma, Stress, and Deprivation Disorders* provides an empirically based diagnostic framework as a basis for important clinical formulation and treatment interventions (ZERO TO THREE, 2016).

Psychodynamic Diagnostic Manual

The *Psychodynamic Diagnostic Manual* (PDM) was the response of the psychoanalytic community to the categorical, descriptive approach of the DSM-III. Five psychoanalytic organizations contributed to the ideographically oriented PDM. The PDM is designed to promote "thoughtful individual case formulation" (p.112, McWilliams N, 2011); as such, it is "a taxonomy of people" (p. 13, APA &APO, 2006). The PDM is formulated to expand on the DSM/ICD categorical descriptions with a focus "on the full range of mental functioning" (p. 2). After extensive revisions, the PDM-2 was published in 2017 (ICDLD-PDM, 2017).

The PDM-2 addresses the lifespan with five sections (Adulthood, Adolescence, Childhood, Infancy and Early Childhood, and Later Life). A sixth section is devoted to assessment and clinical illustrations. PDM-2 diagnoses are listed on three axes *Personality Syndromes* (P), *Profile for Mental Functioning* (M), and *Symptoms Patterns: The Subjective Experience* (S) (ICDLD-PDM, 2017). Part IV, *Infancy and Early Childhood*, classifies mental health diagnosis on Axis I and contributing factors on Axes II through V, capturing *functional emotional development capacities* (II), *regulatory-sensory processing capacities* (III), *relational patterns and disorders* (IV), and *other medical and neurological disorders* (V) (p. 827). The PDM-2 includes autism and Asperger syndrome as developmental disorders (Lingiardi & McWilliams, 2017).

PDM-2 diagnoses are presented as a narrative description followed by a clinical illustration. In alignment with the PDM-2 goals of supporting clinical and research endeavors, a section called *relevant assessment tools* accompanies the descriptions. An appendix with age-group-specific psychodiagnostic charts is included for documentation. The charts allow DSM/ICD/DC:0-5 diagnoses to be incorporated into the psychodiagnostic formulation and captures the range of axes presentation on Likert scales (Lingiardi & McWilliams, 2017). The PDM-2 does not include ID as a specific diagnosis but recognizes the importance of intellectual and adaptive assessments and offers a rich framework for capturing a person's unique needs.

International Classification of Sleep Disorders

While the DSM and ICD address sleep disorders, the American Academy of Sleep Medicine's International Classification of Sleep Disorders (ICSD) is considered the authoritative diagnostic system. (See Chap. 26.)

Precision Medicine

In 2011 the National Institutes of Health charged the National Research Council with exploring:

> a New Taxonomy of human disease based on molecular biology [with the goal of] understand[ing] how a person's genetics, environment, and lifestyle can help determine the best

> approach to prevent or treat disease. (p.1, "What is the Precision Medicine Initiative?," 2018)

Precision medicine supports the use of large data sets, big data, and advanced analytical techniques to understand the needs of subpopulations and promote specific treatments. To support the development of this new taxonomy, the federal government has funded the Precision Medicine Initiative and a database called the Precision Medicine Cohort (Precision Medicine, 2018 ghr.nlm.nih.gov) operationalized as the All of Us Research Program (allofus.nih.gov, joinallofus.org).

The need for research regarding general health and mental illness in persons with ID is widely recognized (Krahn, Walker, & Correa-De-Araujo 2015), and applying the principles of precision medicine to the health-care needs of persons with ID is critical (Sabatello, 2018). To this end, NIH has established a registry called DS-Connect® to support better understanding of the health-care needs of persons with Down syndrome (dsconnect.nih.gov/). The National Fragile X Foundation has established the NFXF Biobank™ to promote the identification of biomarkers (https://fragilex.org/our-research/nfxf/biobank/). Conditions which disproportionately occur in person with ID such as epilepsy (Zuberi & Brunklaus, 2018) and gastrointestinal disorders (Kuntz & Gilbert, 2017) are the focus of precision medicine research. As precision medicine explores specific health conditions, it is critical that subpopulations with ID be included.

The Legal Classification of Mental Disorders

The DSM-5 includes a *Cautionary Statement for Forensic Use* which highlights the clinical orientation of the DSM and alerts "to dangers that arise because of the imperfect fit between the questions of ultimate concern to the law and information contained in a clinical diagnosis" (p 25). The clinician who ventures into the legal arena must heed this admonition and be prepared to adopt the language of the court; always keeping in mind that DSM diagnoses are not synonymous with the legal definitions. As examples of legal terminology, "mentally ill person," "insanity," and "mental disease or defect" are briefly reviewed before introducing the landmark US Supreme Court rulings on the definition of intellectual disability in death penalty cases.

While mental health and legal classification systems are dynamic systems, their basis, evolution and purpose are quite different. Clinical diagnostic systems value empirical knowledge and provide a basis for treatment to alleviate suffering. Legal systems are based on statutes and precedents and are designed to promote justice. Because legal definitions vary by jurisdiction and legal context, the clinician should consult with an attorney to be clear on applicable law before addressing a legal question. In general, statutes defining mental illness or disability typically describe the impact the disorder has on an individual's understanding of reality or functioning in a specific legal context. For example, when considering commitment to a treatment facility, Louisiana defines a "mentally ill person" as "any person with a psychiatric disorder which has substantial adverse effects on his ability to function and who requires care and treatment" [LA Rev Stat § 28:2 (14)]. Note that under this definition, a person may have a DSM diagnosis but would not be classified as a "mentally ill person" under the law if the disorder did not render the person unable to function to the point of grave disability. In contrast to civil commitment, the definition used to determine "insanity" in a criminal proceeding requires a "mental disease or mental defect" render "the offender incapable of distinguishing between right and wrong" [LA Rev Stat § 14:14]. The "mental disease or mental defect" language dates to precedent set by English Common Law. In modern contexts it is defined by state law. In general, the law considers a mental disease a disorder that may respond to treatment while a mental defect represents a stable condition.

In the 2002 landmark case *Atkins v. Virginia*, the US Supreme Court ruled it unconstitutional to execute persons with intellectual disability. The

decision of the Court did not define intellectual disability but opted to leave this definition to individual states. Twelve years later in the case *Hall v. Florida*, the US Supreme Court ruled that states must base the legal determination of intellectual disability in capital cases on clinical diagnostic standards and must consider a test's standard deviation for error in that determination (*Hall* 572 U.S. ___). The US Supreme Court reaffirmed the importance of reliance on prevailing professional guidelines in the assessment of intellectual and adaptive functioning in *Moore v. Texas* (2017) (Bundrick & Martinez, 2017). The US Supreme Court considered the written testimony of professional organizations including the American Psychological Association, American Psychiatric Association, and American Association on Intellectual and Developmental Disabilities when deliberating these landmark decisions.

These brief paragraphs on legal classifications of mental disorders and intellectual disability are introductory. The intention is to alert the reader that clinical classification systems are preparation for clinical work. Clinicians interested in legal matters are advised to consider forensic training.

Conclusions

Names and nosology make a tangible difference in people's lives on many levels. From the systems that deliver mental health care to the systems that determine supports and service eligibility to personal identity and responsibility, names and nosology matter.

The significance of naming is illustrated by the evolution of terminology for intellectual disability. As science defined intellect and society stigmatized specific names, *intellectual disability* replaced earlier terms. Educational, legal, and statutory terminology describing intellectual disability has been influenced by official classification systems. The 2010 passage of the Twenty-First Century Communications and Video Accessibility Act or Rosa's Law replaced the term *mental retardation* with *intellectual disability* and is an example of society's influence on terminology. The law's namesake, Rosa Marcellino, was aided by her family to change the law in her home state of Maryland leading to changing US law. Rosa's brother, Nick, spoke of his sister with Down syndrome stating:

> What you call people is how you treat them. If we change the words, maybe it will be the start of a new attitude towards people with disabilities. ("Remarks by the President at the Signing of the 21st Century Communications and Video Accessibility Act of 2010," 2010)

The classification of mental disorders is a dynamic process driven by science and technology and influenced by society. Future classification systems will borrow from the categorical systems of past and the emerging multidimensional systems to offer new dimensions in diagnosis, holding promise for the advancement of research and patient care.

References

Achenbach, T. M. (1966). The classification of children's psychiatric symptoms: a factor-analytic study. *Psychological Monographs: general and applied, 80*(7), 1

Achenbach, T. M. (2009). The Achenbach system of empirically based assessment (ASEBA): Development, findings, theory, and applications. University of Vermont, Research Center for Children, Youth, & Families.

Alexopoulos, G. S., & Arean, P. (2014). A model for streamlining psychotherapy in the RDoC era: The example of 'Engage'. *Molecular Psychiatry, 19*(1), 14–19. https://doi.org/10.1038/mp.2013.150

American Academy of Sleep Medicine. (2014). *International classification of sleep disorders* (3rd ed.). Darien, IL: American Academy of Sleep Medicine.

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

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

American Psychiatric Association. (1980). *Diagnostic and statistical manual of mental disorders*, (2nd ed.). American Psychiatric Association.

American Psychiatric Association. (1987). *Diagnostic and Statistical Manual of Mental Health Disorders* (DSM-III-R). American Psychiatric Association.

American Psychiatric Association. (1994). *Diagnostic and Statistical Manual of Mental Health Disorders* (DSM-IV). American Psychiatric Association.

American Psychiatric Association. (2013). *Diagnostic and statistical manual of mental disorders* (DSM-5®). American Psychiatric Pub.

American Psychoanalytic Association, & Alliance of Psychoanalytic Organizations. (2006). Psychodynamic diagnostic manual (PDM). Interdisciplinary Council on.

Ashley, E. A. (2015). The precision medicine initiative: A new national effort. *JAMA, 313*(21), 2119–2120. https://doi.org/10.1001/jama.2015.3595

Bertelli, M. O., Munir, K., Harris, J., & Salvador-Carulla, L. (2016). "Intellectual developmental disorders": Reflections on the international consensus document for redefining "mental retardation-intellectual disability" in ICD-11. *Advances in Mental Health and Intellectual Disabilities, 10*(1), 36–58. https://doi.org/10.1108/amhid-10-2015-0050

Blashfield, R. K., Keeley, J. W., Flanagan, E. H., & Miles, S. R. (2014). The cycle of classification: DSM-I through DSM-5. *Annual Review of Clinical Psychology, 10*(1), 25–51.

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. https://doi.org/10.1080/19315864.2011.651682

Bundrick, J., & Martinez, R. (2017). Determination of intellectual disability in death penalty cases. *Journal of the American Academy of Psychiatry and the Law Online, 45*(4), 505.

Burns, C. O., & Matson, J. L. (2017). An evaluation of the clinical application of the DSM-5 for the diagnosis of autism spectrum disorder. *Expert Review of Neurotherapeutics, 17*(9), 909–917. https://doi.org/10.1080/14737175.2017.1351301

Carcone, D., & Ruocco, A. C. (2017). Six years of research on the National Institute of Mental Health's Research Domain Criteria (RDoC) initiative: A systematic review. *Frontiers in Cellular Neuroscience, 11*, 46–46. https://doi.org/10.3389/fncel.2017.00046

Caspi, A., Houts, R. M., Belsky, D. W., Goldman-Mellor, S. J., Harrington, H., Israel, S., ... & Moffitt, T. E. (2014). The p factor: one general psychopathology factor in the structure of psychiatric disorders?. *Clinical Psychological Science, 2*(2), 119–137.

Caspi, A., & Moffitt, T. E. (2018). All for One and one for all: Mental disorders in one dimension. *The American Journal of Psychiatry, 175*(9), 831–844. https://doi.org/10.1176/appi.ajp.2018.17121383

Clark, L. A., Cuthbert, B., Lewis-Fernández, R., Narrow, W. E., & Reed, G. M. (2017). Three approaches to understanding and classifying mental disorder: ICD-11, DSM-5, and the National Institute of Mental Health's Research Domain Criteria (RDoC). *Psychological Science in the Public Interest, 18*(2), 72–145. https://doi.org/10.1177/1529100617727266

Clementz, B. A., Sweeney, J. A., Hamm, J. P., Ivleva, E. I., Ethridge, L. E., Pearlson, G. D., … Tamminga, C. A. (2016). Identification of distinct psychosis biotypes using brain-based biomarkers. *The American Journal of Psychiatry, 173*(4), 373–384. https://doi.org/10.1176/appi.ajp.2015.14091200

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 Disability Research : JIDR, 47*(Suppl 1), 3–15. https://doi.org/10.1046/j.1365-2788.47.s1.2.x

Cuthbert, B. N. (2014). The RDoC framework: Facilitating transition from ICD/DSM to dimensional approaches that integrate neuroscience and psychopathology. *World Psychiatry, 13*(1), 28–35. https://doi.org/10.1002/wps.20087

Cuthbert, B. N., & Insel, T. R. (2013). Toward the future of psychiatric diagnosis: The seven pillars of RDoC. *BMC Medicine, 11*, 126–126. https://doi.org/10.1186/1741-7015-11-126

Desmond-Hellman, & Sawyers. (2011). *Toward precision medicine: Building a knowledge network for biomedical research and a new taxonomy of disease*. In. Retrieved from https://www.ncbi.nlm.nih.gov/books/NBK92146/

Diagnostic manual--intellectual disability: A textbook of diagnosis of mental disorders in persons with intellectual disability. (2007). Kingston, NY: National Association for the Dually Diagnosed.

Engel, G. L. (1977). The need for a new medical model: A challenge for biomedicine. *Science, 196*(4286), 129–136.

Feighner, J. P., Robins, E., Guze, S. B., Woodruff, R. A., Winokur, G., & Munoz, R. (1972). Diagnostic criteria for use in psychiatric research. *Archives of General Psychiatry, 26*(1), 57–63.

Fletcher, R., Barnhill, J., & McCarthy, S.-A. (2016). *Diagnostic manual-intellectual disability (DM-ID): A clinical guide for diagnosis of mental disorders in persons with intellectual disability* (2nd ed.). Kingston, NY: NADD.

Fletcher, R. J., Barnhill, J., McCarthy, J., & Strydom, A. (2016). From DSM to DM-ID. *Journal of Mental Health Research in Intellectual Disabilities, 9*(3), 189–204. https://doi.org/10.1080/19315864.2016.1185324

Fletcher, R. J., Havercamp, S. M., Ruedrich, S. L., Benson, B. A., Barnhill, L. J., Cooper, S. A., & Stavrakaki, C. (2009). Clinical usefulness of the diagnostic manual-intellectual disability for mental disorders in persons with intellectual disability: Results from a brief field survey. *The Journal of Clinical Psychiatry, 70*(7), 967–974. https://doi.org/10.4088/jcp.08m04429

Fonagy, P., & Luyten, P. (2018). Conduct problems in youth and the RDoC approach: A developmental, evolutionary-based view. *Clinical Psychology Review, 64*, 57–76. https://doi.org/10.1016/j.cpr.2017.08.010

Garvey, M. A., & Cuthbert, B. N. (2017). Developing a motor systems domain for the NIMH RDoC program

Glahn, D. C., Knowles, E. E., & Pearlson, G. D. (2016). Genetics of cognitive control: Implications for Nimh's research domain criteria initiative. *American Journal of Medical Genetics. Part B, Neuropsychiatric Genetics, 171B*(1), 111–120. https://doi.org/10.1002/ajmg.b.32345

Gottesman, I. I., & Gould, T. D. (2003). The endophenotype concept in psychiatry: Etymology and strategic intentions. *The American Journal of Psychiatry, 160*(4), 636–645. https://doi.org/10.1176/appi.ajp.160.4.636

Grob, G. N. (1985). The origins of American psychiatric epidemiology. *American Journal of Public Health, 75*(3), 229–236.

Haller, J. S. (1972). The Negro and the southern physician: A study of medical and racial attitudes 1800–1860. *Medical History, 16*(3), 238–253. https://doi.org/10.1017/S0025727300017737

Halter, M. J., Rolin-Kenny, D., & Grund, F. (2013). DSM-5: Historical perspectives. *Journal of Psychosocial Nursing and Mental Health Services, 51*(4), 22–29. https://doi.org/10.3928/02793695-20130226-03

Henje Blom, E., Duncan, L. G., Ho, T. C., Connolly, C. G., LeWinn, K. Z., Chesney, M., … Yang, T. T. (2014). The development of an RDoC-based treatment program for adolescent depression: "Training for Awareness, Resilience, and Action" (TARA). *Frontiers in Human Neuroscience, 8*, 630. https://doi.org/10.3389/fnhum.2014.00630

Hershenberg, R., & Goldfried, M. R. (2015). Implications of RDoC for the research and practice of psychotherapy. *Behavior Therapy, 46*(2), 156–165. https://doi.org/10.1016/j.beth.2014.09.014

Houts, A. C. (2000). Fifty years of psychiatric nomenclature: Reflections on the 1943 war department technical bulletin, medical 203. *Clinical Psychology, 56*(7), 925–934.

Policy Recommendations: Addressing the Mental Health and Wellness of Individuals with Intellectual Disabilities. In. Retrieved from https://utw10282.utweb.utexas.edu/wp-content/uploads/2015/09/MH_IDD-Policy-Rec_0801141.pdf

Human Connectome Project: About. Retrieved from http://www.humanconnectomeproject.org/about/

Insel, T., Cuthbert, B., Garvey, M., Heinssen, R., Pine, D. S., Quinn, K., … Wang, P. (2010). Research domain criteria (RDoC): Toward a new classification framework for research on mental disorders. *The American Journal of Psychiatry, 167*(7), 748–751. https://doi.org/10.1176/appi.ajp.2010.09091379

Jablensky, A., & Waters, F. (2014). RDoC: a roadmap to pathogenesis?. *World Psychiatry, 13*(1), 43.

Kawa, S., & Giordano, J. (2012). A brief historicity of the diagnostic and statistical manual of mental disorders: Issues and implications for the future of psychiatric canon and practice. *Phiolosophy, Ethics, and Humanities in Medicine, 7*, 2.

Kelly, J. R., Clarke, G., Cryan, J. F., & Dinan, T. G. (2016). Brain-gut-microbiota axis: challenges for translation in psychiatry. *Annals of Epidemiology, 26*(5), 366–372.

Kendler, K. S. (2009). An historical framework for psychiatric nosology. *Psychological Medicine, 39*(12), 1935–1941. https://doi.org/10.1017/S0033291709005753

Kendler, K. S. (2016). The transformation of American psychiatric nosology at the dawn of the twentieth century. *Molecular Psychiatry, 21*(2), 152–158. https://doi.org/10.1038/mp.2015.188

Keyes, D. E., Ltaief, H., & Turkiyyah, G. (2020). Hierarchical algorithms on hierarchical architectures. Philosophical Transactions of the Royal Society A, 378(2166), 20190055.

Kirmayer, L. J., & Crafa, D. (2014). What kind of science for psychiatry?. *Frontiers in Human Neuroscience, 8*, 435.

Koslowski, N., Klein, K., Arnold, K., Kosters, M., Schutzwohl, 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. *The British Journal of Psychiatry, 209*(6), 469–474. https://doi.org/10.1192/bjp.bp.114.162313

Kotov, R., Krueger, R. F., Watson, D., Achenbach, T. M., Althoff, R. R., Bagby, R. M., … Zimmerman, M. (2017). The Hierarchical Taxonomy of Psychopathology (HiTOP): A dimensional alternative to traditional nosologies. *Journal of Abnormal Psychology, 126*(4), 454–477. https://doi.org/10.1037/abn0000258

Krahn, G. L., Walker, D. K., & Correa-De-Araujo, R. (2015). Persons with disabilities as an unrecognized health disparity population. *American journal of public health, 105*(S2), S198–S206.

Krueger, R. F., Kotov, R., Watson, D., Forbes, M. K., Eaton, N. R., Ruggero, C. J., ... & Bagby, R. M. (2018). Progress in achieving quantitative classification of psychopathology. *World Psychiatry, 17*(3), 282–293.

Kozak, M. J., & Cuthbert, B. N. (2016). The NIMH research domain criteria initiative: Background, issues, and pragmatics. *Psychophysiology, 53*(3), 286–297. https://doi.org/10.1111/psyp.12518

Kuntz, T. M., & Gilbert, J. A. (2017). Introducing the microbiome into precision medicine. *Trends in Pharmacological Sciences, 38*(1), 81–91. https://doi.org/10.1016/j.tips.2016.10.001

Lahey, B. B., Applegate, B., Hakes, J. K., Zald, D. H., Hariri, A. R., & Rathouz, P. J. (2012). Is there a general factor of prevalent psychopathology during adulthood? *Journal of Abnormal Psychology, 121*(4), 971–977. https://doi.org/10.1037/a0028355

Landis, C., & Page, J. (1938). *Modern society and mental disease*. New York, NY: Farrar and Rinehart.

Lapointe, F. H. (1972). Who originated the term "psychology"? *Journal of the History of the Behavioral Sciences, 8*, 328–335.

Lilienfeld, S. O., & Treadway, M. T. (2016). Clashing diagnostic approaches: DSM-ICD versus RDoC. *Annual*

Review of Clinical Psychology, 12, 435–463. https://doi.org/10.1146/annurev-clinpsy-021815-093122

Lingiardi, V., & McWilliams, N. (Eds.). (2017). Psychodynamic diagnostic manual: PDM-2. Guilford Publications.

Litwack, L. (1958). The Federal Government and the Free Negro, 1790-1860. *Journal of Negro History, 43*(4), 263–268.

Luckasson, R., & Schalock, R. L. (2013). Defining and applying a functionality approach to intellectual disability. *Journal of Intellectual Disability Research, 57*(7), 657–668. https://doi.org/10.1111/j.1365-2788.2012.01575.x

Lupien, S. J., Sasseville, M., Francois, N., Giguere, C. E., Boissonneault, J., Plusquellec, P., … Lesage, A. (2017). The DSM5/RDoC debate on the future of mental health research: Implication for studies on human stress and presentation of the signature bank. *Stress, 20*(1), 95–111. https://doi.org/10.1080/10253890.2017.1286324

Marneros, A. (2008). Psychiatry's 200th birthday. *The British Journal of Psychiatry, 193*(1), 1–3. https://doi.org/10.1192/bjp.bp.108.051367

McCord, D. M., Achee, M. C., Cannon, E. M., Harrop, T. M., & Poynter, W. D. (2017). Using the research domain criteria framework to explore associations between MMPI-2-RF constructs and physiological variables assessed by eye-tracker technology. *Journal of Personality Assessment, 99*(4), 363–374. https://doi.org/10.1080/00223891.2016.1228067

McWilliams, N. (2011). The Psychodynamic Diagnostic Manual: An effort to compensate for the limitations of descriptive psychiatric diagnosis. *Journal of Personality Assessment, 93*(2), 112–122.

Millon, T., Krueger, R., & Simonsen, E. (2010). *Contemporary directions in psychopathology scientific foundations of the DSM-V and ICD-11*. New York, NY: Guilford Press.

Mittal, V. A., & Wakschlag, L. S. (2017). Research domain criteria (RDoC) grows up: Strengthening neurodevelopmental investigation within the RDoC framework. *Journal of affective disorders, 216*, 30.

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), 95–102. https://doi.org/10.1097/YCO.0000000000000236

National Institutes of Mental Health. *Research domain criteria*. Retrieved from https://www.nimh.nih.gov/research-priorities/rdoc/index.shtml

National Research Council. (2011). *Toward precision medicine: Building a knowledge network for biomedical research and a new taxonomy of disease*. Washington, D.C.: National Academies Press.

O'Connor, B. P. (2005). Graphical analyses of personality disorders in five-factor model space. *European journal of personality: Published for the European association of personality psychology*, 19(4), 287–305.

Palm, U., & Möller, H. J. (2011). Reception of Kraepelin's ideas 1900-1960. *Psychiatry and Clinical Neurosciences, 65*(4), 318–325. https://doi.org/10.1111/j.1440-1819.2011.02226.x

Parnas, J. (2014). The RDoC program: Psychiatry without psyche? *World Psychiatry, 13*(1), 46–47. https://doi.org/10.1002/wps.20101

Patrick, C. J., Durbin, C. E., & Moser, J. S. (2012). Reconceptualizing antisocial deviance in neurobehavioral terms. *Development and Psychopathology, 24*(3), 1047–1071. https://doi.org/10.1017/s0954579412000533

Patrick, C. J., Venables, N. C., Yancey, J. R., Hicks, B. M., Nelson, L. D., & Kramer, M. D. (2013). A construct-network approach to bridging diagnostic and physiological domains: Application to assessment of externalizing psychopathology. *Journal of Abnormal Psychology, 122*(3), 902–916. https://doi.org/10.1037/a0032807

Reference guide. (2018). In. Retrieved from https://icd.who.int/browse11/content/refguide.ICD11_en/html/index.html

Remarks by the president at the signing of the 21st century communications and Video Accessibility Act of 2010. (2010). Retrieved from https://obamawhitehouse.archives.gov/the-press-office/2010/10/08/remarks-president-signing-21st-century-communications-and-video-accessib

Sabatello, M. (2018). Precision medicine, health disparities, and ethics: the case for disability inclusion. *Genetics in Medicine, 20*(4), 397–399.

Sanislow, C. A. (2016). Updating the research domain criteria. *World Psychiatry, 15*(3), 222.

Schalock, R. L., Borthwick-Duffy, S. A., Bradley, V. J., Buntinx, W. H., Coulter, D. L., Craig, E. M., & Shogren, K. A. (2010). *Intellectual disability: Definition, classification, and Systems of Supports*. Washington, D.C.: American Association on Intellectual and Developmental Disabilities.

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: British Medical Journal, 351*, h4326.

Shorter, E. (1997). *A history of psychiatry: From the era of the asylum to Prozac*. New York, NY: Wiley.

Spitzer, R. L., Endicott, J., & Robins, E. (1978). Research diagnostic criteria: rationale and reliability. *Archives of General Psychiatry, 35*(6), 773–782.

Stengel, E. (1959). Classification of mental disorders. *Bulletin of the World Health Organization, 21*, 601–663.

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, 51*(2), 127–131. https://doi.org/10.1352/1934-9556-51.2.127

Tassé, M. J., Luckasson, R., & Schalock, R. L. (2016). The relation between intellectual functioning and adaptive behavior in the diagnosis of intellectual dis-

ability. *Intellectual and developmental disabilities, 54*(6), 381–390.

Telles-Correia, D. (2018). Mental disorder: Are we moving away from distress and disability? *Journal of Evaluation in Clinical Practice, 24*(5), 973–977. https://doi.org/10.1111/jep.12871

Thompson, B., Schalock, S., Tassé, W., … Rotholz. (2017). *Person-centered planning with the supports intensity scale-adult version™: A guide for planning teams*. Washington D.C: AAIDD.

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. https://doi.org/10.1016/j.ridd.2014.01.027

Tyrer, P., Crawford, M., Sanatinia, R., Tyrer, H., Cooper, S., Muller-Pollard, C., … Weich, S. (2014). Preliminary studies of the ICD-11 classification of personality disorder in practice. *Personality and Mental Health, 8*(4), 254–263. https://doi.org/10.1002/pmh.1275

Ustün, T. B., Chatterji, S., Kostanjsek, N., Rehm, J., Kennedy, C., Epping-Jordan, J., … Project, W. N. J. (2010). Developing the World Health Organization disability assessment schedule 2.0. *Bulletin of the World Health Organization, 88*(11), 815–823. https://doi.org/10.2471/BLT.09.067231

Weine, S. M., Langenecker, S., & Arenliu, A. (2018). Global mental health and the National Institute of Mental Health Research domain criteria. *The International Journal of Social Psychiatry, 64*(5), 436–442. https://doi.org/10.1177/0020764018778704

Widiger, T. A., Lynam, D. R., Miller, J. D., & Oltmanns, T. F. (2012). Measures to assess maladaptive variants of the five-factor model. *Journal of Personality Assessment, 94*(5), 450–455.

What is the precision medicine initiative?. (2018). Retrieved from https://ghr.nlm.nih.gov/primer/precisionmedicine/initiative

WHO. (1949). *Manual of the international statistical classification of diseases, injuries, and causes of death* (6th ed.). Geneva, Switzerland: World Health Organization.

WHO. (1967). *Manual of the international statistical classification of diseases, injuries, and causes of death* (8th ed.). Geneva, Switzerland: World Health Organization.

WHO. (1975). *Manual of the international statistical classification of diseases, injuries, and causes of death* (9th ed.). Geneva, Switzerland: World Health Organization.

WHO. (2017). *International classification of functioning, disability and health*. Geneva, Switzerland: World Health Organization.

WHO. (2018). *Manual of the international statistical classification of diseases, injuries, and causes of death* (11th ed.). Geneva, Switzerland: World Health Organization.

World Health Organization. (1992). *The ICD-10 classification of mental and behavioral disorders: Clinical descriptions and diagnostic guidelines*. Geneva, Switzerland: World Health Organization.

World Health Organization. (1993). *The ICD-10 classification of mental and behavioural disorders: Diagnostic criteria for research*. Geneva, Switzerland: World Health Organization.

Zeanah, C. H., Carter, A. S., Cohen, J., Egger, H., Gleason, M. M., Keren, M., ... & Oser, C. (2016). Diagnostic classification of mental health and developmental disorders of infancy and early childhood dc: 0–5: Selective reviews from a new nosology for early childhood psychopathology. *Infant mental health journal, 37*(5), 471–475.

Zero to Three (2016). Diagnostic classification of mental health and developmental disorders of infancy and early childhood: Revised edition DC: 0-5. ZERO TO THREE Press, Washington, DC.

Zuberi, S. M., & Brunklaus, A. (2018). Epilepsy in 2017: Precision medicine drives epilepsy classification and therapy. *Nature Reviews. Neurology, 14*(2), 67–68. https://doi.org/10.1038/nrneurol.2017.190

Challenging Behaviors and Dual Diagnosis

3

Matthew J. O'Brien

Introduction

It is a universal finding that individuals diagnosed with an intellectual disability (ID) are more likely to display challenging behaviors than peers without cognitive impairment (Baker et al., 2003; Dekker, Koot, Ende, & Verhulst, 2002; Einfeld, Ellis, & Emerson, 2011; Emerson et al., 2001). Outcomes for those exhibiting challenging behavior are unfavorable and include poorer quality of life (Murphy, 2009), negative perceptions and burnout for their care providers (Ko, Lunsky, Hensel, & Dewa, 2012; Mitchell & Hastings, 2001; Zijlmans, Embregts, Bosman, & Willems, 2012), increased likelihood of placement out of the family home (Chan & Sigafoos, 2000), and higher costs associated with the increased care needed to support them (Allen, Lowe, Moore, & Brophy, 2007; Knapp, Comas-Herrera, Astin, Beecham, & Pendaries, 2005). There is also a large body of research that suggests individuals with ID are more likely to be diagnosed with comorbid psychiatric disorders than persons without an ID diagnosis (Axmon, Bjorne, Nylander, & Ahlstrom, 2018; Cooper, Smiley, Morrison, Williamson, & Allan, 2007; Dekker et al., 2002; Smiley et al., 2007; Whitaker & Read, 2006). Although the relationship between challenging behavior, ID, and mental health is poorly understood (Grey, Pollard, McClean, MacAuley, & Hastings, 2010), given the relationships between ID and challenging behaviors and ID and psychopathology, it seems that the topic of challenging behavior is central to understanding the dual diagnosis population (i.e., intellectual disability and psychopathology). The purpose of this chapter is to provide an overview of challenging behaviors in ID populations with comorbid mental health diagnoses, including commonly observed challenging behaviors, problem behavioral profiles and mention of specific diagnoses, risk factors associated with developing challenging behaviors, and causes of such behavior.

Defining Challenging Behavior in Dual Diagnosis

Before surveying the existing research on challenging behaviors in individuals with dual diagnosis, it is important to consider how these topics of interest are defined. A considerable amount of research has been conducted on the risks, causes, and prevalence of challenging behavior among individuals with ID (much less research has been done on challenging behavior in individuals with dual diagnosis); however, consistent findings have been elusive. For example,

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several studies have reported prevalence rates of problem behavior among individuals with ID below 10% (e.g., Emerson & Bromley, 1995; Holden & Gitlesen, 2006; Lowe et al., 2007), while other studies have suggested that the prevalence rate is greater than 50% (e.g., Dekker et al., 2002; Dworschak, Ratz, & Wagner, 2016; Poppes, van der Putten, & Vlaskamp, 2010). There are multiple reasons for variable results, most of which lies in the different research methods employed in such studies. Much of the discrepancy seems to involve the lack of a uniform definition for challenging behavior. In a summary of findings on the prevalence of challenging behavior in the ID population, Koritsas and Iacono (2012) highlighted this problem and enumerated the various ways that studies have constituted "challenging behavior." They reported that some studies define challenging behavior based largely upon its impact on the individual who exhibits challenging behavior and those caring for that individual. A number of studies include a broad variety of behavioral topographies, while others are much more targeted (e.g., aggression and self-injury only). Moreover, definitions of specific topographies vary greatly. For example, while most studies of self-injurious behavior (SIB) define such behavior based upon whether it causes tissue damage or injury to the individual exhibiting the behavior, as Koritsas and Iacono point out, there are studies that have included less common behaviors such as polydipsia (i.e., excessive thirst or excessive drinking) and rumination in their definitions of SIB (Griffin, Williams, Stark, Altmeyer, & Mason, 1986; Maurice & Trudel, 1982; Rojahn, 1986; Schroeder, Schroeder, Smith, & Dalldorf, 1978). Furthermore, for a behavior to be considered, SIB several studies have required that the behavior be repetitive (Maurice & Trudel, 1982; Oliver, Murphy, & Corbett, 1987; Rojahn, 1986). Even when studies are in agreement on the topographies for inclusion, behavioral dimensions, such as the severity or intensity of a behavior, the frequency, and the temporal parameters (e.g., behavior occurring in the past week, 2 months, or at any point during a lifetime), may vary greatly.

Inconsistent behavioral definitions are not the only barrier to synthesizing the research on challenging behavior in individuals with dual diagnosis. Differences in the definition for intellectual disability and determining what constitutes "dual diagnosis" have also led to less than reliable and sometimes contradictory findings. Changes in diagnostic requirements, such as the recent shift in diagnostic criteria for ID that focuses more on an individual's adaptive functioning than IQ score (American Psychiatric Association, 2013), can make it difficult to compare causes, risk factors, and prevalence rates for a heterogeneous group across studies over time. Despite the differences mentioned, much can still be gleaned from the research. A small and slowly increasing number of studies are exploring the connection between challenging behavior and psychopathology in the ID population, yet a substantial amount of investigation has already been carried out on the relationship between challenging behavior and intellectual disability without consideration for co-occurring psychiatric symptoms. To avoid the risk of providing a very narrow and somewhat incomplete look at challenging behavior in individuals with dual diagnosis, this chapter will draw largely from the literature on challenging behavior in individuals diagnosed with ID, without unwarranted discussion on the various definition challenges described above. Whenever applicable, research specific to dual diagnosis will be incorporated, and any differences between what is known about challenging behavior in the ID population and what is known about challenging behavior in those with dual diagnosis will be highlighted.

Common Challenging Behaviors

As noted previously, the definition of challenging behavior is not universal. What may be considered an adaptive behavior can quickly be deemed problematic when displayed at an excessive frequency or duration and during contextually inappropriate times. Take, for example, perseverative thinking and behavior, which is not uncommon in individuals with autism (Leekam, Prior, &

Uljarevic, 2011). If a child were to ask a parent about the following day's schedule of activities, it would likely be considered an appropriate display of curiosity. Yet, if a child were to ask a parent that same question hundreds of times throughout a single day or interrupt a classroom discussion to ask the teacher that question, it may be considered aberrant, if not problematic. Using this logic, nearly any behavior may be considered maladaptive depending on the form and context in which that behavior is displayed; however, there are several challenging behaviors that would not be considered acceptable under any condition, regardless of the frequency, duration, or context. A broad range of maladaptive behaviors has been documented in the literature on challenging behavior and ID or dual diagnosis population, but aggression, self-injury, property destruction, and stereotyped behaviors are cited more frequently than any other challenging behavior. A description of these behaviors, including the specific topographies, prevalence rates, and diagnoses commonly associated with each, is presented next.

Aggression

Studies of aggression may include a fairly wide range of topographies, including various forms of physical aggression, verbal aggression, property destruction, and self-injurious behavior (sometimes called "autoaggression") but often with little or no attempt to distinguish between them. Physical aggression involves physical acts and/or threats to commit physical acts toward others in a manner that is likely to inflict harm. Verbal aggression is poorly defined in the research literature but may include screaming, swearing, abusive language, or verbal threats directed at others. Common forms of physical aggression include hitting, kicking, scratching, pinching, biting, and throwing objects at others; however, a broad range of topographical forms have been studied, including pushing, punching, pulling other's hair, grabbing other's clothing, headbutting, flicking hair into other's faces, choking, using objects as weapons, spitting, grabbing materials or food from others, poking others' eyes, and inappropriately touching or fondling others in a sexual manner (Arron, Oliver, Moss, Berg, & Burbidge, 2011; Crocker et al., 2006; Finucane, Konar, Haas-Givler, Kurtz, & Scott Jr., 1994; Hessl et al., 2001; Lowe et al., 2007; Oliver, Oxener, Hearn, & Hall, 2001; Petty, Bacarese-Hamilton, Davies, & Oliver, 2014; Ross Collins & Cornish, 2002; Sloneem, Oliver, Udwin, & Woodcock, 2011; Wigren & Heimann, 2001). Hitting is often identified as the most common form of aggression in individuals with ID (Lowe et al., 2007).

Prevalence rates of aggression among individuals with ID vary considerably, depending upon the definitions utilized and the population sampled. Rates range from as low as 2% (Borthwick-Duffy, 1994) to as high as 51% (Crocker et al., 2006). Prevalence studies reporting higher rates tend to include samples from individuals living in institutions and other restrictive settings. Among specific diagnoses, higher prevalence rates are seen in Smith-Magenis syndrome, Angelman syndrome, fragile X syndrome, and Prader-Willi syndrome (Arron et al., 2011; Powis & Oliver, 2014), while lower rates of aggression are observed in Down syndrome and Williams syndrome (Powis & Oliver, 2014).

Unlike other forms of challenging behavior, the fact that aggression involves the targeting of others may negatively impact social relationships to a greater degree than other challenging behaviors (Cooper, Smiley, Jackson, et al., 2009; Antonacci, Manuel, & Davis, 2008; Allen, 2000). Additionally, aggression negatively affects the educational and vocational opportunities of individuals with ID and contributes to frequent changes in residential placement (Jahoda, Willner, Pert, & MacMahon, 2013). In fact, in comparison with other challenging behaviors, aggression has been cited as requiring the greatest amount of support to manage (Ruddick, Davies, Bacarese-Hamilton, & Oliver, 2015).

Little research has been conducted specifically on aggression in individuals with dual diagnosis; however, some have cited aggression among individuals with ID as the most common reason for referral to a mental health specialist (Harper, 2019; Jahoda et al., 2013). Van den

Bogaard, Nijman, Palmstierna, and Embregts (2018) conducted a study of aggression among dual diagnosis individuals in a residential setting, resulting in several interesting findings. They noted that individuals with dual diagnosis were much more likely to exhibit verbal aggression than physical aggression. They also found that the common target of aggression was staff members, with very few incidents targeting other residents. Although it was common for others (e.g., staff and other residents) to feel a sense of threat from the aggressive acts, physical injury or pain only occurred in a relatively small percentage of incidents.

Self-Injury

Like aggression, self-injurious behavior (SIB) may include a broad range of topographical forms depending upon how it is defined. Generally, any behavior directed toward oneself with the potential to result in tissue damage is considered self-injurious (Tate & Baroff, 1966). Unfortunately, numerous methods of harming oneself have been documented in ID research, including object/hand-to-head/body hitting or slapping, head-to-surface banging, hair pulling, lip or body biting, eye poking, skin picking, scratching, compulsive eating, bruxism, rumination, aerophagia, inserting items into the body/rectum, pinching, hand mouthing, and rectal digging (Hessl et al., 2001; Huisman et al., 2018; Kahng, Iwata, & Lewin, 2002; Rojahn & Esbensen, 2002; Ross Collins & Cornish, 2002; Symons, Harper, Shinde, Clary, & Bodfish, 2010). Definitions of SIB often consider behavior that has the *potential* for damage, as well as other important dimensions of the behavior, such as the frequency, the targeted area of the body, and the intensity of the behavior.

In total population studies, prevalence rates of SIB in persons with ID range from less than 3% (Holden & Gitlesen, 2006) to roughly 30% (Emerson et al., 2001; Lundqvist, 2013). Higher rates are found in a number of specific diagnoses associated with ID, including autism (Buono, Scannella, & Palmigiano, 2010), Cornelia de Lange syndrome (Arron et al., 2011), cri du chat syndrome (Ross Collins & Cornish, 2002), fragile X syndrome (Hall, Lightbody, & Reiss, 2008), Lesch-Nyhan syndrome (Anderson & Ernst, 1994), Prader-Willi syndrome (Didden, Korzilius, & Curfs, 2007), Rett syndrome (Sansom, Krishnan, Corbett, & Kerr, 1993), and Smith-Magenis syndrome (Arron et al., 2011). By some estimates, fewer individuals with Down syndrome, Tuberous sclerosis, and Williams syndrome display SIB than the general ID population (Huisman et al., 2018). Like most challenging behaviors, self-injury is more prevalent among those living in institutionalized or other restricted settings (Borthwick-Duffy, 1994).

Among SIB topographies, several studies have identified head hitting as the most common form of SIB (Lowe et al., 2007; Mulick, Dura, Rasnake, & Callahan, 1986; Rojahn, 1984; Rojahn, 1986), while others have suggested biting is more prevalent (Bodfish et al., 1995; Emberson & Walker, 1990; Griffin et al., 1987; Hillery & Mulcahy, 1997; Rojahn, 1986). Regardless, many individuals who exhibit SIB display multiple topographies (Emberson & Walker, 1990; Griffin et al., 1986; Oliver et al., 1987). In fact, Bodfish et al. (1995) reported that more than a third of individuals with ID exhibit three or more forms of SIB, and Rojahn (1984) found that more than half did so.

As noted above, pica is often considered a form of self-injury due to the potential for a myriad of harmful outcomes, including poisoning, infection, intestinal blockages or perforations, and even death (Sturmey & Williams, 2016). Although reported prevalence rates of pica in the ID population have been as high as 31% (Danford & Huber, 1982; Lowe et al., 2007), most studies have found rates near or below 15% (Griffin et al., 1987; Hove, 2004; Rojahn, 1984). There is some evidence that individuals with autism, cri du chat syndrome, and Prader-Willi syndrome are more likely to display pica than other individuals with ID (Clark, Vandermeer, Simonetti, & Buka, 2010; Dykens, 2000; Ross Collins & Cornish, 2002).

Forms of self-mutilation, such as skin cutting and skin burning, are associated with psychiatric

disorders, including post-traumatic disorder, eating disorders, and intermittent explosive disorder (Zlotnick, Mattia, & Zimmerman, 1999). As noted previously, individuals with ID are highly susceptible to psychopathology, which may increase the likelihood that forms of self-mutilation would occur in individuals with dual diagnosis; however, the connection between ID, psychopathology, and self-mutilation has not received attention in the literature on challenging behavior in individuals with dual diagnosis, and there is no reason to believe that it is more common in individuals with ID.

Property Destruction

Many studies incorporate acts involving property destruction into their definition of aggression. Indeed, property destruction includes many of the same topographical forms (e.g., hitting, kicking, biting), as well as tearing, throwing, stomping, and slamming, though the target of these behaviors is an object rather than a person. Prevalence studies that have made the distinction between aggression and property destruction have found property destruction to be relatively common, suggesting that it could stand alone as a challenging behavior. In fact, at least one study (Borthwick-Duffy, 1994) found destruction to be the most likely problem behavior observed in individuals with ID. In general, property destruction is found to be as common or more so than self-injury (Crocker et al., 2006; Emerson et al., 2001; Holden & Gitlesen, 2006; Lowe et al., 2007; Matson & Rivet, 2008). Furthermore, in terms of the effects of property destruction, Ruddick et al. (2015) found that among children and adolescents with ID, property destruction required more substantial support to manage than self-injury, and not much less than aggression.

Stereotyped Behaviors

Although several terms have been used interchangeably with stereotypy (e.g., "self-stimulation" or "stimming," "sensory seeking") and many definitions exist, stereotyped behaviors are generally identified as repetitive motor or vocal behaviors occurring with little variance. To add to this general definition, Rapp and Lanovaz (2016) suggested the requirement that the behavior persists in the absence of social consequences. Common examples of stereotyped behavior include body rocking, hand flapping, finger flicking, spinning, twirling, grunting, echolalia, and humming (Bodfish, 2007; Lanovaz & Sladeczek, 2012). Whether a stereotypy is considered a challenging behavior depends upon the degree to which the behavior interferes with daily functioning. Stereotyped behaviors have the potential to be highly maladaptive, as they have been associated with disrupted learning (Klintwall & Eikeseth, 2012), poorer social integration (Jones, Wint, & Ellis, 1990), and the development of self-injurious behaviors (Kennedy, 2002).

Stereotyped behaviors are common in individuals with ID. Prevalence rates in total population samples range from 10% (Bowring, Totsika, Hastings, Toogood, & Griffith, 2017) to 34% (Lundqvist, 2013); however, in the latter study, only 6% of individuals with ID were considered to have stereotyped behavior severe enough to meet their definition of challenging behavior. Most studies have found the prevalence much higher, often above 50% (Bodfish et al., 1995; Bodfish, Symons, Parker, & Lewis, 2000; Medeiros, Curby, Bernstein, Rojahn, & Schroeder, 2013; Poppes et al., 2010). Individuals with ID, who also have a diagnosis of autism, are highly likely to exhibit stereotyped behaviors (Lewis & Bodfish, 1998), as well as other diagnoses associated with ID, including Smith-Magenis syndrome, fragile X syndrome, Rett syndrome, and Prader-Willi syndrome (Dykens & Smith, 1998; Turner, 1999).

Risk Factors

Dual diagnosis has been identified as a possible risk factor for challenging behavior in individuals with ID (Folch et al., 2018); yet, little research

has focused on the risk factors of challenging behavior among individuals with a dual diagnosis. Numerous studies have been conducted on the putative risk factors for challenging behavior in individuals with ID, some of which provide insight into the relationship between dual diagnosis and challenging behavior. The most common risk factors studied have been related to individual demographics, skills, or traits. A smaller number of studies have evaluated the association between challenging behavior and a variety of factors considered supervenient to the individual, such as type of residential setting or usage of psychotropic medications. It is unlikely that any one risk factor will prove highly predictive of challenging behavior for individuals with ID or dual diagnosis; rather, a multifactorial model that accounts for the interaction of various risk factors is likely necessary to fully understand why challenging behavior is exhibited by some individuals with ID or dual diagnosis and not others (Sigafoos, Arthur, & O'Reilly, 2003). Whether one factor or multiple factors are considered, any attempt to consolidate the existing research on risk factors will undoubtedly find the methodological and definitional differences across studies a barrier too difficult to reconcile.

There is little indication that many of the studied risk factors should be considered causal, and for some factors, such as placement in a residential setting or use of prescription medicine, the direction of the relationship is questionable (e.g., does challenging behavior lead to increased risk of residential placement or vice versa?). Nonetheless, what follows is a look at the research on specific potential risk factors for challenging behavior in individuals with ID or dual diagnosis. Most of the research presented is on challenging behavior as a whole, but findings on selected topographies are also included. The factors below have been classified into three categories: those inherent to the individual (i.e., a demographic, skill, or trait unlikely to change), factors extraneous to the individual (i.e., factors subject to change and/or potentially controlled by others), or factors related to psychiatric symptoms or diagnoses.

Risk Factors Inherent to the Individual

Age For total population studies, the relationship between age and challenging behavior is always a focal point, while studies sampling from a restricted age range have less freedom to explore this relationship. Most often, studies that look at age as a risk factor find a significant relationship; however, nuances, such as differences in the relationship based upon the severity of ID, are sometimes found (e.g., Emerson et al., 1997).

Total population studies by Emerson et al. (2001) and Holden and Gitlesen (2006) have found that for individuals with ID, challenging behavior was more likely to be observed in adolescents and young adults, but rates decline among individuals entering later adulthood (i.e., 40–50 years of age and beyond). Gitlesen and Holden noted that this pattern was distinct for those exhibiting "more demanding" challenging behavior (based upon frequency and negative consequences) as the rates for those with "less demanding" challenging behavior were evenly distributed across the life span until age 60, at which time milder forms of challenging behavior were nearly nonexistent. Increased prevalence of challenging behavior among adolescents and young adults juxtaposed by lower prevalence among older adults has been supported by studies utilizing a narrower population sample (Lowe et al., 2007). The decline in challenging behavior at later adulthood has been hypothesized as a result of limited activity levels often observed in older adults (Koritsas & Iacono, 2012); however, this has not been confirmed in any studies.

Few studies have found a relationship between age and challenging behavior in individuals with ID. A recent total population study by Bowring et al. (2017) failed to find an association between age and challenging behavior, as well as age and specific topographies of aggression, SIB, and stereotyped behavior. Studies with more restricted age ranges (including those examining only children and those examining only older adults) have also reported no significant association between age and challenging behavior (Davies & Oliver,

2016; Dworschak et al., 2016; Koritsas & Iacono, 2015; Schützwohl et al., 2016).

Among specific topographies of challenging behavior, multiple studies have established a relationship between age and aggression and/or self-injury. Davies and Oliver (2013) conducted a review of the literature on age as a risk for aggression and SIB in individuals with ID. For both topographies, convergent evidence suggested a curvilinear relationship whereby an increase in the risk of problem behavior was observed from childhood through mid-to-late adulthood before decreasing thereon.

A couple of studies have assessed age as a risk factor for challenging behavior specifically in individuals with dual diagnosis. Dinya, Csorba, Suli, and Grosz (2012) found a significant correlation between younger age and ratings of self-injury and aggression/destruction for adolescents with dual diagnosis but failed to find a correlation between age and stereotypic behavior. Using a broader age sample, Dudley, Ahlgrim-Delzell, and Calhoun (1999) did not find a relationship between age and aggressive or destructive behavior but noted that the older an individual was, the greater the likelihood that he or she fits into a subgroup that exhibited pica and/or SIB.

Gender Males are more likely than females to be diagnosed with ID (Drews, Yeargin-Allsopp, Decouflé, & Murphy, 1995; Maulik, Mascarenhas, Mathers, Dua, & Saxena, 2011), but even in studies controlling for this difference, determination of whether gender is a risk factor for challenging behavior in individuals with ID or dual diagnosis remains unclear. The majority of studies have not found any connection between gender and overall challenging behavior in the ID population (Bowring et al., 2017; Felce & Kerr, 2013; Holden & Gitlesen, 2006; Koritsas & Iacono, 2015; Schützwohl et al., 2016), but of the studies that have identified gender as a risk, all but a few exceptions (e.g., Jones et al., 2008) have suggested males are more likely to demonstrate challenging behavior than females (Dworschak et al., 2016; Einfeld et al., 2011; Emerson et al., 2001).

It is possible that gender is not a risk factor for challenging behavior in general but still a risk factor for specific topographies of challenging behavior. For example, Tyrer et al. (2006) found that males were more likely to display aggression than females in an adult population with ID, while Davies and Oliver (2016) found the same thing in children with ID. Additionally, a meta-analytic study involving samples across the life span by McClintock, Hall, and Oliver (2003) indicated that males were more likely than females to engage in aggression. By contrast, Dudley et al. (1999) reported that females with a dual diagnosis were more likely to exhibit specific topographies of aggressive behavior, including temper tantrums and screaming, than males with the same condition. Moreover, a total population study conducted by Lundqvist (2013) reported that adult females with ID were more likely than adult males to display various forms of physical and verbal aggression. Nonetheless, most studies continue to find no connection between gender and specific challenging behaviors, including stereotypies (Lundqvist, 2013), destruction (Davies & Oliver, 2016; Lowe et al., 2007), self-injury (Davies & Oliver, 2016; Dudley et al., 1999; Lowe et al., 2007; Lundqvist, 2013; McClintock, et al. 2003), and noncompliance (Lowe et al., 2007).

Severity of ID Among all risk factors studied, the greatest consensus lies on the association between ID severity and challenging behavior. Specifically, the more severe the level of ID or impairment, the greater the likelihood of challenging behavior. This finding has been true in total population studies (Bowring et al., 2017; Holden et al., 2006; Jones et al., 2008), meta-analyses (McClintock et al., 2003), and studies targeting specific age groups, including adult-only populations (Koritsas & Iacono, 2015), children (Chadwick, Piroth, Walker, Bernard, & Taylor, 2000), and older adults (Axmon et al., 2018; O'Dwyer et al., 2018).

Studies on specific topographies of challenging behavior have identified a greater risk for those with more severe ID in relationship to self-injury (Bowring et al., 2017; Cooper, Smiley,

Allan et al., 2009; Folch et al., 2018; Lowe et al., 2007; Lundqvist, 2013; McClintock et al., 2003; Tsiouris, Kim, Brown, & Cohen, 2011) and stereotyped behaviors (Bowring et al., 2017; Lundqvist, 2013; McClintock et al., 2003). The association between ID level and aggression is less robust, with studies that support a connection (Bowring et al., 2017; Cooper, Smiley, Jackson et al., 2009; Tyrer et al., 2006) and studies failing to find a connection (Davies & Oliver, 2016; Lundqvist, 2013; McClintock et al., 2003). Among individuals with a dual diagnosis, more severe ID has also been associated with self-injury and stereotyped behaviors (Dinya et al., 2012), as well as pica (Dudley et al., 1999). Level of ID does not appear to be associated with aggression in dual diagnosis studies (Dinya et al., 2012; Dudley et al., 1999).

Communication Communication difficulties are common in individuals with ID (Pinborough-Zimmerman et al., 2007) and an important component of theories on the cause of challenging behavior (see Causes). Thus, it should not be a surprise that communication has been identified as a risk factor for challenging behavior in individuals with ID in several studies (Bowring et al., 2017; Dworschak et al., 2016; Emerson et al., 2001; Holden & Gitlesen, 2006; Lundqvist, 2013; McClintock et al., 2003). In general, these studies have focused on expressive communication as a risk factor, but even receptive language deficits has been suggested as a risk factor (Bowring et al., 2017; Dworschak et al., 2016; Emerson et al., 2001; McClintock et al., 2003). Nonetheless, a few exceptions have not found an increased risk of challenging behavior with greater communication challenges (Koritsas & Iacono, 2015; O'Dwyer et al., 2018) or with a specific topography of challenging behavior (i.e., aggression) and expressive communication deficits (McClintock et al., 2003).

Of the few studies focused on specific topographies of challenging behavior and communication, the most consistent finding is that communication deficits are a risk factor for self-injury (Bowring et al., 2017; Lowe et al., 2007; McClintock et al., 2003). Interestingly, Danquah et al. (2009) found that individuals with poor expressive communication were twice as likely to display persistent self-injury (i.e., continued self-injury over a 2-year period) than those with stronger communication skills.

Communication has not been explored as a risk factor in the dual diagnosis population, apart from Dudley et al. (1999). Dudley and colleagues found that weaker communication was associated with pica, but not with aggression.

Risks Related to Psychiatry Symptoms/Psychiatric Diagnosis

Psychiatric symptoms and diagnoses were purposely excluded from the other two categories of risk factors in an attempt to highlight the inherent connection of challenging behavior and dual diagnosis. The relationship between challenging behavior and psychopathology in ID and dual diagnosis populations has been a topic of debate. While there are numerous studies that have suggested psychiatric symptoms may be a risk factor for challenging behavior, there is disagreement on the direction of the relationship and whether a causal link can be made. Additionally, when the relationship between challenging behavior and psychiatric diagnoses is considered, there are inherent problems of circular causality. For example, one might reason that an individual with autism demonstrates stereotypies because that individual has autism and also reason that she or he is diagnosed with autism, in part, because he or she exhibits stereotypies. This type of logic is highlighted by Rojahn, Matson, Naglieri, and Mayville (2004) in a study on the relationship between psychiatric conditions and problem behaviors exhibited by those with ID. In summarizing their results, they concluded that "… behavior problems are associated with psychiatric conditions that are traditionally known to be associated with behavior problems…" (p. 29). Further discussion on psychopathology as a cause of challenging behavior is offered later in this chapter.

An additional barrier to understanding the connection between psychiatric symptoms and challenging behavior is the questionable validity of diagnostic evaluations dependent upon self-report (Finlay & Lyons, 2001). For many individuals with ID, the ability to communicate symptoms associated with psychopathology may be poor (Witwer, Lawton, & Aman, 2014), and for those who are completely nonverbal, there is a necessary reliance on caregiver perceptions and/or behavioral observation to identify symptomology associated with psychiatric diagnoses. Several standardized instruments have been developed to assist in determining psychopathology in individuals with ID, such as the Assessment of Dual Diagnosis (ADD; Matson & Bamburg, 1998), the Diagnostic Assessment for the Severely Handicapped-II (DASH-II; Matson et al., 1996), and the Reiss Screen for Maladaptive Behavior (Havercamp & Reiss, 1997).

Numerous studies have evaluated the relationship between challenging behavior, ID, and psychopathology but often in different ways, leading to nuanced results. For example, Grey et al. (2010) examined whether psychiatric symptoms were associated with "more severe" and "less severe" challenging behavior and identified a higher risk of psychiatric disorder only for those with more severe behaviors. Rojahn et al. (2004) also found a strong relationship between psychopathology and challenging behavior; however, they noted that the challenging behavior was only related to psychiatric diagnoses where such behaviors are likely to be part of the diagnostic makeup and not related to other diagnoses that fail to include challenging behavior as part of the diagnostic criteria (e.g., depression). Among middle-age and older adults, Axmon et al. (2018), Felce, Kerr, and Hastings (2009), and O'Dwyer et al. (2018) found a significant risk of challenging behavior among individuals with a psychiatric diagnosis, and Axmon et al. further specified that the highest odds of challenging behavior were associated with psychotic, affective, and anxiety disorders. With regard to specific topographies, Folch et al. (2018) identified dual diagnosis as a risk factor specifically for self-injury. Nonetheless, at least one study (i.e., Lowe et al., 2007) considered the risk of having any psychiatric disorder on various topographies of challenging behavior and found no association with any.

Autism is the most common comorbid diagnosis associated with challenging behavior. It has been associated with a greater risk of challenging behavior in general, in total population studies (Holden & Gitlesen, 2006; Lundqvist, 2013), in adults (Axmon et al., 2018; Felce & Kerr, 2013; McCarthy et al., 2010), and across specific topographies, including self-injury (Folch et al., 2018; McClintock et al., 2003; Richards, Oliver, Nelson, & Moss, 2012; Tsiouris et al., 2011), aggression (McClintock et al., 2003), stereotypy (Felce & Kerr, 2013), pica (Dudley et al., 1999), and destruction (McClintock et al., 2003). Despite these findings, Goldin, Matson, and Cervantes (2014) examined the effect of ID on children and adolescents with autism and found that the combination of ID and autism did not lead to a greater number or intensity of challenging behaviors, a finding also obtained by Jones et al. (2008) in adults with ID.

Anxiety Among specific psychiatric symptoms, anxiety has been identified as a risk factor in several studies (Holden & Gitlesen, 2003; Koritsas & Iacono, 2015; Moss et al., 2000; Myrbakk & von Tetzchner, 2008; Tsiouris et al., 2011). Notwithstanding the overall evidence connecting anxiety and challenging behavior, in a review of studies evaluating this connection, Pruijssers, van Meijel, Maaskant, Nijssen, and van Achterberg (2014) made clear that the strength of the association is highly variable and may be easily influenced by stressful life events and thus transient.

Mood Low mood (i.e., depression) and excessively elevated mood (i.e., mania), as well as diagnoses consisting of these symptoms (e.g., major depressive disorder, bipolar disorder), have been associated with an increased risk of challenging behavior (Hayes, McGuire, O'Neill, Oliver, & Morrison, 2011; Moss et al., 2000; Myrbakk & von Tetzchner, 2008; Tsiouris et al., 2011; Turygin, Matson, MacMillan, & Konst, 2013). This has led some to suggest that

challenging behaviors may be a symptom of a psychiatric disorder (Myrbakk & von Tetzchner, 2008), called the "behavioral equivalents" hypothesis (The merits of this proposal are discussed later in the section on causes of challenging behavior.). Several studies have failed to find a relationship between mood and challenging behavior (Holden & Gitlesen, 2003; Koritsas & Iacono, 2015; Sturmey, Laud, Cooper, Matson, & Fodstad, 2010).

Impulsivity AD/HD and its associated symptoms have been cited in several studies as a risk factor for challenging behavior in both children and adults with ID. Specifically, hyperactivity and impulsivity have been associated with challenging behavior (Petty et al., 2014) and specific topographies of physical aggression (Davies & Oliver, 2016; Dudley et al., 1999; Rojahn et al., 2004), self-injury (Rojahn et al., 2004; Tsiouris et al., 2011), and destruction (Davies & Oliver, 2016) in children with ID. Among adults, Cooper, Smiley, Jackson, et al. (2009) and Cooper, Smiley, Allan, et al. (2009) found an increased risk of aggression and self-injury, respectively, with a comorbid diagnosis of AD/HD, a finding supported by Jones et al. (2008). Despite the research evidence, the connection between impulsivity or hyperactivity and challenging behavior is still not fully understood and devoid of strong theories explaining the association (Davies & Oliver, 2016).

Extraneous Risk Factors

Some putative risk factors are more transient, exogenous, or likely a consequence of challenging behavior and thus considered extraneous. For example, psychotropic medications may be a risk factor for challenging behavior, but unlike factors such as gender or age, the decision to use psychotropic medication is more probably made by the caregivers and is often an intended remedy for challenging behavior rather than the cause of such behavior. Many of the following putative risk factors have less overall research to support or refute their association with challenging behavior, but nonetheless remain important factors to consider.

Psychotropic Medication The use of psychotropic medication for individuals with dual diagnosis and challenging behavior is ubiquitous (Matson & Neal, 2009; Matson & Wilkins, 2008). Given the mental health conditions inherent in dual diagnosis, the use of medications intended to treat a variety of psychiatric symptoms, such as anxiety and depression (e.g., SSRIs) and impulsivity (e.g., stimulants), should not be a surprise; however, a large proportion of individuals with ID or dual diagnosis are prescribed medications targeting irritability and challenging behavior as well. Commonly, these medications include typical antipsychotics (e.g., haloperidol) and atypical antipsychotics (e.g., risperidone) (Matson & Neal, 2009). Indeed, multiple studies have found psychotropic medication use to be a risk factor for challenging behavior (Bowring et al., 2017; O'Dwyer et al., 2018), but the relationship may be dependent upon topography. More specifically, while Folch et al. (2018), Lundqvist (2013), and Bowring et al. (2017) found a higher risk of self-injury in adults with ID who were taking psychotropic medications, the latter two studies did not find the same association with stereotyped behaviors. To add nuance, Lundqvist (2013) did not find psychotropic medication to be a risk factor for aggression, while Bowring et al. (2017) did. Although psychotropic medication usage is widespread in children and adolescents with ID (Bramble, 2007; Haw & Stubbs, 2005; Matson et al., 2000), no studies have reproduced the above findings with younger populations. Though the logical direction of the relationship is that the presence of challenging behavior leads to psychotropic medication usage, the fact that negative side effects associated with medication can lead to challenging behavior (Valdovinos, Caruso, Roberts, Kim, & Kennedy, 2005) may make this association difficult to parse out.

Residential Setting A large proportion of individuals with dual diagnosis live in a group home setting (Gentile, Gillig, Stinson, & Jensen, 2014), and placement outside of the family home is but one negative consequence of challenging behavior (Chan & Sigafoos, 2000). Like psychotropic medications, it seems likely that the presence of challenging behavior may influence the residential setting of many individuals with ID rather than the other way around. Several studies have found living in a congregate care setting to be a significant risk factor for challenging behavior (Holden & Gitlesen, 2006; Jones et al., 2008; Lowe et al., 2007; O'Dwyer et al., 2018; Schützwohl et al., 2016). However, this finding has not been unanimous (see Koritsas & Iacono, 2015 for exception), particularly across various behavioral topographies. Bowring et al. (2017), for example, found congregate housing to be a significant risk for self-injury and stereotyped behaviors, while Folch et al. (2018) did not find such a relationship. Given that most children and adolescents are likely to live in their family home, it should not be a surprise that only one study has shown an increased risk of challenging behavior related to living in a residential setting for younger populations (Dworschak et al., 2016).

Biomedical Conditions Few biomedical conditions or complications have proven to be a risk factor for challenging behavior in the ID and dual diagnosis populations. Probably the most studied condition is epilepsy, which most studies have not found to be a risk factor (Cooper, Smiley, Jackson, et al., 2009; de Winter, Jansen, & Evenhuis, 2011; Folch et al., 2018; Holden & Gitlesen, 2006; Jones et al., 2008; O'Dwyer et al., 2018). Mixed findings have been obtained with regard to urinary or bowel incontinence (Bowring et al., 2017; de Winter et al., 2011; Jones et al., 2008; O'Dwyer et al., 2018), mobility issues or physical disability (Bowring et al., 2017; de Winter et al., 2011; Holden & Gitlesen, 2006; Jones et al., 2008), and visual impairment (Bowring et al., 2017; de Winter et al., 2011; Holden & Gitlesen, 2006; Jones et al., 2008), while no support has been evident for hearing impairment (Bowring et al., 2017; de Winter et al., 2011; Holden & Gitlesen, 2006) as a risk factor for challenging behavior.

Daytime Activity Engagement Involvement in daytime activities, such as day habilitation, vocational training, voluntary work, or education, has been explored as a risk factor in a number of studies. Generally, lack of daytime activity engagement has been identified as a risk factor for challenging behavior (Bowring et al., 2017; Lowe et al., 2007). An exception is a study by O'Dwyer et al. (2018) that failed to identify an association between daytime occupation and challenging behavior; however, this study only included individuals over the age of 40 years, which may be less capable of enjoying daytime engagement. It seems likely that either daytime activity engagement protects against challenging behavior by keeping an individual busy during the day or that challenging behavior precludes involvement in daytime activities.

Summary of Risk Factors

Studies on the risk factors associated with challenging behavior in individuals with ID have identified few robust associations and virtually no putative risk factors without some contradictory evidence. Among the most consistent findings are that challenging behavior is more likely among individuals with more severe ID, greater communication deficits, general psychopathology, and residing in congregate housing. Too few studies specific to challenging behavior in dual-diagnosed individuals has been conducted to provide strong evidence of risk factors. Possible associations exist with regard to severity of ID, gender, age, and residential setting, but more study is necessary for confirmation.

The Etiology of Challenging Behavior

Unlike risk factors, theories on the causes of challenging behavior assist in explaining why a

behavior is emitted, rather than merely indicating an association between behavior and another variable. Like risk factors, it is unlikely that a monolithic theory of causality will explain all challenging behavior observed in individuals with ID or dual diagnosis. Several prominent theories on causality have been offered, with varying levels of research to support them. A few theories described later may be more relevant to dual diagnosis because there is a focus on the overlap between challenging behavior and psychopathology, but others are germane to the ID population and do not involve discussion on psychopathology as part of the cause. It is important to point out that theories of causality for psychopathology (and thus dual diagnosis) also exist, but the following section will only attempt to describe theories used to explain challenging behavior among individuals with ID and dual diagnosis without conflating the two. The most common theories are outlined next.

Challenging Behavior as a Learned Response

The most prominent theory with the greatest research support explains challenging behavior as a learned response through operant conditioning (see Skinner, 1938). In this theoretical model, challenging behavior is a product of the environmental stimuli that evoke it (i.e., the antecedents) and the environmental stimuli that maintain or reinforce it (i.e., consequences). Thus, behavior is said to be selected by the consequences of the environment, otherwise called reinforcement. Basically, an individual is more likely to display a behavior in the future if the occurrence of that behavior is followed by a beneficial outcome (i.e., reinforcement) and less likely to display a behavior in the future if the beneficial outcome is absent (i.e., extinction) or if it is followed by an aversive outcome (i.e., punishment).

When a challenging behavior is emitted, it is the product of a history of reinforcement, which suggests it has a purpose or function. Challenging behavior, like adaptive behavior, may be maintained via social reinforcement, automatic reinforcement, or both. When challenging behavior is socially maintained, the behavior serves to communicate to or influence another person to produce the reinforcement. Broad classes of social reinforcement include attention, preferred items or activities, and escape or avoidance of aversive contexts (e.g., academic demands, loud sporting event). Attention and preferred items/activities are considered positive reinforcers, and escape/avoidance is considered negative reinforcement. When challenging behavior is automatically maintained, the behavior produces its own beneficial outcome, such as pleasurable sensory stimulation (automatic positive reinforcement) or reduction of an aversive state or sensation (automatic negative reinforcement).

Identification of the function of challenging behavior is done through functional behavioral assessment (FBA). The FBA process may include indirect methods, such as rating scales or caregiver interviews about contexts that might occasion challenging behavior, or direct observation, including correlational descriptive assessments and functional (experimental) analyses of the challenging behavior. Experimental analyses are the most rigorous method of assessment and are considered the "gold standard" of behavioral assessment for challenging behavior (Oliver, Pratt, & Normand, 2015). Iwata, Dorsey, Slifer, Bauman, and Richman (1982/1994) developed a standardized method of experimental analysis, called a functional analysis (FA), which involves systematic manipulation of antecedents and consequences within a multielement single-case design (Ulman & Sulzer-Azaroff, 1975). In the seminal study describing the FA, Iwata and colleagues compared rates of self-injury in three test conditions to a control condition to determine whether self-injury in nine individuals with developmental delay was maintained by social positive reinforcement, social negative reinforcement, and/or automatic reinforcement. For eight of nine participants, the rates of self-injury were sufficiently divergent from the control condition to suggest a function or functions. Since Iwata et al.'s original study, thousands of replications and extensions have been conducted, including numerous refinements and variations to assist in

the identification of function(s) for a variety of challenging behaviors across a variety of populations and settings (Beavers, Iwata, & Lerman, 2013; Lydon, Healy, O'Reilly, & Lang, 2012).

Support for this theory comes from research on treatments utilizing operant learning principles to reduce challenging behavior and/or increase adaptive behaviors. Treatments tied to the outcome of FBA have proven to be more effective than those developed without regard to the function of the challenging behavior (Newcomer & Lewis, 2004; Payne, Scott, & Conroy, 2007). In the ID population in particular, treatments utilizing a function-based approach for challenging behavior have been superior to traditional psychotherapies (Didden, Korzilius, Van Oorsouw, & Sturmey, 2006; Doehring, Reichow, Palka, Phillips, & Hagopian, 2014; Rush & Frances, 2000). Common function-based treatments often involve a combination of reinforcing more appropriate behaviors or the absence of challenging behavior (i.e., differential reinforcement) and withholding the known functional reinforcer for a targeted challenging behavior (i.e., extinction; Vollmer & Iwata, 1992). For example, functional communication training or FCT (Carr & Durand, 1985) is a function-based treatment that entails differential reinforcement of communication behavior matched to the function of the targeted challenging behavior and extinction of the challenging behavior (Tiger, Hanley, & Bruzek, 2008). Using FCT, a child who displays property destruction to get attention may be taught to press a microswitch for attention instead, while the property destruction would be ignored and/or blocked. Given the high prevalence of communication challenges in the ID population (Pinborough-Zimmerman et al., 2007) and concomitant challenging behavior, this approach is well suited for individuals with ID. Indeed, FCT is an evidence-based approach for challenging behavior in the ID population (Gerow, Davis, Radhakrishnan, Gregori, & Rivera, 2018).

Despite an exceptionally large quantity of studies using function-based approaches to treat challenging behavior, the majority utilize single-case design, which has been a barrier to substantiating the effectiveness of this approach (Stoiber, Purdy, & Klingbeil, 2016). Nonetheless, large meta-analyses and systematic reviews which have consistently shown treatments based on an operant learning model are effective in treating a wide range of challenging behaviors among individuals with ID (for an overview see Sturmey & Hamelin, 2014), providing strong support for this theoretical explanation.

Psychiatric/Behavioral Equivalents Explanations

For individuals with ID or dual diagnosis, challenging behavior may be a symptom of a psychiatric disorder, according to a theory offered by Emerson (2001). In this theory, such behavior represents an atypical manifestation of a mental illness. According to this theory, individuals with and without ID are exposed to the same life stressors, such as loss of a loved one, but display a different response. While such life stressors may result in symptoms of a psychiatric disorder in most individuals, in someone with ID, it may be expressed as a challenging behavior. The challenging behaviors substituting typical symptoms of psychopathology have been called "behavioral equivalents."

Support for a behavioral equivalents theory comes from studies that have shown an increased risk of psychopathology in individuals with ID (Holden & Gitlesen, 2003; Moss et al., 2000). Most studies of behavioral equivalents have focused on the connection between depression and challenging behavior (Meins, 1995). For example, Reiss and Rojahn (1993) reported that individuals with ID who exhibited aggression were more likely to have a diagnosis of depression. Some have countered that the high rates of psychiatric diagnoses in the ID population is an overestimate due to the misperception that challenging behavior is a behavioral equivalent and an indicator of psychopathology (Whitaker & Read, 2006). Moreover, numerous studies have failed to demonstrate support for this theory (Grieve, Jones, & Slupek, 2007; McBrien, 2003; Melville et al., 2016; Rojahn et al., 2004; Rojahn,

Borthwick-Duffy, & Jacobson, 1993; Tsiouris, Mann, Patti, & Sturmey, 2003).

Emerson (2001) has suggested two additional explanations for the occurrence of challenging behavior in individuals with ID. One explanation is that challenging behavior is a secondary symptom of psychopathology. According to this hypothesis, the challenging behavior is not necessarily a direct symptom of psychopathology but an inability to express feelings associated with the psychopathology due to skill deficits. For example, an individual with depression who is feeling sadness may display aggression toward others due to the inability to communicate this feeling. This explanation for challenging behavior has not been a focus of much research and thus has not garnered much research support.

A third explanation offered by Emerson (2001) extends the operant learning model by suggesting that psychiatric symptoms serve as setting events for challenging behavior. Based upon this explanation, individuals experiencing psychopathology may have a reduced tolerance for aversive environmental stimuli (e.g., an academic demand) and thus increase the value of reinforcement associated with its termination (e.g., a break from the demand). There is a plethora of studies that have established poor physical health as a setting event for challenging behavior (see next section) but very little examining mental health as a setting event. A study by Sovner, Fox, Lowry, and Lowry (1993) provides indirect support for this explanation. Sovner and colleagues showed that for two adults ID with depressive symptoms, concomitant SIB reduced substantially via antidepressant medication (i.e., fluoxetine), as did their depressive features. They suggested that the depressive symptoms served as a setting event for challenging behavior and was attenuated with the medication.

Biomedical and Neurobiological Models

Biomedical and neurobiological models attempt to explain challenging behavior as a result of biomedical conditions and/or brain dysfunction. While research supporting these models has generally focused on challenging behavior as a whole, there is considerably more research on biomedical and neurobiological models of self-injury than other topographies.

Dysregulation among neurotransmitters, which are thought of as chemical messengers in the brain, has been implicated in several theories specific to self-injurious behavior. One prominent theory of SIB offered by Sandman (1990) and dubbed the "opiate hypothesis" suggests that for some individuals with ID, there is an insensitivity to pain (hypoalgesia), and acts of SIB result in a release of endogenous opiates. This release produces a euphoria "high," increasing the likelihood of additional SIB (i.e., SIB is automatically maintained and repeated). There is support for this hypothesis involving studies of naltrexone, an opiate antagonist, as a treatment for SIB. Several studies have reported decreases in SIB among individuals with ID-prescribed naltrexone (Roy, Roy, Deb, Unwin, & Roy, 2015; Sandman & Kemp, 2011). Additional theories involving dysregulation of other neurotransmitters, including reduced dopamine (Breese et al., 1995) and reduced serotonin (Kolevzon et al., 2014), exist, but with less research support.

Several regions of the brain have been linked to challenging behavior, but support is limited, or it is speculatively based on nonhuman animal research. Poor inhibitory control associated with frontal cortex dysfunction, as well as increased activation of the amygdala (fear responding) and the hypothalamus (emotional processing), has been suggested as contributors to aggression (Anderson, Bechara, Damasio, Tranel, & Damasio, 1999; Siever, 2008). Studies of repetitive behaviors and stereotypy suggest that impairment in the basal ganglia and the fronto-striatal circuits (motor movement and inhibition) may be the origin of such behavior (Bodfish & Lewis, 2002). The basal ganglia, and specifically the striatum, has also been a region of focus in the study of self-injury (Turner & Lewis, 2002), which provides credence to theories that stereotyped behaviors evolve into self-injury for some individuals (Kennedy, 2002).

The link between challenging behavior and biomedical conditions and complications has been explained in two ways: first, some behaviors may be reinforced via the attenuation of unpleasant sensations associated with a medical condition. For example, studies by Christensen et al. (2009) and Swender, Matson, Mayville, González, and McDowell (2006) found that SIB was automatically maintained but correlated with bouts of constipation and gastroesophageal reflux, respectively. Presumably, the SIB was negatively reinforced by the reduction of pain of their medical conditions. In fact, in the Christensen study, a bowel clean-out using a laxative resulted in a reduction of SIB, providing support for this hypothesis.

A second manner that biomedical conditions may contribute to challenging behavior is by acting as setting events. As noted previously, setting events are distal factors that change someone's threshold for aversive environmental events. For example, Kennedy and Meyer (1996) reported on the results of an assessment for an adolescent with ID who showed SIB to escape demands but did so only when allergy symptoms were present. Studies with similar findings have been reported for individuals with sleep problems (O'Reilly, 1995; O'Reilly & Lancioni, 2000), ear infections (Luiselli, Cochran, & Huber, 2005; O'Reilly, 1997), and onset of menses (Carr, Smith, Giacin, Whelan, & Pancari, 2003). Unlike the reduction of pain model above, this hypothesis does not suggest that biomedical conditions cause challenging behavior; rather, they increase the likelihood that such behavior will occur under specific motivating conditions.

Genetics and Behavioral Phenotypes

Challenging behavior is more common in certain genetic syndromes, giving rise to a genetic explanation of such behavior (Oliver et al., 2013). This model is concerned with behavioral phenotypes, which are specific behavioral patterns associated with particular genetic syndromes often accompanied by ID. Unlike environmental theories, explanations based on behavioral phenotypes suggest that individuals with ID or dual diagnosis exhibit challenging behaviors due to a variety of possible deviations from the normal DNA sequence. More specifically, genetic syndromes are a result of gene mutations (e.g., fragile X syndrome) or chromosomal abnormalities (e.g., Cornelia de Lange syndrome), which leads to higher (or sometimes lower) probabilities of challenging behavior.

Numerous genetic syndromes have a strong association with higher rates of aggression, self-injury, or both in comparison with the non-syndromic ID population. Lesch-Nyhan syndrome, for example, is the most prominent diagnosis associated with self-injury, with estimates of nearly 100% of individuals with this diagnosis displaying SIB (Fu & Jinnah, 2012). Arron et al. (2011) surveyed the caregivers of nearly 800 individuals with one of several genetic syndromes known to have high rates of challenging behavior and compared rates of SIB and aggression to a large group of individuals with heterogeneous ID etiology. They found significantly higher prevalence rates of individuals exhibiting SIB with a diagnosis of cri du chat, Cornelia de Lange, fragile X, Prader-Willi, Lowe, and Smith-Magenis syndromes, with the rates observed in Smith-Magenis and cri du chat syndromes, at 92.9% and 75.8%, respectively. Two syndromes, Angelman and Smith-Magenis, exhibited significantly higher rates of physical aggression (73%) than the control group. Additionally, some syndromes in the Arron study were significantly more likely to be associated with specific topographies of self-injury (e.g., "rubs or scratches self" was significantly higher in individuals with cri du chat and Prader-Willi syndromes). Studies, such as one by Arron et al., which demonstrate a relationship between a genetic syndrome and challenging behavior above chance, provide support for the behavioral phenotype explanation.

Some genetic syndromes are also more likely to be associated with various types of psychopathology (and dual diagnosis), which may function as a mediating or moderating variable to help explain challenging behavior. An example of this relationship is seen in Cornelia de Lange and

fragile X syndromes. Both of these diagnoses are associated with greater risk of social anxiety, which in turn may lead to more challenging behavior to escape or avoid social situations (Hall, DeBernardis, & Reiss, 2006; Richards, Moss, O'Farrell, Kaur, & Oliver, 2009). Angelman syndrome (Clayton-Smith, 2001), Prader-Willi syndrome (O'Brien & Bevan, 2011), Williams syndrome (O'Brien & Bevan, 2011), and Smith-Magenis syndrome (Dykens & Smith, 1998) are among the genetic syndromes with higher risk of psychopathology. Interestingly, all but one of these diagnoses (Williams syndrome) are associated with higher rates of challenging behavior, providing support for a link between psychopathology and challenging behavior.

Mixed Models of Causation

There is great support for an environmental explanation of challenging behavior in the ID and dual diagnosis populations, especially through the lens of the operant learning model. There is also plenty of evidence to support the contribution of genetics and biomedical conditions to the etiology of challenging behavior. It is unlikely that the etiology of all challenging behavior in ID and dual diagnosis populations can be found in one model or theory. Rather, a comprehensive model that incorporates both environmental and organic etiological explanations will likely be more fruitful and create a stronger understanding of the cause(s) and therefore the best approaches to treatment. Recent efforts at mixed-model explanations exist. For example, Tunnicliffe and Oliver (2011) conducted a review on operant studies with individuals diagnosed with genetic syndromes and reported that challenging behavior across a broad range of genetic syndromes may be highly influenced by the environment. Moreover, they point out the possible causal pathways associated with particular genetic syndromes. To illustrate, Tunnicliffe and Oliver report that individuals with Angelman and Smith-Magenis syndrome have behavioral phenotypes with a predilection to seek out the attention of others, which in turn results in more challenging behavior to gain others' attention. Impulsivity often observed in Soto's syndrome, reduced pain perception in Smith-Magenis syndrome, and social anxiety in fragile X syndrome are additional examples of biobehavioral factors that may influence challenging behavior probabilities under certain environmental conditions. Even so, more research is needed to fully understand this relationship and to build a comprehensive biobehavioral model.

Conclusion

Differing definitions, research methodologies, and findings have led to difficulties in synthesizing the research literature on challenging behavior in individuals with ID or dual diagnosis. However, some conclusions may be brought forth through the research that exists. It is apparent that prevalence rates of challenging behaviors are relatively high in individuals with ID, and in some diagnoses where ID is common, challenging behaviors may be significantly more or less likely than the general ID population. Although risk for challenging behavior is multifactorial, it is apparent that individuals with more severe ID, greater communication deficits, co-occurring psychopathology, and living in institutional or restricted residential settings are more likely to exhibit challenging behavior. Finally, while no solitary theory can claim to explain the etiology of all challenging behavior in individuals with ID or dual diagnosis, a biobehavioral model that acknowledges both environmental and organic contributions shows promise.

Despite what we know about the relationship between challenging behavior and intellectual disability, the connection between intellectual disability, psychopathology, and challenging behavior is deserving of more research. Given the high comorbidity rate associated with ID, it is not unreasonable to make assumptions regarding the relationship between dual diagnosis and challenging behavior based upon research that does not consider co-occurring psychopathology. Indeed, this chapter has largely relied on research

literature of this type. However, an improved understanding of the role psychopathology plays in the development and maintenance of challenging behavior and the direction of the relationship will be beneficial for the future development of preventive and treatment efforts.

References

Allen, D. (2000). Recent research on physical aggression in persons with intellectual disability: An overview. *Journal of Intellectual and Developmental Disability, 25*(1), 41–57. https://doi.org/10.1080/132697800112776

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. https://doi.org/10.1111/j.1365-2788.2006.00877.x

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

Anderson, L. T., & Ernst, M. (1994). Self-injury in Lesch-Nyhan disease. *Journal of Autism and Developmental Disorders, 24*(1), 67–81. https://doi.org/10.1007/bf02172213

Anderson, S. W., Bechara, A., Damasio, H., Tranel, D., & Damasio, A. R. (1999). Impairment of social and moral behavior related to early damage in human prefrontal cortex. *Nature Neuroscience, 2*(11), 1032–1037. https://doi.org/10.1038/14833

Antonacci, D. J., Manuel, C., & Davis, E. (2008). Diagnosis and treatment of aggression in individuals with developmental disabilities. *Psychiatric Quarterly, 79*(3), 225–247. https://doi.org/10.1007/s11126-008-9080-4

Arron, K., Oliver, C., Moss, J., Berg, K., & Burbidge, C. (2011). The prevalence and phenomenology of self-injurious and aggressive behaviour in genetic syndromes. *Journal of Intellectual Disability Research, 55*(2), 109–120. https://doi.org/10.1111/j.1365-2788.2010.01337.x

Axmon, A., Bjorne, P., Nylander, L., & Ahlstrom, G. (2018). Psychiatric diagnoses in relation to severity of intellectual disability and challenging behaviors: A register study among older people. *Aging & Mental Health, 22*(10), 1344–1350. https://doi.org/10.1080/13607863.2017.1348483

Baker, B., McIntyre, 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*, 217–230. https://doi.org/10.1046/j.1365-2788.2003.00484.x

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

Bodfish, J. W. (2007). Stereotypy, self-injury, and related abnormal repetitive behaviors. In *Handbook of intellectual and developmental disabilities* (pp. 481–505). New York, NY: Springer.

Bodfish, J. W., Crawford, T. W., Powell, S. B., Parker, D. E., Golden, R. N., & Lewis, M. H. (1995). Compulsions in adults with mental retardation: Prevalence, phenomenology, and comorbidity with stereotypy and self-injury. *American Journal of Mental Retardation, 100*(2), 183–192.

Bodfish, J. W., & Lewis, M. H. (2002). Self-injury and comorbid behaviors in developmental, neurological, psychiatric, and genetic disorders. In *Self-injurious behavior: Gene-brain-behavior relationships* (pp. 23–39). Washington, DC: American Psychological Association.

Bodfish, J. W., Symons, F. J., Parker, D. E., & Lewis, M. H. (2000). Varieties of repetitive behavior in autism: Comparisons to mental retardation. *Journal of Autism and Developmental Disorders, 30*(3), 237–243. https://doi.org/10.1023/a:1005596502855

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

Bowring, D. L., Totsika, V., Hastings, R. P., Toogood, S., & Griffith, G. 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*(1), 16–32. https://doi.org/10.1111/bjc.12118

Bramble, D. (2007). Psychotropic drug prescribing in child and adolescent learning disability psychiatry. *Journal of Psychopharmacology, 21*(5), 486–491. https://doi.org/10.1177/0269881106075642

Breese, G. R., Criswell, H. E., Duncan, G. E., Moy, S. S., Johnson, K. B., Wong, D. F., & Mueller, R. A. (1995). Model for reduced brain dopamine in Lesch-Nyhan syndrome and the mentally retarded: Neurobiology of neonatal-6-hydroxydopamine-lesioned rats. *Mental Retardation and Developmental Disabilities Research Reviews, 1*(2), 111–119. https://doi.org/10.1002/mrdd.1410010207

Buono, S., Scannella, F., & Palmigiano, M. B. (2010). Self-injurious behavior: A comparison between Prader-Willi syndrome, Down syndrome and Autism. *Life Span and Disability, 13*, 187–201.

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., Smith, C. E., Giacin, T. A., Whelan, B. M., & Pancari, J. (2003). Menstrual discomfort as a biological setting event for severe problem behavior: Assessment and intervention. *American Journal on*

Mental Retardation, 108(2), 117–133. https://doi.org/10.1352/0895-8017(2003)108<0117:MDAABS>2.0.CO;2

Chadwick, O., Piroth, N., Walker, J., Bernard, S., & Taylor, E. (2000). Factors affecting the risk of behaviour problems in children with severe intellectual disability. *Journal of Intellectual Disability Research, 44*, 108–123.

Chan, J., & Sigafoos, J. (2000). A review of child and family characteristics related to the use of respite care in developmental disability services. *Child & Youth Care Forum, 29*(1), 27–37. https://doi.org/10.1023/A:1009420206722

Christensen, T. J., Ringdahl, J. E., Bosch, J. J., Falcomata, T. S., Luke, J. R., & Andelman, M. S. (2009). Constipation associated with self-injurious and aggressive behavior exhibited by a child diagnosed with autism. *Education and Treatment of Children, 32*(1), 89–103.

Clark, B., Vandermeer, B., Simonetti, A., & Buka, I. (2010). Is lead a concern in Canadian autistic children? *Paediatrics & Child Health, 15*(1), 17–22. https://doi.org/10.1093/pch/15.1.17

Clayton-Smith, J. (2001). Angelman syndrome: Evolution of the phenotype in adolescents and adults. *Developmental Medicine and Child Neurology, 43*(7), 476–480.

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

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. https://doi.org/10.1111/j.1365-2788.2008.01127.x

Cooper, S.-A., Smiley, E., Morrison, J., Williamson, A., & Allan, L. (2007). An epidemiological investigation of affective disorders with a population-based cohort of 1023 adults with intellectual disabilities. *Psychological Medicine, 37*(6), 873–882. https://doi.org/10.1017/S0033291707009968

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. https://doi.org/10.1111/j.1365-2788.2006.00815.x

Danford, D. E., & Huber, A. M. (1982). Pica among mentally retarded adults. *American Journal of Mental Deficiency, 87*(2), 141–146.

Danquah, A., Limb, K., Chapman, M., Burke, C., Flood, A., Gore, S., … Hare, D. J. (2009). An investigation of factors predictive of continued self-injurious behaviour in an intellectual disability service. *Journal of Applied Research in Intellectual Disabilities, 22*(4), 395–399. https://doi.org/10.1111/j.1468-3148.2008.00470.x

Davies, L., & Oliver, C. (2013). The age related prevalence of aggression and self-injury in persons with an intellectual disability: A review. *Research in Developmental Disabilities, 34*(2), 764–775. https://doi.org/10.1016/j.ridd.2012.10.004

Davies, L. E., & Oliver, C. (2016). Self-injury, aggression and destruction in children with severe intellectual disability: Incidence, persistence and novel, predictive behavioural risk markers. *Research in Developmental Disabilities, 49-50*, 291–301. https://doi.org/10.1016/j.ridd.2015.12.003

de Winter, C. F., Jansen, A. A. C., & Evenhuis, H. M. (2011). Physical conditions and challenging behaviour in people with intellectual disability: A systematic review. *Journal of Intellectual Disability Research, 55*(7), 675–698. https://doi.org/10.1111/j.1365-2788.2011.01390.x

Dekker, M. C., Koot, H. M., Ende, J. V. D., & Verhulst, F. C. (2002). Emotional and behavioral problems in children and adolescents with and without intellectual disability. *Journal of Child Psychology and Psychiatry, 43*(8), 1087–1098. https://doi.org/10.1111/1469-7610.00235

Didden, R., Korzilius, H., & Curfs, L. M. G. (2007). Skin-picking in individuals with Prader-Willi syndrome: Prevalence, functional assessment and its comorbidity with compulsive and self-injurious behaviours. *Journal of Applied Research in Intellectual Disabilities, 20*, 409–419. https://doi.org/10.1111/j.1468-3148.2007.00388.x

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 of Mental Retardation, 111*(4), 290–298. https://doi.org/10.1352/0895-8017(2006)111[290:Btocbi]2.0.Co;2

Dinya, E., Csorba, J., Suli, A., & Grosz, Z. (2012). Behaviour profile of Hungarian adolescent outpatients with a dual diagnosis. *Research in Developmental Disabilities, 33*(5), 1574–1580. https://doi.org/10.1016/j.ridd.2012.03.001

Doehring, P., Reichow, B., Palka, T., Phillips, C., & Hagopian, L. (2014). Behavioral approaches to managing severe problem behaviors in children with autism spectrum and related developmental disorders: A descriptive analysis. *Child and Adolescent Psychiatric Clinics of North America, 23*(1), 25–40. https://doi.org/10.1016/j.chc.2013.08.001

Drews, C. D., Yeargin-Allsopp, M., Decouflé, P., & Murphy, C. C. (1995). Variation in the influence of selected sociodemographic risk factors for mental retardation. *American Journal of Public Health, 85*(3), 329–334. https://doi.org/10.2105/AJPH.85.3.329

Dudley, J. R., Ahlgrim-Delzell, L., & Calhoun, M. (1999). Diverse diagnostic and behavioural patterns amongst people with a dual diagnosis. *Journal of Intellectual Disability Research, 43*, 70–79.

Dworschak, W., Ratz, C., & Wagner, M. (2016). Prevalence and putative risk markers of challenging behavior in students with intellectual disabilities. *Research in Developmental Disabilities, 58*, 94–103. https://doi.org/10.1016/j.ridd.2016.08.006

Dykens, E., & Smith, A. (1998). Distinctiveness and correlates of maladaptive behaviour in children and adolescents with Smith-Magenis syndrome. *Journal of Intellectual Disability Research, 42*, 481–489.

Dykens, E. M. (2000). Contaminated and unusual food combinations: What do people with Prader-Willi syndrome choose? *Mental Retardation, 38*(2), 163. https://doi.org/10.1352/0047-6765(2000)038<0163:CAUFCW>2.0.CO

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. https://doi.org/10.1080/13668250.2011.572548

Emberson, J., & Walker, E. (1990). Self-injurious behaviour in people with a mental handicap. *Nursing Times, 86*(23), 43–46.

Emerson, E. (2001). *Challenging behaviour: Analysis and intervention in people with severe intellectual disabilities*. Cambridge, UK: Cambridge University Press.

Emerson, E., Alborz, A., Reeves, D., Mason, H., Swarbrick, R., Kiernan, C., & Mason, L. (1997). *The HARC challenging behaviour project. Report 2: The prevalence of challenging behaviour*. Manchester, UK: Hester Adrian Research Centre, University of Manchester.

Emerson, E., & Bromley, J. (1995). The form and function of challenging behaviors. *Journal of Intellectual Disability Research, 39*, 388–398.

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

Felce, D., & Kerr, M. (2013). Investigating low adaptive behaviour and presence of the triad of impairments characteristic of autistic spectrum disorder as indicators of risk for challenging behaviour among adults with intellectual disabilities. *Journal of Intellectual Disability Research, 57*(2), 128–138. https://doi.org/10.1111/j.1365-2788.2011.01524.x

Felce, D., Kerr, M., & Hastings, R. P. (2009). A general practice-based study of the relationship between indicators of mental illness and challenging behaviour among adults with intellectual disabilities. *Journal of Intellectual Disability Research, 53*(3), 243–254. https://doi.org/10.1111/j.1365-2788.2008.01131.x

Finlay, W., & Lyons, E. (2001). Methodological issues in interviewing and using self-report questionnaires with people with mental retardation. *Psychological Assessment, 13*(3), 319–335. https://doi.org/10.1037/1040-3590.13.3.319

Finucane, B. M., Konar, D., Haas-Givler, B., Kurtz, M. B., & Scott, C. I., Jr. (1994). The spasmodic upper-body squeeze: A characteristic behavior in Smith-Magenis syndrome. *Developmental Medicine and Child Neurology, 36*(1), 78–83. https://doi.org/10.1111/j.1469-8749.1994.tb11770.x

Folch, A., Cortes, M. J., Salvador-Carulla, L., Vicens, P., Irazabal, M., Munoz, S., … Martinez-Leal, R. (2018). Risk factors and topographies for self-injurious behaviour in a sample of adults with intellectual developmental disorders. *Journal of Intellectual Disability Research, 62*(12), 1018–1029. https://doi.org/10.1111/jir.12487

Fu, R., & Jinnah, H. A. (2012). Genotype-phenotype correlations in Lesch-Nyhan disease moving beyond the gene. *Journal of Biological Chemistry, 287*(5), 2997–3008.

Gentile, J. P., Gillig, P. M., Stinson, K., & Jensen, J. (2014). Toward impacting medical and psychiatric comorbidities in persons with intellectual/developmental disabilities: An initial prospective analysis. *Innovations in Clinical Neuroscience, 11*(11–12), 22–26.

Gerow, S., Davis, T., Radhakrishnan, S., Gregori, E., & Rivera, G. (2018). Functional communication training: The strength of evidence across disabilities. *Exceptional Children, 85*(1), 86–103. https://doi.org/10.1177/0014402918793399

Goldin, R. L., Matson, J. L., & Cervantes, P. E. (2014). The effect of intellectual disability on the presence of comorbid symptoms in children and adolescents with autism spectrum disorder. *Research in Autism Spectrum Disorders, 8*(11), 1552–1556. https://doi.org/10.1016/j.rasd.2014.08.006

Grey, I., Pollard, J., McClean, B., MacAuley, N., & Hastings, R. (2010). Prevalence of psychiatric diagnoses and challenging behaviors in a community-based population of adults with intellectual disability. *Journal of Mental Health Research in Intellectual Disabilities, 3*(4), 210–222. https://doi.org/10.1080/19315864.2010.527035

Grieve, A., Jones, A., & Slupek, S. (2007). A study of the relationships between mental illness, epilepsy and challenging behaviour in a sample of people with learning disabilities living in the Highlands of Scotland. *Clinical Psychology and People with Learning Disabilities, 5*, 2–9.

Griffin, J. C., Ricketts, R. W., Williams, D. E., Locke, B. J., Altmeyer, B. K., & Stark, M. T. (1987). A community survey of self-injurious behavior among developmentally disabled children and adolescents. *Psychiatric Services, 38*(9), 959–963. https://doi.org/10.1176/ps.38.9.959

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. https://doi.org/10.1016/0270-3092(86)90022-6

Hall, S., DeBernardis, M., & Reiss, A. (2006). Social escape behaviors in children with fragile X syndrome. *Journal of Autism and Developmental Disorders, 36*(7), 935–947.

Hall, S. S., Lightbody, A. A., & Reiss, A. L. (2008). Compulsive, self-injurious, and autistic behavior in children and adolescents with fragile X syndrome. *American Journal of Mental Retardation, 113*(1), 44–53. https://doi.org/10.1352/0895-8017(2008)113[44:Csaabi]2.0.Co;2

Harper, K. (2019). Aggression. In J. P. Gentile, A. E. Cowan, & D. W. Dixon (Eds.), *Guide to intellectual disabilities: A clinical handbook* (pp. 101–120). Cham, Switzerland: Springer International Publishing.

Havercamp, S. M., & Reiss, S. (1997). The Reiss screen for maladaptive behavior: Confirmatory factor analysis. *Behaviour Research and Therapy, 35*(10), 967–971. https://doi.org/10.1016/S0005-7967(97)00043-0

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*(11), 858–864. https://doi.org/10.1111/j.1365-2788.2005.00723.x

Hayes, S., McGuire, B., O'Neill, M., Oliver, C., & Morrison, T. (2011). Low mood and challenging behaviour in people with severe and profound intellectual disabilities. *Journal of Intellectual Disability Research, 55*(2), 182–189. https://doi.org/10.1111/j.1365-2788.2010.01355.x

Hessl, D., Dyer-Friedman, J., Glaser, B., Wisbeck, J., Barajas, R. G., Taylor, A., & Reiss, A. L. (2001). The influence of environmental and genetic factors on behavior problems and autistic symptoms in boys and girls with fragile X syndrome. *Pediatrics, 108*(5), E88. https://doi.org/10.1542/peds.108.5.e88

Hillery, J., & Mulcahy, M. (1997). Self-injurious behaviour in persons with a mental handicap: An epidemiological study in an Irish population. *Irish Journal of Psychological Medicine, 14*(1), 12–15. https://doi.org/10.1017/S0790966700002834

Holden, B., & Gitlesen, J. P. (2003). Prevalence of psychiatric symptoms in adults with mental retardation and challenging behaviour. *Research in Developmental Disabilities, 24*(5), 323–332.

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

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

Huisman, S., Mulder, P., Kuijk, J., Kerstholt, M., van Eeghen, A., Leenders, A., … Hennekam, R. (2018). Self-injurious behavior. *Neuroscience & Biobehavioral Reviews, 84*, 483–491. https://doi.org/10.1016/j.neubiorev.2017.02.027

Iwata, B. A., Dorsey, M. F., Slifer, K. J., Bauman, K. E., & Richman, G. S. (1994). Toward a functional analysis or self-injury. *Journal of Applied Behavior Analysis, 27*(2), 197–209. https://doi.org/10.1901/jaba.1994.27-197

Jahoda, A., Willner, P., Pert, C., & MacMahon, K. M. A. (2013). From causes of aggression to interventions: The importance of context. In R. Hastings & J. Rojahn (Eds.), *International review of research in developmental disabilities* (Vol. 44, pp. 69–104). Amsterdam: Elsevier.

Jones, R., Wint, D., & Ellis, N. C. (1990). The social effects of stereotyped behavior. *Journal of Mental Deficiency Research, 34*, 261–268.

Jones, S., Cooper, S. A., Smiley, E., Allan, L., Williamson, A., & Morrison, J. (2008). Prevalence of, and factors associated with, problem behaviors in adults with intellectual disabilities. *Journal of Nervous and Mental Disease, 196*(9), 678–686. https://doi.org/10.1097/NMD.0b013e318183f85c

Kahng, S., Iwata, B. A., & Lewin, A. B. (2002). Behavioral treatment of self-injury, 1964 to 2000. *American Journal of Mental Retardation, 107*(3), 212–221. https://doi.org/10.1352/0895-8017(2002)107<0212:BTOSIT>2.0.CO

Kennedy, C. H. (2002). Evolution of stereotypy into self-injury. In *Self-injurious behavior: Gene-brain-behavior relationships* (pp. 133–143). Washington, DC: American Psychological Association.

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

Klintwall, L., & Eikeseth, S. (2012). Number and controllability of reinforcers as predictors of individual outcome for children with autism receiving early and intensive behavioral intervention: A preliminary study. *Research in Autism Spectrum Disorders, 6*(1), 493–499. https://doi.org/10.1016/j.rasd.2011.07.009

Knapp, M., Comas-Herrera, A., Astin, J., Beecham, J., & Pendaries, C. (2005). Intellectual disability, challenging behaviour and cost in care accommodation: What are the links? *Health & Social Care in the Community, 13*(4), 297–306. https://doi.org/10.1111/j.1365-2524.2005.00539.x

Ko, C., Lunsky, Y., Hensel, J., & Dewa, C. S. (2012). Burnout among summer camp staff supporting people with intellectual disability and aggression. *Intellectual and Developmental Disabilities, 50*(6), 479–485. https://doi.org/10.1352/1934-9556-50.06.479

Kolevzon, A., Lim, T., Schmeidler, J., Martello, T., Cook, E. H., Jr., & Silverman, J. M. (2014). Self-injury in autism spectrum disorder: An effect of serotonin transporter gene promoter variants. *Psychiatry Research, 220*(3), 987–990. https://doi.org/10.1016/j.psychres.2014.09.018

Koritsas, S., & Iacono, T. (2012). Challenging behaviour and associated risk factors: An overview (part I). *Advances in Mental Health and Intellectual Disabilities, 6*(4), 199–214. https://doi.org/10.1108/20441281211236643

Koritsas, S., & Iacono, T. (2015). Predictors of challenging behaviour in adults with intellectual disability. *Advances in Mental Health and Intellectual Disabilities, 9*(6), 312–326. https://doi.org/10.1108/AMHID-06-2015-0029

Lanovaz, M. J., & Sladeczek, I. E. (2012). Vocal stereotypy in individuals with autism spectrum disorders: A review of behavioral interventions. *Behavior Modification, 36*(2), 146–164. https://doi.org/10.1177/0145445511427192

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*(4), 562–593. https://doi.org/10.1037/a0023341

Lewis, M. H., & Bodfish, J. W. (1998). Repetitive behavior disorders in autism. *Mental Retardation and Developmental Disabilities Research Reviews, 4*(2), 80–89. https://doi.org/10.1002/(sici)1098-2779(1998)4:2<80::Aid-mrdd4>3.0.Co;2-0

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., Cochran, M., & Huber, S. (2005). Effects of otitis media on a child with autism receiving behavioral intervention for self-injury. *Child & Family Behavior Therapy, 27*, 51–56. https://doi.org/10.1300/J019v27n02_05

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. https://doi.org/10.1016/j.ridd.2013.01.010

Lydon, S., Healy, O., O'Reilly, M. F., & Lang, R. (2012). Variations in functional analysis methodology: A systematic review. *Journal of Developmental and Physical Disabilities, 24*(3), 301–326. https://doi.org/10.1007/s10882-012-9267-3

Matson, J. L., Baglio, C. S., Smiroldo, B. B., Hamilton, M., Packlowskyj, T., Williams, D., & Kirkpatrick-Sanchez, S. (1996). Characteristics of autism as assessed by the diagnostic assessment for the severely handicapped-II (DASH-II). *Research in Developmental Disabilities, 17*(2), 135–143. https://doi.org/10.1016/0891-4222(95)00044-5

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., Bamburg, J. W., Mayville, E. A., Pinkston, J., Bielecki, J., Kuhn, D., … Logan, J. R. (2000). Psychopharmacology and mental retardation: A 10 year review (1990–1999). *Research in Developmental Disabilities, 21*(4), 263–296. https://doi.org/10.1016/S0891-4222(00)00042-1

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. https://doi.org/10.1016/j.ridd.2008.08.007

Matson, J. L., & Rivet, T. T. (2008). Characteristics of challenging behaviours in adults with autistic disorder, PDD-NOS, and intellectual disability. *Journal of Intellectual and Developmental Disability, 33*(4), 323–329. https://doi.org/10.1080/13668250802492600

Matson, J. L., & Wilkins, J. (2008). Antipsychotic drugs for aggression in intellectual disability. *The Lancet, 371*(9606), 9–10. https://doi.org/10.1016/S0140-6736(08)60046-X

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

Maurice, P., & Trudel, G. (1982). Self-injurious behavior prevalence and relationships to environmental events. *Monographs of the American Association on Mental Deficiency, 5*, 81–103.

McBrien, J. (2003). Assessment and diagnosis of depression in people with intellectual disability. *Journal of Intellectual Disability Research, 47*(1), 1–13.

McCarthy, 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. https://doi.org/10.1016/j.ridd.2009.10.009

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. https://doi.org/10.1046/j.1365-2788.2003.00517.x

Medeiros, K., Curby, T. W., Bernstein, A., Rojahn, J., & Schroeder, S. R. (2013). The progression of severe behavior disorder in young children with intellectual and developmental disabilities. *Research in Developmental Disabilities, 34*(11), 3639–3647. https://doi.org/10.1016/j.ridd.2013.08.002

Meins, W. (1995). Symptoms of major depression in mentally retarded adults. *Journal of Intellectual Disability Research, 39*(1), 41–45. https://doi.org/10.1111/j.1365-2788.1995.tb00912.x

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. https://doi.org/10.1016/j.ridd.2016.03.007

Mitchell, G., & Hastings, R. P. (2001). Coping, burnout, and emotion in staff working in community services for people with challenging behaviors. *American Journal on Mental Retardation, 106*(5), 448–459. https://doi.org/10.1352/0895-8017(2001)106<0448:Cbaeis>2.0.Co;2

Moss, S., Emerson, E., Kiernan, C., Turner, S., Hatton, C., & Alborz, A. (2000). Psychiatric symptoms in adults with learning disability and challenging behaviour. *The British Journal of Psychiatry, 177*(5), 452–456.

Mulick, J. A., Dura, J. R., Rasnake, K., & Callahan, C. (1986). *Prevalence of SIB in institutionalized nonambulatory profoundly retarded people*. Paper presented at the annual meeting of the American Psychological Association, Washington, DC.

Murphy, G. (2009). Challenging behavior: A barrier to inclusion? *Journal of Policy and Practice in Intellectual Disabilities, 6*(2), 89–90. https://doi.org/10.1111/j.1741-1130.2009.00216.x

Myrbakk, E., & von Tetzchner, S. (2008). Psychiatric disorders and behavior problems in people with intellectual disability. *Research in Developmental Disabilities, 29*(4), 316–332. https://doi.org/10.1016/j.ridd.2007.06.002

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*(3), 168–181. https://doi.org/10.1177/10634266040120030401

O'Brien, G., & Bevan, R. (2011). Recent advances in behavioural phenotypes as they affect adults. *Advances in Mental Health and Intellectual Disabilities, 5*, 5–14. https://doi.org/10.1108/20441281111165553

O'Dwyer, C., McCallion, P., Burke, É., 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. https://doi.org/10.1016/j.ridd.2018.05.007

O'Reilly, M. F. (1995). Functional analysis and treatment of escape-maintained aggression correlated with sleep deprivation. *Journal of Applied Behavior Analysis, 28*(2), 225–226. https://doi.org/10.1901/jaba.1995.28-225

O'Reilly, M. F. (1997). Functional analysis of episodic self-injury correlated with recurrent otitis media. *Journal of Applied Behavior Analysis, 30*(1), 165–167. https://doi.org/10.1901/jaba.1997.30-165

O'Reilly, M. F., & Lancioni, G. (2000). Response covariation of escape-maintained aberrant behavior correlated with sleep deprivation. *Research in Developmental Disabilities, 21*(2), 125–136. https://doi.org/10.1016/S0891-4222(00)00029-9

Oliver, A. C., Pratt, L. A., & Normand, M. P. (2015). A survey of functional behavior assessment methods used by behavior analysts in practice. *Journal of Applied Behavior Analysis, 48*(4), 817–829. https://doi.org/10.1002/jaba.256

Oliver, C., Adams, D., Allen, D., Bull, L., Heald, M., Moss, J., ... & Woodcock, K. (2013). Causal models of clinically significant behaviors in Angelman, Cornelia de Lange, Prader–Willi and Smith–Magenis syndromes. In International Review of Research in Developmental Disabilities (Vol. 44, pp. 167–211). Academic Press. https://doi.org/10.1016/B978-0-12-401662-0.00006-3

Oliver, C., Murphy, G. H., & Corbett, J. A. (1987). Self-injurious behaviour in people with mental handicap: A total population study. *Journal of Mental Deficiency Research, 31*(Pt 2), 147–162.

Oliver, C., Oxener, G., Hearn, M., & Hall, S. (2001). Effects of social proximity on multiple aggressive behaviors. *Journal of Applied Behavior Analysis, 34*(1), 85–88. https://doi.org/10.1901/jaba.2001.34-85

Payne, L. D., Scott, T. M., & Conroy, M. (2007). A school-based examination of the efficacy of function-based intervention. *Behavioral Disorders, 32*(3), 158–174.

Petty, J. L., Bacarese-Hamilton, M., Davies, L. E., & Oliver, C. (2014). Correlates of self-injurious, aggressive and destructive behaviour in children under five who are at risk of developmental delay. *Research in Developmental Disabilities, 35*(1), 36–45. https://doi.org/10.1016/j.ridd.2013.10.019

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. https://doi.org/10.1044/1058-0360(2007/039)

Poppes, P., van der Putten, A. 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. https://doi.org/10.1016/j.ridd.2010.07.017

Powis, L., & Oliver, C. (2014). The prevalence of aggression in genetic syndromes: A review. *Research in Developmental Disabilities, 35*(5), 1051–1071. https://doi.org/10.1016/j.ridd.2014.01.033

Pruijssers, A. C., van Meijel, B., Maaskant, M., Nijssen, W., & van Achterberg, T. (2014). The relationship between challenging behaviour and anxiety in adults with intellectual disabilities: A literature review. *Journal of Intellectual Disability Research, 58*(2), 162–171. https://doi.org/10.1111/jir.12012

Rapp, J., & Lanovaz, M. (2016). Stereotypy. In N. Singh (Ed.), *Handbook of evidence-based practices in intellectual and developmental disabilities* (pp. 751–779). New York, NY: Springer. https://doi.org/10.1007/978-3-319-26583-4_28

Reiss, S., & Rojahn, J. (1993). Joint occurrence of depression and aggression in children and adults with mental retardation. *Journal of Intellectual Disability Research, 37*(3), 287–294. https://doi.org/10.1111/j.1365-2788.1993.tb01285.x

Richards, C., Moss, J., O'Farrell, L., Kaur, G., & Oliver, C. (2009). Social anxiety in Cornelia de Lange syndrome. *Journal of Autism and Developmental Disorders, 39*(8), 1155–1162.

Richards, C., Oliver, C., Nelson, L., & Moss, J. (2012). Self-injurious behaviour in individuals with autism spectrum disorder and intellectual disability. *Journal of Intellectual Disability Research, 56*(5), 476–489. https://doi.org/10.1111/j.1365-2788.2012.01537.x

Rojahn, J. (1984). Self-injurious behavior in institutionalized, severely/profoundly retarded adults—Prevalence data and staff agreement. *Journal of Behavioral Assessment, 6*(1), 13–27. https://doi.org/10.1007/BF01321457

Rojahn, J. (1986). Self-injurious and stereotypic behavior of noninstitutionalized mentally retarded people: Prevalence and classification. *American Journal of Mental Deficiency, 91*(3), 268–276.

Rojahn, J., Borthwick-Duffy, S. A., & Jacobson, J. W. (1993). The association between psychiatric diagnoses and severe behavior problems in mental retardation. *Annals of Clinical Psychiatry, 5*(3), 163–170. https://doi.org/10.3109/10401239309148980

Rojahn, J., & Esbensen, A. J. (2002). *Epidemiology of self-injurious behavior in mental retardation: A review*. Washington, DC: American Psychological Association.

Rojahn, J., Matson, J. L., Naglieri, J. A., & Mayville, E. (2004). Relationships between psychiatric conditions and behavior problems among adults with mental retardation. *American Journal on Mental Retardation, 109*(1), 21–33. https://doi.org/10.1352/0895-8017(2004)109<21:RBPCAB>2.0.CO;2

Ross Collins, M., & 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.

Roy, A., Roy, M., Deb, S., Unwin, G., & Roy, A. (2015). Are opioid antagonists effective in attenuating the core symptoms of autism spectrum conditions in children: A systematic review. *Journal of Intellectual Disability Research, 59*(4), 293–306. https://doi.org/10.1111/jir.12122

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, 45–46*, 307–315. https://doi.org/10.1016/j.ridd.2015.07.019

Rush, A., & Frances, A. E. (2000). Expert consensus guideline series: Treatment of psychiatric and behavioral problems in mental retardation. *American Journal on Mental Retardation, 105*(3), 159–226.

Sandman, C. A. (1990). The opiate hypothesis in autism and self-injury. *Journal of Child and Adolescent Psychopharmacology, 1*(3), 237–248. https://doi.org/10.1089/cap.1990.1.237

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. https://doi.org/10.3390/ph4020366

Sansom, D., Krishnan, V., Corbett, J., & Kerr, A. (1993). Emotional and behavioral aspects of Rett syndrome. *Developmental Medicine and Child Neurology, 35*(4), 340–345.

Schroeder, S. R., Schroeder, C. S., Smith, B., & Dalldorf, J. (1978). Prevalence of self-injurious behaviors in a large state facility for the retarded: A three-year follow-up study. *Journal of Autism and Childhood Schizophrenia, 8*(3), 261–269.

Schützwohl, M., Koch, A., Koslowski, N., Puschner, B., Voß, E., Salize, H., … 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. https://doi.org/10.1007/s00127-016-1197-4

Siever, L. J. (2008). Neurobiology of aggression and violence. *The American Journal of Psychiatry, 165*(4), 429–442. https://doi.org/10.1176/appi.ajp.2008.07111774

Sigafoos, J., Arthur, M., & O'Reilly, M. (2003). Challenging behavior and developmental disability. Baltimore: Paul H Brooks Publishing Co.

Skinner, B. F. (1938). *The behavior of organisms: An experimental analysis*. Oxford, UK: Appleton-Century.

Sloneem, J., Oliver, C., Udwin, O., & Woodcock, K. A. (2011). Prevalence, phenomenology, aetiology and predictors of challenging behaviour in Smith-Magenis syndrome. *Journal of Intellectual Disability Research, 55*(2), 138–151. https://doi.org/10.1111/j.1365-2788.2010.01371.x

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. *British Journal of Psychiatry, 191*(4), 313–319. https://doi.org/10.1192/bjp.bp.106.031104

Sovner, R., Fox, C. J., Lowry, M. J., & Lowry, M. A. (1993). Fluoxetine treatment of depression and associated self-injury in two adults with mental retardation. *Journal of Intellectual Disability Research, 37*(Pt 3), 301–311.

Stoiber, K. C., Purdy, S., & Klingbeil, D. A. (2016). Evidence-based practices. In N. N. Singh (Ed.), *Handbook of evidence-based practices in intellectual and developmental disabilities* (pp. 41–68). Cham, Switzerland: Springer International Publishing.

Sturmey, P., & Hamelin, J. P. (2014). Psychological treatments. In *Handbook of psychopathology in intellectual disability: Research, practice, and policy* (pp. 325–357). New York, NY: Springer Science + Business Media.

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. https://doi.org/10.1016/j.ridd.2010.04.018

Sturmey, P., & Williams, D. E. (2016). *Pica in individuals with developmental disabilities*. Cham, Switzerland: Springer International Publishing.

Swender, S., Matson, J., Mayville, S., González, M., & McDowell, D. (2006). A functional assessment of hand mouthing among persons with severe and profound intellectual disability. *Journal of Intellectual & Developmental Disability, 31*, 95–100. https://doi.org/10.1080/13668250600710880

Symons, F. J., Harper, V., Shinde, S. K., Clary, J., & Bodfish, J. W. (2010). Evaluating a sham-controlled sensory-testing protocol for nonverbal adults with neurodevelopmental disorders: Self-injury and gender effects. *Journal of Pain, 11*(8), 773–781. https://doi.org/10.1016/j.jpain.2009.11.011

Tate, B. G., & Baroff, G. S. (1966). Aversive control of self-injurious behavior in a psychotic boy. *Behaviour Research and Therapy, 4*(1), 281–287. https://doi.org/10.1016/0005-7967(66)90084-2

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

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

Tsiouris, J. A., Mann, R., Patti, P. J., & Sturmey, P. (2003). Challenging behaviors should not be considered as depressive equivalents in individuals with intellectual disability. *Journal of Intellectual Disability Research, 47*(1), 14–21. https://doi.org/10.1046/j.1365-2788.2003.00456.x

Tunnicliffe, P., & Oliver, C. (2011). Phenotype-environment interactions in genetic syndromes associated with severe or profound intellectual disability. *Research in Developmental Disabilities, 32*(2), 404–418. https://doi.org/10.1016/j.ridd.2010.12.008

Turner, C., & Lewis, M. (2002). Dopaminergic mechanisms in self-injurious behavior and related disorders. In *Self-injurious behavior: Gene-brain-behavior relationships* (pp. 165–179). Washington, DC: American Psychological Association. https://doi.org/10.1037/10457-011

Turner, M. (1999). Annotation: Repetitive behaviour in autism: A review of psychological research. *Journal of Child Psychology and Psychiatry and Allied Disciplines, 40*(6), 839–849.

Turygin, N., Matson, J., MacMillan, K., & Konst, M. (2013). The relationship between challenging behavior and symptoms of depression in intellectually disabled adults with and without autism spectrum disorders. *Journal of Developmental and Physical Disabilities, 25*(4), 475–484. https://doi.org/10.1007/s10882-012-9321-1

Tyrer, F., McGrother, C. W., Thorp, C. F., Donaldson, M., Bhaumik, S., Watson, J. M., & Hollin, C. (2006). Physical aggression towards others in adults with learning disabilities: Prevalence and associated factors. *Journal of Intellectual Disability Research, 50*(Pt 4), 295–304. https://doi.org/10.1111/j.1365-2788.2005.00774.x

Ulman, J. D., & Sulzer-Azaroff, B. (1975). Multielement baseline design in educational research. In E. Ramp & G. Semb (Eds.), *Behavior analysis: Areas of research and application* (pp. 377–391). Englewood Cliffs, NJ: Prentice-Hall.

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. https://doi.org/10.1352/0895-8017(2005)110<164:Mabsap>2.0.Co;2

Van den Bogaard, K. J. H. M., Nijman, H. L. I., Palmstierna, T., & Embregts, P. J. C. M. (2018). Characteristics of aggressive behavior in people with mild to borderline intellectual disability and co-occurring psychopathology. *Journal of Mental Health Research in Intellectual Disabilities, 11*, 124–142. https://doi.org/10.1080/19315864.2017.1408726

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. https://doi.org/10.1016/0891-4222(92)90013-V

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: The Guilford Press.

Whitaker, S., & Read, S. (2006). The prevalence of psychiatric disorders among people with intellectual disabilities: An analysis of the literature. *Journal of Applied Research in Intellectual Disabilities, 19*(4), 330–345. https://doi.org/10.1111/j.1468-3148.2006.00293.x

Wigren, M., & Heimann, M. (2001). Excessive picking in Prader-Willi syndrome: A pilot study of phenomenological aspects and comorbid symptoms. *International Journal of Disability, Development and Education, 48*(2), 129–142. https://doi.org/10.1080/10349120120053621

Zijlmans, L. J. M., Embregts, P. J. C. M., Bosman, A. M. T., & Willems, A. P. A. M. (2012). The relationship among attributions, emotions, and interpersonal styles of staff working with clients with intellectual disabilities and challenging behavior. *Research in Developmental Disabilities, 33*(5), 1484–1494. https://doi.org/10.1016/j.ridd.2012.03.022

Zlotnick, I. C., Mattia, I. J., & Zimmerman, I. M. (1999). Clinical correlates of self-mutilation in a sample of general psychiatric patients. *The Journal of Nervous & Mental Disease, 187*(5), 296–301. https://doi.org/10.1097/00005053-199905000-00005

4 Genetic Disorders and Dual Diagnosis: Building Clinical Management on Etiology and Neurocognition

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Introduction

Over the past two decades, it has become evident that genetic disorders, particularly when accompanied by intellectual disabilities, can manifest firstly as psychiatric symptoms and/or behavioral disorders. It was around the 1960s, when the term "dual diagnosis" (DD) was coined for conditions where intellectual disabilities co-occurred with behavioral and/or mental health problems. One of the best known examples is the 22q11.2 microdeletion syndrome that is highly associated with psychiatric diseases within the schizophrenic, bipolar, or autistic spectrum. However, for numerous genetic syndromes, specifically the relatively new ones, studies have mainly focused on somatic complications and to a lower extent on behavioral characteristics while still only occasionally addressing intellectual (dis)abilities, cognition, emotion, and psychopathology. Moreover, most neuropsychiatric studies in this field are limited to children only, and – if cognitive functioning is evaluated at all – reports are often limited to single-case studies with only one or a few instruments: mostly questionnaires filled in by parents or teachers. Systematic, controlled, and multimethod assessment of cognitive and

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social-emotional functioning is rarely performed.

In research on the genetic underpinnings of neuropsychiatric disorders, three major problems are typically encountered. First, there is the large degree of genetic heterogeneity, which means that different genes can contribute in a varying way to the emergence of a single disorder. A second difficulty is the polygenetic inheritance, that is, the simultaneous presence of multiple genetic vulnerabilities that may be responsible for the development of a particular syndrome. And a third problem lies in the well-known interaction between environmental and genetic factors during development from early conception on. The fact that neuropsychiatric disorders are hard to understand from genetic anomalies only drives us to bridge the broad gap between fundamental research (e.g., at the detailed biological level), and applied clinical research (e.g., on diagnosis and treatment efficacy). In order to do so, and overcome the diagnostic artifacts of DSM-classified psychiatric syndromes, we should study neuropsychological phenotypes, that is, profiles of cognitive functions that emerge and can be regarded as an intermediate level between the genetic makeup and the syndrome-related neuropsychiatric disorders.

Neuropsychological assessment provides insights into current and potential functioning of individual patients as well as (personal or contextual) modifiable factors. Cognitive phenotypes can differentiate diagnostic human groups but also connect the basic and clinical sciences by studying them in the wider area of more, different species such as research on the cognitive phenotypes of, e.g., rodents and fish. In addition, recurrent neurocognitive assessment may be useful to monitor changes over time across the life span. The ultimate goal of neuropsychological (cognitive and behavioral) research is to learn about the contribution of both genetic and contextual factors in the variation of syndrome phenotypes. It can shed light on the mechanisms that link the human genome to complex psychological syndromes and psychiatric disorders, and in that way, it can help to develop novel treatments for neuropsychiatric disorders.

In this chapter, we focus mainly on psychopathological and neuropsychological phenotyping of genetic syndromes. Before doing so, we first give an overview of the most important genetic diagnostic achievements during the past decades and its interplay with clinical practice in order to understand the evolving neuropsychological studies of genetic disorders. We conclude this chapter with suggestions concerning strategies for further research and clinical practice.

Genetic Disorders Among Persons with ID: From Karyotyping to Whole Exome Sequencing

Clinical genetic diagnostics

Until the end of the past millennium, genetic diagnostics of patients with neurodevelopmental disorders was performed mainly by means of karyotyping, *fluorescence* in situ *hybridization* (FISH), *sanger sequencing,* and *microarray.* Karyotyping refers to a method that measures the number and appearance of the chromosomes. Based on this method deviations in the number of chromosomes are detected, and specific genetic syndromes such as Down syndrome (Trisomy 21), Edwards syndrome (Trisomy 13), Turner syndrome (monosomy X), and Klinefelter syndrome (47,XXY) have been identified. The etiology of syndromes such as Cri-du-chat-syndrome (5p) and fragile X syndrome has been identified based on aberrations in parts of chromosomes. S*anger sequencing* refers to a method that detects DNA aberrations, which enables clinical geneticists to test for genetic syndromes that are caused by aberrations in single genes.

Since the last decade, a new genetic test, *whole exome sequencing*, enables unbiased testing for DNA aberrations in all genes simultaneously. Due to this genetic technique, many new monogenetic syndromes (syndromes caused by a mutation in a single gene) have been identified, e.g., KBG syndrome (*ANKRD11*), Kleefstra syndrome (*EHMT1)*, and Kabuki syndrome (*KMT2D*). Consequently, this technique has led to an increase in the number of identified causes of neurodevelopmental disorders.

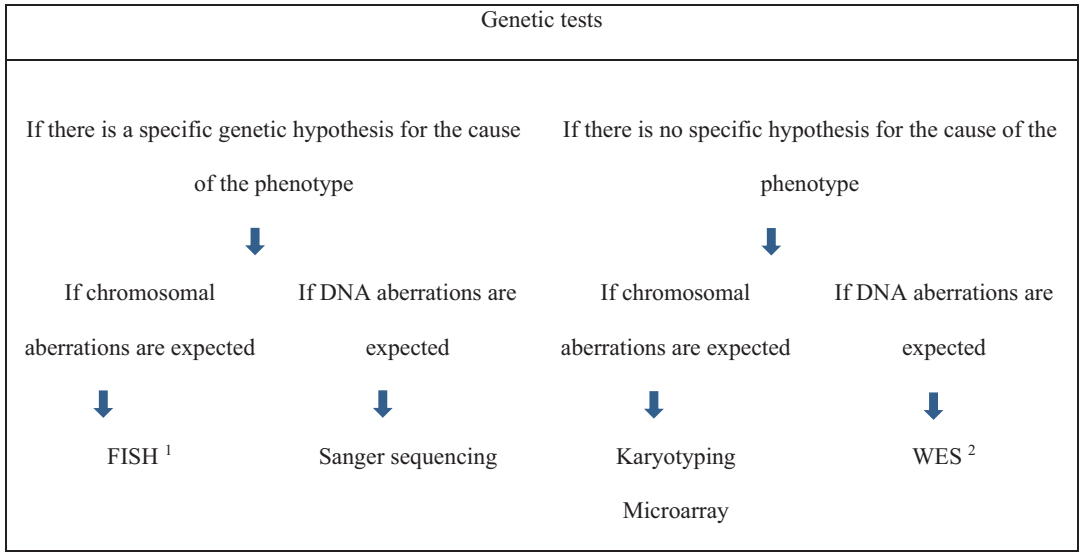

Fig. 4.1 Overview of the diagnostic genetic process. [1]Fluorescence-in-situ-hybridization; [2]Whole exome sequencing

The choice for the type of diagnostic test firstly depends on the presence or absence of a specific hypothesis derived from the phenotype. Secondly, this decision depends on the expected size of the defect (either on chromosomal level or DNA level). For an overview of this diagnostic genetic process, see Fig. 4.1. Phenotype-first approach refers to a diagnostic process in which the clinician tests for a specific genetic abnormality based on expected syndrome that fits the phenotypical (medical and behavioral) presentation of the patient. *Whole exome sequencing* enables an alternative diagnostic approach in which there is no single hypothesis based on the clinical phenotype, and multiple genes have to be assessed in parallel (genotype-first approach).

The rapid increase of identified genetic syndromes precipitated multiple initiatives to document all these pathogenic gene variations and their corresponding phenotypes. One of the most frequently used is Online Mendelian Inheritance in Man (OMIM), a web-based database that contains information on all known genetic disorders and was originally initiated in the early 1960s ("OMIM® – Online Mendelian Inheritance in Man", 2019). Although newly discovered genetic syndromes are often primarily described in terms of facial dysmorphisms and other medical characteristics, neuropsychological profiling of individuals with a suspected genetic disorder may contribute to the genetic diagnostic process.

Research in patient groups

Clinical research on aforementioned monogenetic disorders focuses on describing the (neuropsychological) phenotypes of these patient groups. In contrast, research on specific psychiatric disorders (e.g., ADHD, schizophrenia) that are caused by an interaction of multiple genetic and environmental factors focuses on identification of the potential multiple contributing genes. Studies that apply this latter approach are referred to as *genome-wide association studies* (GWAS). Results of these studies only concern patient groups and are not transferrable to individual patients. However, the remaining paragraphs of this chapter are solely focused on the description of the behavioral phenotype of monogenetic syndromes.

Some Examples of Psychopathological and Neuropsychological Phenotyping of (Rare) Genetic Syndromes

Fragile X Syndrome

The Fragile X syndrome (FXS; OMIM: 300624) is an X-linked disorder with an incidence of about 1 in 4000 newborn males. It was first described in 1969 (Lubs, 1969) and is caused by anomalies in gene *FMR1* on chromosome Xq27.3. This leads to a shortage or complete loss of the FMR1 protein which is essential for dendrite formation, synapse formation, and experiential learning (Hernandez et al., 2009; Marco & Skuse, 2006). The severity of intellectual disability and the intensity of related behavior problems are known to be proportional to the number of repeats. As to the central nervous system, various structural abnormalities have been demonstrated in patients with FXS, in particular the enlargement of hippocampus, amygdala, caudatus, and thalamus with a reduction of the cerebellar vermis (Hessl, Rivera, & Reiss, 2004). Epileptic phenomena are frequent.

Affected males show distinctive dysmorphic features such as a long face, and prominent ears, and a variable degree of developmental delay and behavior problems. Further, female carriers present with or without impaired levels of intelligence.

Weaknesses in levels of general intellectual functioning are accompanied by specific impairments in cognitive domains as attention, (working) memory, mathematical skills, executive functioning, and social cognition.

The psychopathological phenotype includes various autism-related symptoms such as social anxiety and withdrawal behavior, stereotypies like flapping or biting of the hands, perseverations, extreme sensitivity to environmental stimuli, and, in general, decreased social reciprocity with an avoidance of eye contact (Hagerman, Ono, & Hagerman, 2005). Furthermore, multiform anxiety symptoms, obsessive-compulsive characteristics, hyperactivity/impulsivity, and aggression have been described. Female carriers with normal levels of intelligence have an increased risk of mood and anxiety disorders and a schizotypal personality disorder (Franke et al., 1999).

Multiple studies have reported initiatives for specific interventions that target cognitive functioning and educational learning in FXS (Raspa, Wheeler, & Riley, 2017). For example, computer-based cognitive methods in which specific skills (math, geography) or cognitive functions (working memory) are used. These computerized tools are generally highly motivating and reduce social pressure, which may affect learning in children whose anxiety is increased with social interactions. Further, learning strategies in FXS that incorporate visually based, experiential, or holistic learning were reported to be the most successful.

Noonan Syndrome

Noonan syndrome (NS; OMIM: 163950) is a genetically heterogeneous disorder with different causative mutations in the RAS-MAPK pathway.[1] The estimated incidence of NS is between 1:1000 and 1:2500 live births (Allanson et al., 2010). NS may occur on a sporadic basis or in a pattern consistent with autosomal dominant inheritance, with a predominance of maternal transmission. In approximately 50% of the patients, a missense mutation is found in the *PTPN11* gene on chromosome 12 (12q24.1). Germline mutations in 12 other genes of the Ras-MAPK pathway have been identified as causative in NS and closely related disorders.[2]

[1] Syndromes with shared pathogenetic mechanisms and clinical overlap with NS include cardiofaciocutaneous (CFC) syndrome, Costello syndrome, neurofibromatosis type 1 (NF1), Noonan syndrome with multiple lentigines (formerly called LEOPARD syndrome), Noonan syndrome-like disorder with loose anagen hair (NSLH), and CBL-associated syndrome. They are grouped into the neurocardiofacialcutaneous syndrome family, or the Ras-opathies.

[2] *SOS1*, *RAF1, KRAS, NRAS, BRAF*, *SHOC2, MAP2K1 (MEK1)*, *MAP2K2, CBL*, *RIT1*, *A2ML1, SPRED1,* and *HRAS*.

The following characteristics have been described: typical facial features (hypertelorism, low set ears, ptosis, strabismus), congenital heart defects, broad and/or webbed neck, small stature, skeletal malformations, cryptorchidism, lymphatic dysplasia, and abnormal bleeding (Verhoeven, Wingbermühle, Egger, Van der Burgt, & Tuinier, 2008). Also, hearing loss due to recurrent otitis media, feeding problems, and ophthalmic abnormalities are reported frequently. The syndrome was first described in the 1970s, and diagnosis was primarily made on a set of clinical criteria (Noonan, 1968; Van der Burgt et al., 1994). The most widely used scoring system was developed in 1994 and updated and validated in 2010 (Dyscerne, 2010; Van der Burgt et al., 1994). There are only a limited number of case studies on structural brain abnormalities in NS, in which macrocephaly, hydrocephaly, epilepsy and hemangioma, and, in a few individuals, also structural defects of the cerebellum and vascular changes are described (Wingbermühle, Egger, van der Burgt, & Verhoeven, 2009).

In people with NS, a variable level of intelligence has been found, varying from mentally retarded to superior, with a mildly lowered average level (Verhoeven et al., 2008; Wingbermühle et al., 2009). Language and motor development are often delayed in childhood, but no longer constrained in adulthood. Furthermore, in adulthood prolonged mild problems have been found in selective and sustained attention, as well as suboptimal organization skills and compromised abilities to structure complex information. Memory function (storage and retrieval of information) in adults is not affected (Wingbermühle et al., 2009). Compared to healthy adults, those with NS show lowered information processing speed (Wingbermuhle et al., 2012).

NS in adults has been associated with (moderate) deficiencies in social and emotional recognition and expression (Verhoeven et al., 2008). Adults with NS show higher levels of cognitive alexithymia compared to healthy adults. This means that they face more difficulties in identifying, analyzing, and verbalizing their own emotions. Mild signs of anxiety and lowered mood have been found in NS (Verhoeven et al., 2008). The increased risk to develop affective disorders (like mood, anxiety, and compulsive disorders) may be associated with the frequent co-occurrence of alexithymia in adulthood (Wingbermühle et al., 2009) as well as frequently being bullied, for example, because of dysmorphia (Wingbermühle, Egger, Verhoeven, Van der Burgt, & Kessels, 2012).

Recent research has revealed that interventions in improving social cognition in adult neuropsychiatric patients are effective. In case of alexithymia in patients with NS, it is important to help patients to give words to their inner experiences and improve their social cognition (e.g., emotion recognition) by dedicated interventions (Roelofs, Wingbermühle, Kessels, & Egger, 2019).

Kleefstra Syndrome

The autosomal dominant disorder Kleefstra syndrome (KS; OMIM #610253) is caused by a microdeletion at 9q34.3 or pathogenic variants in *EHMT1* (OMIM #607001). Although the current prevalence is unknown, based on data from other rare disorders involving intellectual disability, KS is estimated to affect at least 1:200,000 individuals. The syndrome is characterized by developmental delay/intellectual disability, childhood hypotonia, microcephaly, and distinctive facial dysmorphisms (Kleefstra, Nillesen, & Yntema, 2010). KS is also associated with heart defects (50% of patients), renal anomalies (10–20%), urologic impairments, genital anomalies in males (30% of males), severe respiratory infections, and obesity. Multiple studies describe both structural and functional anomalies of the brain in KS. Epileptic seizures have been described in 30% of the individuals, which compromises tonic-clonic seizures, absence seizures, and complex partial epilepsy (Kleefstra et al., 2010). As for brain imaging studies, anomalies have been found in half of the patients with KS. These anomalies comprise mostly white matter abnormalities and cortical atrophy (frontal, cerebral, or cerebellar) or indirect evidence of such atrophy (prominent ventricles, increased cerebrospinal

fluid spaces) (Ciaccio et al., 2018). A disturbed sleep pattern has been described and includes frequent and enduring nocturnal awakenings accompanied by daytime sleepiness (Verhoeven, Kleefstra, & Egger, 2010).

Developmental delay is present for both motor and language functioning, whereby the receptive language is superior to the weak or even absence expressive language. The level of intellectual functioning in KS ranges from moderate to severe ID, with only a minority of reported individuals displaying a mild ID (Kleefstra et al., 2010). The behavior phenotype in children comprises attention problems, autism spectrum disorders, stereotypies, and self-injurious behavior (Vermeulen et al., 2015). A significant number of patients display behavior difficulties such as aggression, impulsivity, and depressive and chaotic behaviors. During adolescence, a gradual decline of previously learned motor, language, and communication skills becomes prominent. Furthermore, regression is observed in terms of goal-directed behavior (performing daily skills) and goal-directed cognition (interest and curiosity) as well as in an almost complete lack of emotional and social reciprocity (Verhoeven et al., 2010; Verhoeven, Egger, Vermeulen, van de Warrenburg, & Kleefstra, 2011; Vermeulen et al., 2017). The decline in functioning is expected to be an expression of a psychotic episode followed by sleep disturbances and for that reason needs to be treated with antipsychotics in order to restore sleep and halt further regression (Vermeulen et al., 2017; Vermeulen, de Boer, et al., 2017). Therefore, it is recommended to periodically asses the level of adaptive functioning and in relation to maladaptive functioning in all patients in order to both detect regression and overcome under- or overstimulation in counseling en school functioning.

KBG Syndrome

KBG syndrome (OMIM #148050) is named after the surname initials of the first three families described in 1975 (Herrmann, Pallister, Tiddy, & Opitz, 1975). Today, approximately 218 patients have been reported worldwide. As the clinical phenotype is rather mild, the autosomal dominant disorder was hardly recognized until the molecular identified cause in 2011 (heterozygous mutation in *ANKRD11* (OMIM #611192) (Sirmaci et al., 2011). A tremendous increase in molecularly confirmed cases has been achieved since then. *ANKRD11* was even identified as the most frequently mutated gene in a cohort of 400 patients with neurodevelopmental disorders, establishing KBG syndrome as one of the most prevalent dominant neurodevelopmental syndromes. Although clinical features may vary, KBG syndrome is characterized by developmental delay/ID, macrodontia of upper central permanent incisors, triangular facies, brachycephaly, hypertelorism, protruding ears, and upturned nose with full nasal tip. Other recognized features include short stature, palate abnormalities, skeletal anomalies, cardiac abnormalities, partial hearing loss, and persistent/large fontanel.

In a study of mice and human cortical, neural precursors is indicated that *ANKRD11* is a chromatin regulator that controls histone acetylation and gene expression during neural development (Gallagher et al., 2015). Using neuroimaging techniques, various structural abnormalities of the central nervous system in patients with KBG syndrome can be demonstrated, in particular small cerebelli, hypoplasia of the cerebellar vermis (Goldenberg et al., 2016; Kim, Cho, Park, Im, & Kim, 2015; Tunovic, Barkovich, Sherr, & Slavotinek, 2014), enlargement of the ventricles, partial agenesis of the corpus callosum, and white matter anomalies around the ventricles and near the gray-white junction in both hemispheres (Low et al., 2016; Miyatake et al., 2013; Skjei, Martin, & Slavotinek, 2007). As to neurological findings, abnormal electroencephalogram patterns both with and without tonic-clonic, complex partial, and absence seizures are described (Kim et al., 2015; Ockeloen et al., 2015; Skjei et al., 2007; Walz et al., 2015).

Psychopathology that has been described in patients with KBG syndrome include attention deficit hyperactivity disorder (ADHD), autism spectrum disorder (ASD), anxiety, as well as aggression or compulsive behavior (Goldenberg et al., 2016; Hah et al., 2009; Low et al., 2016;

Murray et al., 2017; Novara et al., 2017; Ockeloen et al., 2015; Skjei et al., 2007). Systematic investigation of the behavioral phenotype pointed, however, to a more specific behavioral profile that primarily includes distractibility, impulsivity, and restless behavior. Furthermore, though more social problems were indeed found compared to normative means, patients displayed fewer social difficulties when compared to a more appropriate control group with a similar level of intellectual functioning (van Dongen et al., 2019). As for cognitive functioning, in the group of patients with KBG syndrome, a wide range in the level of intellectual functioning is present, varying from a moderate intellectual disability to average levels of intelligence, with a majority of patients demonstrating a mild intellectual disability. Compared to a mixed control group of patients with other genetic syndromes, patients with KBG syndrome displayed weaknesses in sustained attention, shifting and visuoconstruction, and relative strengths in memory and social cognitive functioning, where the inhibition problems may also underlie their weaker visuoconstruction performance (van Dongen et al., 2019). These findings on social (cognitive) functioning argue against the previously described ASD symptomatology in patients with KBG syndrome; reported social difficulties in patients with KBG syndrome may well be related to their ID rather than a result of a ASD. Further, found weaknesses in attention in executive functioning highlight the importance of providing external structure (including tasks monitoring and controlling of processing speed) in educational trajectories of patients with KBG syndrome in order to maximize their learning and development.

Phelan-McDermid Syndrome

Phelan-McDermid syndrome (PMS; OMIM: #606232) is a microdeletion syndrome resulting from loss of the terminal end of chromosome 22 by a simple deletion, an unbalanced translocation, ring chromosome formation, or other unbalanced structural changes typically comprising the SH3 and multiple ankyrin repeat domains 3 (SHANK3) gene located in 22q13.33 (Luciani et al., 2003; Mitz et al., 2018; Phelan, 2008). PMS can also be caused by a pathogenic nucleotide variant in this critical gene (De Rubeis et al., 2018; Kolevzon et al., 2014; Wilson et al., 2003). The phenotype was first described in the late 1980s by Phelan and co-workers (Phelan, Rogers, & Stevenson, 1988). Nowadays, the phenotypic presentation of PMS is characterized by neonatal hypotonia, recurrent upper airway infections, global developmental delay, impaired to absent speech and expressive language, increased sensitivity to sensory stimuli and sudden environmental events, as well as symptoms from the autism spectrum in the absence of major dysmorphisms (Cusmano-Ozog, Manning, & Hoyme, 2007; Phelan, 2008; Phelan & McDermid, 2011; Philippe et al., 2008). In addition, congenital cardiac and urogenital anomalies may be present as well as hypothyroidism, lymphedema, decreased perspiration and a high pain threshold (Costales & Kolevzon, 2015; Kolevzon, Angarita, et al., 2014; Sarasua et al., 2014). Moreover, a variety of epileptic manifestations may be present in about one third of patients with PMS (Figura et al., 2014; Holder & Quach, 2016) as well as structural cerebral abnormalities such as cerebellar vermis hypoplasia, thinning of the corpus callosum, and ventricular dilatation (Aldinger et al., 2013; Philippe et al., 2008; Srivastava et al., 2019).

With respect to the course and the prognosis of the syndrome, several authors mentioned SHANK3-related neurobehavioral regression, i.e., loss of acquired skills either permanently or for an extended period and often with co-occurring catatonic phenomena, which may be triggered by stressful events, seizures, or infection (Breckpot et al., 2016; De Rubeis et al., 2018; Denayer et al., 2012; Serret et al., 2015). Most important, however, are the recurrent reports in the literature about psychopathology from the autism spectrum (Oberman, Boccuto, Cascio, Sarasua, & Kaufmann, 2015; Tabet et al., 2017; Uchino & Waga, 2015) and the bipolar affective domain (Denayer et al., 2012; Egger, Verhoeven, Groenendijk-Reijenga, & Kant,

2017; Egger, Zwanenburg, van Ravenswaaij-Arts, Kleefstra, & Verhoeven, 2016; Guilmatre, Huguet, Delorme, & Bourgeron, 2014; Verhoeven, Egger, Cohen-Snuijf, Kant, & de Leeuw, 2013; Verhoeven, Egger, Willemsen, de Leijer, & Kleefstra, 2012). As for therapeutic interventions, short-term studies (with a limited number of children with PMS) indicate that injections with insulin-like growth factor 1 or intranasal administration of insulin may have some beneficial effects on aspects of social functioning (Kolevzon et al., 2014; Schmidt, Kern, Giese, Hallschmid, & Enders, 2009; Zwanenburg et al., 2016).

Prader-Willi Syndrome

Genetic Information

Prader-Willi syndrome (PWS; OMIM: 176270) or Prader-Labhart-Willi syndrome is first described in 1956 by the Swiss medical doctors Prader, Labhart, and Willi. This genetic neurocognitive developmental disorder results from a lack of expression of paternally derived genes from the 15q11-13 region of the paternally inherited chromosome 15. The absence of expression of the genes *SNORD116*, *MAGEL2*, and *IPW* at locus 15q11-13, singularly or in combination, are considered central to PWS.

The following three mechanisms account for most cases of PWS: First, a deletion (DEL subtype) of the 15q11-13 region from the paternal chromosome (~55–70% of cases). The DEL subtype is divided according to deletion size into two different subtypes (see Table 4.1). Second, a maternal uniparental disomy (UPD subtype), where both chromosomes have genetic material from the mother (~25–40%). Third, a small minority of cases (<5%) are caused by imprinting center defects (IC subtype) or translocations. Both the UPD subtype and IC subtype are sometimes also referred to as "disomy group."

The birth incidence rate of PWS is about 1:25,000. The estimated population prevalence of PWS varies from between 1 in 10,000 and 1 in 50,000 (Bennett, Germani, Haqq, & Zwaigenbaum, 2015; Whittington & Holland, 2018). There is a relatively high mortality rate compared to both the general population and to other groups of people with intellectual disabilities. Today, the average age is approximately 40–50 years (Butler et al., 2018; Sinnema, Schrander-Stumpel, Maaskant, Boer, & Curfs, 2012).

Table 4.1 PWS subtypes

Group	Subtype	Genetic cause	Percentage of patients with PWS with this cause
–	DEL: Type I Type II	Deletion of the 15q11-13 region from the paternal chromosome: deletion between breakpoint 1 and 3 deletion between breakpoint 2 and 3	~55–70%
Disomy group	UPD	Maternal uniparental disomy, both chromosomes have genetic material from the mother	~25–40%
	IC	Imprinting center defects	<5%
	–	Translocations	

Medical Conditions

The PWS phenotype includes facial dysmorphic features (narrow forehead, almond-shaped eyes, and triangular mouth, thin and down-turned upper lip), short stature, small hands and feet, hypothalamic dysfunction, and impaired sexual development, due to growth and sex hormone deficiency. Other physical features are eye problems (especially squint) and scoliosis, which are sometimes present from birth or may emerge later.

The hypothalamic dysfunction is responsible for hormonal dysfunctions, causing deficiencies of thyroid hormone, cortisol, sex hormones testosterone/estrogen, and IGF-1 ("growth hormone"). Furthermore, the hypothalamic dysfunction causes also dysregulation of appetite due to absence of the sense of satiety, dysregulation of body temperature (inability to produce fever), disturbed pain perception, central apneas,

sleep abnormalities (including excessive daytime sleepiness), and behavioral problems.

After birth people with PWS are characterized by failure to thrive, extreme hypotonia (low muscle tone) and associated sucking deficit, and feeding difficulties. Besides hypotonia and lower motor skills, in early childhood (from 3 to 4 years and older; usually between 2 and 6 years of age) children with PWS start suffering from hyperphagia, due to insatiable hunger, preoccupation by food, and/or food-seeking behavior. Therefore, people with PWS are highly at risk for marked obesity and diabetes, partly because their bodies need less calories.

As for brain structure and functioning, aberrations in both subcortical and higher-order structures PWS have been reported, including those involved in processing reward, motivation, affect, and higher-order cognitive functions (Manning & Holland, 2015). More specific, disturbances have been found in the limbic cortico-striatal-pallido-thalamic loops,[3] which are related to emotional cognition and regulation, and the dorsolateral prefrontal network,[4] which has been strongly associated with executive functions (including set shifting). A preliminary trial of three individuals with PWS indicates some improvements in maladaptive behavior, temperament, social functioning, and food-seeking behavior due to vagus nerve stimulation (Manning et al., 2016).

Cognitive Functioning

Intelligence

Generally, individuals with PWS have an intellectual disability (ID) with average full size IQ of 60–70. The level of ID is mild or moderate in most people with PWS (Bennett et al., 2015). The IQ distribution is, in substance, a normal curve, but the distribution is shifted with about 40 points to the lower end (Whittington & Holland, 2010). Some persons with PWS have IQ scores that are within what is considered the expected range for the general population, but nevertheless nearly always face learning issues. Note that cognitive strengths and weaknesses vary in severity and from person to person.

Several studies have been done to investigate difference in general levels of abilities or more specific intellectual profiles between genetic PWS subtypes. For example, comparison of Wechsler subtests or indices has indicated higher information processing speed (subtest "Coding") and higher performance IQ in the DEL subtype compared to the UPD subtype. In the UPD subtype, higher performance on verbal comprehension (subtest "Vocabulary") and higher verbal IQ has been observed, compared to the DEL subtype (Copet et al., 2010; Whittington & Holland, 2010) (see Table 4.2. for a PWS subtype comparison).

Cognitive Domains

With respect to more specific domains of cognitive functioning, difficulties have been found in attention and short-term memory/working memory (Jauregi et al., 2007). Also, auditory processing turned out to be worse than visual processing (Curfs, Wiegers, Sommers, Borghgraef, & Fryns, 1991). Furthermore, weaknesses are found in executive functioning (e.g., cognitive flexibility) compared both to the general population and to an IQ and age matched control group of patients with fragile X. Relative strengths are the long-term memory capacities as well as the ability to solitary practice tasks they love (e.g., puzzles) and develop visually oriented strategies in solving them (e.g., using the shape of the pieces rather than the color or visual depiction) (Whittington & Holland, 2017).

Social cognition is, in line with the level of general abilities, usually impaired in individuals

Table 4.2 PWS subtype comparison intelligence

PWS subtype comparison	UPD	DEL
Processing speed	Lower	Higher
Performance IQ	Lower	Higher
Verbal comprehension	Higher	Lower
Verbal IQ	Higher	Lower

[3]The limbic cortico-striatal-pallido-thalamic loops involve the orbitomedial prefrontal cortex, anterior cingulate cortex, ventral striatum, ventral pallidum, and thalamus.

[4]The dorsolateral prefrontal network has connections with premotor and parietal regions and projects to caudate head, pallidum, and thalamus.

with PWS. They face difficulties with theory of mind (understanding the mental states of others), emotion recognition, and application of social norms. Furthermore, they have hardly any peer group relationships. They often prefer the social engagement with older or younger persons (Whittington & Holland, 2010). Some of the social difficulties in PWS are those seen in people with autism spectrum conditions. Emotional development in PWS is in most cases on a much lower level than chronological age (Rice & Einfeld, 2015).

Regarding language development, there are various findings. Expressive language is found to be weaker than receptive language, but the UPD subtype performed better on expressive than receptive language tasks (Chen et al., 2010; Dimitropoulos, Ho, & Feldman, 2013). In daily life, the level of verbal comprehension in PWS is often over- and/or underestimated. There are impairments in receptive language skills due to a tendency to literal interpretation and "concrete" thinking and processing. People with PWS face also difficulties in productive language skills due to speech motor problems (difficulties in articulation) and mental slowness.

Mental Health Disorders

Autism

Atypical social behaviors are common in PWS. Some of the behavioral features apparently overlap with those found in individuals with non-syndromic autism spectrum disorder (ASD). The overall prevalence of ASD-related symptoms in people with PWS has been estimated at 25–27%, which is much higher than is found in typically developing individuals (Bennett et al., 2015; Veltman, Craig, & Bolton, 2005). The prevalence of those symptoms has been shown significantly higher in individuals with the PWS-UPD subtype (35.3%) compared to the PWS-deletion subtype (18.5%) (Bennett et al., 2015).

Different studies, focusing on differentiating ASD symptoms to the genetic subtypes of PWS, have demonstrated significant differences in ASD symptoms in genetic subtype mediated by age or have demonstrated that ASD symptoms directly are more pronounced with increasing age (Lo, Siemensma, Collin, & Hokken-Koelega, 2013; Ogata et al., 2014; Song et al., 2015). The latter found no difference in ASD symptoms between genetic subtypes in children with PWS (n = 22) but did find significant differences between genetic subtypes in adolescents with PWS (n = 23). Consistent with previous findings, the UPD subtype showed significantly higher autistic symptomatology in adolescents with UPD compared to the DEL subtype.

Concerning the restricted or repetitive behaviors, overall, on group level, people with PWS display no more restricted or repetitive behaviors compared to people with ASD or to people with intellectual disability, in terms of met criterium for ASD (Flores et al., 2011; Moss, Oliver, Arron, Burbidge, & Berg, 2009). However, people with PWS do differ in specific aspects of their restricted or repetitive behaviors compared to people with ASD, in that skin picking is the most frequent form of their self-injurious behaviors (Buono, Scannella, & Palmigiano, 2010).

Regarding the social communication impairment (e.g., deficits in reciprocal social interaction), the degree of this symptom is found significantly higher in individuals with PWS UPD subtype compared to individuals with deletion subtype, in both children and (young) adults (Dimitropoulos et al., 2013; Dimitropoulos & Schultz, 2007; Milner et al., 2005; Veltman et al., 2004).

Mood disorders and psychosis

People with PWS often face brief mood swings, changing rapidly, even over the course of the day. Different studies tend to describe the prevalence of mood disorders and psychosis in PWS according to the genetic subtypes and/or age. Mood disorders are frequent in adults with PWS (Verhoeven, Tuinier, & Curfs, 2003; Vogels et al., 2004). Depressive illness is relatively more common in the DEL subtype group (compared to other subtypes), and its prevalence is about the same to that of the ID population (Soni et al., 2007; Soni et al., 2008). Research into psychopathology in adults has been shown that 56% of the DEL subtype was diagnosed with depressive

illness without psychotic symptoms and 85% of the UPD subtype had psychotic symptoms with or without affective component (Sinnema et al., 2011).

In the UPD and IC subtype, the first affective disorder or psychosis is often described in young adulthood, suggesting heightened vulnerabilities for psychiatric disorders in this life stage (Boer et al., 2002; Ho & Dimitropoulos, 2010). The rate of psychosis in PWS has been estimated at 60–100% lifetime prevalence in the UPD subtype and the imprinting center defect (IC) subtype (Whittington & Holland, 2018). Furthermore, the rate of psychosis in the DEL subtype is not higher than that in the intellectually disabled (ID) population. This suggests a strong genetic component in the cause of psychotic symptoms in PWS subtypes with a double expression of a paternally imprinted gene on chromosome 15 (Webb et al., 2008).

Challenging Behaviors

Adults with PWS have higher rates of maladaptive behaviors compared to people with ID due to other etiologies (non-specified ID, Trisomy 21, fragile X syndrome) (Sinnema et al., 2011). Maladaptive or challenging behaviors include stubbornness, impulsivity, difficulty with change in routine, tantrums/temper outbursts, aggression, repetitive and ritualistic behaviors, and self-injurious behaviors (such as skin picking). These features are sometimes described as obsessive compulsive traits. We will describe these features below and demonstrate that these behaviors from a developmental perspective preferably should not be considered as classified psychiatric disorders like OCD (DSM-5) in the general population.

Temper Outbursts and Aggression

Temper outbursts have similar features as those seen in typically developing children, but in PWS these behaviors persist in adolescence and adulthood. Temper outbursts are most often connected with disappointed expectations and are usually associated with routine and food. Young adults show the highest extent behavioral problems (notably stubbornness, mood changes, and biting) compared to children, adolescents, and older adults (Dykens, 2004; Sinnema et al., 2011). During young adulthood, several major changes occur, like finishing school, start working, separation from family life, and moving to living in an institutional accommodation/setting. Additionally, the awareness raises of their dependency as compared to their siblings, as well as their impairments/restrictions and necessary measures. These changes and awareness may lead to an increase in maladaptive behavior. Older adults (30–50 years of age) are described as much less maladaptive, compulsive, and impulsive than young adults. Nevertheless, in older adults with PWS (>45 years of age) behavioral problems are also prevalent but are in this life stage mainly associated with increasing physical morbidity and inactivity (Sinnema et al., 2011).

Restricted and/or Repetitive Behaviors (RRBs) and Self Injurious Behavior

RRBs in PWS are present from an early age (mostly regarding thinking or talking about the same thing repeatedly, hoarding, insistence on routine) but in that stage of life do not apparently differ from typically developing young children. However, in PWS RRBs persist in adolescence and adulthood (Dykens, 2004). These behaviors are also conceptualized as "arrested development" (Holland et al., 2003); particularly an arrest or delay in emotional development that leads to immature coping skills. RRBs correlate negatively with intellectual abilities in PWS (Dykens, Lee, & Roof, 2011). In addition, problems in self-direction (like emotion regulation and executive function) enhance their need for routine.

Skin-picking is the most frequent form of self-injurious behavior in PWS (Buono et al., 2010). Much of the existing skin picking is associated with skin abrasion, and the front of head, arms, legs, hands, and feet are the most common targets (Hustyi, Hammond, Rezvani, & Hall, 2013; Symons, Butler, Sanders, Feurer, & Thompson, 1999). In some cases skin picking has become severe to the extent that medical treatment is required. Skin picking has been interpreted as,

for example, self-harm or OCD-behavior. However, due to hypothalamic dysfunction people with PWS have disturbed pain perception. Skin picking is supposed to be reinforced by automatic sensory stimulation under circumstances in which they are alone or ignored; e.g., it occurs at times of low mood and lack of activity (Hall, Hustyi, Chui, & Hammond, 2014; Whittington & Holland, 2018).

Overall, symptoms of RRBs and skin picking are more prevalent and severe in PWS compared to the general population. Therefore, a strong genetic component related to the PWS region is presumed underlying these behaviors. Taking account of their etiology RRBs and self-injurious behavior in PWS differ from the psychiatric condition OCD (DSM-5). In addition, people in the general population suffer from OCD while they perceive their obsessions aversive, while most people with PWS do not want to change (Whittington & Holland, 2018).

Feeding Problems/Eating Behavior

The tendency to eat excessive amounts of food (hyperphagia) is a core symptom of PWS and is a defining feature of the phenotype. Hyperphagia is caused by the lack of the sense of satiety rather than any psychiatric illness, such as OCD, eating disorders, or addiction (Whittington & Holland, 2018). Without food restrictions and management people with PWS definitely will develop (excessive) behavior problems such as overeating, resulting in marked obesity, cardiovascular diseases, and other medical complications.

Food management should not focus on very strict dietary control (stressing weight control, lower BMI) but instead focus on food planning (offering limited choices of permitted products and measures amounts of food, high in bulk and low in calories), on lowering arousal/stress by closet store of all food stimuli, and on exercising and healthy lifestyle. Weight control/loss is inherently stressful for most persons with PWS, and they often deny food-seeking behavior and prohibited extra food-intake. Caregivers should balance between the need for weight loss and the increased risk of behavioral problems (Table 4.3).

Interdisciplinary Clinical and Research Strategies and Future Vistas

Because of technological and professional innovation and the continued collaboration of clinicians and researchers from various disciplines, deepened insights have been achieved in the genetic, psychopathological, and neuropsychological features of different syndromes. Several examples of this growing knowledge have been presented in this chapter. In this final paragraph, we would like to advocate the continuation of an interdisciplinary strategy, both in clinical and in research settings.

To date, more than 60 years after the first description of Prader-Willi syndrome, several studies (especially including adult patients) seem to be limited by a lack of genetically confirmed diagnosis and/or specification in genetic subtypes. Consequently, syndrome studies may contain heterogeneous populations which complicate generalization of the results. In the interests of further research and patient care, therefore, we encourage clinical genetic testing.

Second, because of the phenotypical similarities between different genetic aberrations, next studies should include genetic comparison groups. Until now, many syndrome-specific studies lack a genetic comparison group or only have a typical developing control group.

Behaviors occur in interaction between biological vulnerability, environmental contingencies, and setting conditions. In genetic syndromes, like PWS, mental health disorders and challenging behaviors become more visible with age, because (a) deficits may not fully manifest until social demands exceed limited capacities; (b) transfer form home to institutional setting is a stressful life event; (c) deficits may be masked by care/coping/containing of primary caregivers; and (d) deficits are less socially accepted outside the primary family/support system when people with genetic syndromes (like PWS) physically impress like adolescents and adults. In PWS, for example, challenging behaviors often reach a peak and psychiatric illness exacerbates in late adolescence and early adult-

Table 4.3 Summary of neuropsychological profiles

Syndrome	ID	Cognitive impairments	DSM symptomatology
Fragile X	Weaknesses in levels of general intellectual functioning Female: normal/ID	Attention (Working) memory Mathematical skills Executive functioning social cognition	ASD Anxiety OCD Mood disorders (females normal intelligence Schizotypal personality disorder (females normal intelligence)
Noonan	Lowered average (mild ID – superior	(Mild) selective and sustained attention (Mild) organization skills Information processing speed (Moderate) social and emotional recognition and expression Alexithymia	Mood disorders Anxiety OCD
Kleefstra	Severe – moderate ID (minority mild ID) (receptive language is superior to the weak/ absence expressive language)		ASD Psychosis
KBG	Mild ID (moderate ID – normal intelligence)	Sustained attention Shifting Visuoconstruction	ADHD ASD Anxiety
PMD	Severe – moderate ID (impaired to absent speech and expressive language)	Increased sensitivity to sensory stimu	ASD Bipolar affective domain
PWS	Mild ID (moderate ID – mild ID) Expressive language is weaker than receptive language (except for UPD subtype)	Attention Working memory Auditory processing (worse than visual processing) Executive functioning (cognitive flexibility) compared to both the general population as Fragile X Emotional development (much lower than chronological age)	ASD (25–27%) Significantly higher symptoms in UPD compared to the DEL Psychosis Rate 60–100% lifetime prevalence in the UPD and IC 85% of UPD has symptoms Rate in DEL subtype is not higher than in ID Depression (56% of DEL)

	Behavior	Interventions
Fragile X	Social anxiety and withdrawal behavior Stereotypies (flapping or biting of the hands) Perseverations Extreme sensitivity to environmental stimuli Decreased social reciprocity with an avoidance of eye contact Hyperactivity/impulsivity Aggression	Computer based cognitive methods that target cognitive functioning and educational learning Learning strategies that incorporate visually based, experiential or holistic learning were reported to be the most successful
Noonan	Mild signs of anxiety and lowered mood	Interventions in improving social cognition in adult neuropsychiatric patients are effective. It is important to help patients to give words to their inner experiences and improve their social cognition (e.g. emotion recognition) by dedicated interventions.

(continued)

Table 4.3 (continued)

	Behavior	Interventions
Kleefstra	Attention problems Stereotypies and self-injurious behavior Behavior difficulties such as aggression, impulsivity, and depressive and chaotic behaviors Regression During adolescence a gradual decline of previously learned motor, language and communication. Regression is observed in terms of goal directed behavior (performing daily skills) and goal directed cognition (interest and curiosity) as well as in an almost complete lack of emotional and social reciprocity	Decline in functioning is expected to be an expression of a psychotic episode followed by sleep disturbances and for that reason needs to be treated with antipsychotics in order to restore sleep and halt further regression. Recommended to periodically asses the level of adaptive functioning and in relation to maladaptive functioning in all patients in order to both detect regression and overcome under- or overstimulation in counseling en school functioning.
KBG	Anxiety Aggression Compulsive behavior In systematic study: distractibility, impulsivity and restless behavior	Importance of providing external structure (including tasks monitoring and controlling of processing speed) in educational trajectories.
PMD	Increased sensitivity to sudden environmental events Neurobehavioral regression, i.e., loss of acquired skills either permanently or for an extended period and often with co-occurring catatonic phenomena, which may be triggered by stressful events, seizures or infection	Short term studies indicate that injections with insulin-like growth factor 1 or intranasal administration of insulin may have some beneficial effects on aspects of social functioning.
PWS	PWS equal restricted or repetitive behaviors compared to people with ASD or to people with ID. Maladaptive or challenging behaviors include stubbornness, impulsivity, difficulty with change in routine, tantrums/temper outbursts, aggression, repetitive and ritualistic behaviors, and self-injurious behaviors ≠ OCD Temper outbursts are most often connected with disappointed expectations and are usually associated with routine and food. Young adults show to the highest extent behavioral problems which may be a result of changes and awareness. Restricted and/or repetitive behaviors (mostly regarding thinking or talking about the same thing repeatedly, hoarding, insistence on routine); an arrest or delay in emotional development that leads to immature coping skills. Skin-picking is the most frequent form of self-injurious behavior (disturbed pain perception) ≠ OCD Hyperphagia (lack of the sense of satiety rather) ≠ OCD, eating disorders, addiction	Do not rely on verbal capacities (only), adapt to slow speed of information processing. Visually support verbal and written communication. Help to structure information Provide external sources of emotion regulation appropriate to the social and emotional developmental age. Skin picking: a sufficient number of caregivers, provide with (relaxation) activities. Food management: food planning, lowering arousal/stress by closet store of all food stimuli, and on exercising and healthy lifestyle. Support caregivers, for example by psycho-education of PWS and understanding the individuals strengths and weaknesses of the PWS patient.

hood. Consequently, thirdly, we recommend early neuropsychological assessment and monitoring behaviors across life span and being sensitive to the different needs at different stages of life. For the same reasons, we also advocate for research studies that are longitudinal in design, not mainly cross-sectional.

As this chapter shows, psychiatric comorbidities, such as autism spectrum disorder (ASD), anxiety disorders, depressions, and attention deficit hyperactivity disorder (ADHD) are highly prevalent in genetic disorders. Symptoms of psychopathology may overlap behavioral problems. Increases in behavioral problems may be early indicators of underlying medical or psychiatric disorders, vice versa. Therefore, fourthly, persons with genetic disorders would benefit from detailed, individual neuropsychological assess-

ment of various domains of cognitive, behavioral, psychiatric strengths and weaknesses, and contextual factors. Consequently, physicians, psychologists, caregivers, and researchers can become more aware of individual strengths and weaknesses in cognitive and behavioral functioning. These assessments also can help characterize and support everyday somatic and functional difficulties. Fifthly, the application of golden standard (symptomatology) assessment tools, like for intelligence, ASD, would be helpful to address under- and over-diagnosis of psychiatric disorders, to further understand somatic comorbidities as well as to develop common clinical guidelines and research frameworks.

As the syndrome descriptions in this chapter demonstrate, many syndromes are characterized by intellectual disability and/or learning problems. Learning difficulties, for example, require attention for special educational needs. At the same time, discerning syndromes show specific cognitive and behavioral profiles and also individual differences within syndromes. This stresses the importance of the development of personalized treatment plans. Together with the aforementioned high rates of psychiatric disorders, the cognitive and behavioral profiles also stress the significant needs for specialized therapies and services, including speech-language, occupational, and physical therapies; special education services; and behavior management. In addition, specialized medical care, such as consultations with developmental-behavioral pediatricians or neurologists, is required.

Challenging behavior typically has the greatest impact on persons with genetic syndromes and in particular their families and caregivers, over other aspects of the syndrome. Psychotropic drugs can only "control" these behaviors to a certain extent. Hence, the importance of contextual interventions (like psycho-education for proxy, systemic therapy, communication strategies, design of residential facilities) is evident, which highlights the significance of personalized counselling based on neuropsychological profiles.

Conclusion

To summarize, this chapter aims at providing an overview of several common and rare genetic syndromes including their neuropsychological and/or psychopathological phenotype that could be delineated by detailed and extensive assessment of neurocognitive functioning and psychiatric symptoms. It may have been demonstrated that the genetic neuropsychological paradigm is of marked scientific and clinical relevance for the study of rare genetic diseases. Moreover, the described developments stress the importance of clinical awareness of psychological and psychiatric manifestations of genetic syndromes in patients with intellectual disability and the increasing importance to implement a structured collaboration between neuropsychologists, psychiatrists, and clinical geneticists.

In all, the etiologically driven established patterns of (dys)functioning point at specific therapeutic options and clinical strategies, implying that the abovementioned paradigm is an essential prerequisite to further clarify genotype-phenotype relations, advance clinical diagnostic methods, and help establishing individualized behavioral and clinical management programs.

References

Aldinger, K. A., Kogan, J., Kimonis, V., Fernandez, B., Horn, D., Klopocki, E., … Millen, K. J. (2013). Cerebellar and posterior fossa malformations in patients with autism-associated chromosome 22q13 terminal deletion. *American Journal of Medical Genetics, Part A, 161*(1), 131–136.

Allanson, J. E., Bohring, A., Dörr, H. G., Dufke, A., Gillessen-Kaesbach, G., Horn, D., … Pauli, S. (2010). The face of Noonan syndrome: Does phenotype predict genotype. *American Journal of Medical Genetics, Part A, 152*(8), 1960–1966.

Bennett, J. A., Germani, T., Haqq, A. M., & Zwaigenbaum, L. (2015). Autism spectrum disorder in Prader-Willi syndrome: A systematic review. *American Journal of Medical Genetics, Part A, 167A*(12), 2936–2944. https://doi.org/10.1002/ajmg.a.37286

Boer, H., Holland, A., Whittington, J., Butler, J., Webb, T., & Clarke, D. (2002). Psychotic illness in people with Prader Willi syndrome due to chromosome 15 maternal uniparental disomy. *The Lancet, 359*(9301), 135–136.

Breckpot, J., Vercruyssen, M., Weyts, E., Vandevoort, S., D'Haenens, G., Van Buggenhout, G., … Renieri, A. (2016). Copy number variation analysis in adults with catatonia confirms haploinsufficiency of SHANK3 as a predisposing factor. *European Journal of Medical Genetics, 59*(9), 436–443.

Buono, S., Scannella, F., & Palmigiano, M. B. (2010). Self-injurious behavior: A comparison between Prader-Willi syndrome, down syndrome and autism. *Life Span and Disability, 2*, 187–201.

Butler, M. G., Kimonis, V., Dykens, E., Gold, J. A., Miller, J., Tamura, R., & Driscoll, D. J. (2018). Prader–Willi syndrome and early-onset morbid obesity NIH rare disease consortium: A review of natural history study. *American Journal of Medical Genetics, Part A, 176*(2), 368–375.

Chen, C.-M., Chen, C.-L., Hou, J.-W., Hsu, H.-C., Chung, C.-Y., Chou, S.-W., … Chen, K.-H. (2010). Developmental profiles and mentality in preschool children with Prader-Willi syndrome: A preliminary study. *Chang Gung Medical Journal, 33*(4), 436–442.

Ciaccio, C., Scuvera, G., Tucci, A., Gentilin, B., Baccarin, M., Marchisio, P., … Milani, D. (2018). New insights into Kleefstra syndrome: Report of two novel cases with previously unreported features and literature review. *Cytogenetic and Genome Research, 156*(3), 127–133. https://doi.org/10.1159/000494532

Copet, P., Jauregi, J., Laurier, V., Ehlinger, V., Arnaud, C., Cobo, A. M., … Thuilleaux, D. (2010). Cognitive profile in a large french cohort of adults with Prader–Willi syndrome: Differences between genotypes. *Journal of Intellectual Disability Research, 54*(3), 204–215.

Costales, J. L., & Kolevzon, A. (2015). Phelan–McDermid syndrome and SHANK3: Implications for treatment. *Neurotherapeutics, 12*(3), 620–630.

Curfs, L., Wiegers, A., Sommers, J., Borghgraef, M., & Fryns, J. (1991). Strengths and weaknesses in the cognitive profile of youngsters with Prader-Willi syndrome. *Clinical Genetics, 40*(6), 430–434.

Cusmano-Ozog, K., Manning, M. A., & Hoyme, H. E. (2007). *22q13.3 deletion syndrome: a recognizable malformation syndrome associated with marked speech and language delay.* Paper presented at the American Journal of Medical Genetics Part C: Seminars in Medical Genetics.

De Rubeis, S., Siper, P. M., Durkin, A., Weissman, J., Muratet, F., Halpern, D., … Wang, A. T. (2018). Delineation of the genetic and clinical spectrum of Phelan-McDermid syndrome caused by SHANK3 point mutations. *Molecular Autism, 9*(1), 31.

Denayer, A., Van Esch, H., De Ravel, T., Frijns, J.-P., Van Buggenhout, G., Vogels, A., … Swillen, A. (2012). Neuropsychopathology in 7 patients with the 22q13 deletion syndrome: Presence of bipolar disorder and progressive loss of skills. *Molecular syndromology, 3*(1), 14–20.

Dimitropoulos, A., Ho, A., & Feldman, B. (2013). Social responsiveness and competence in Prader-Willi syndrome: Direct comparison to autism spectrum disorder. *Journal of Autism and Developmental Disorders, 43*(1), 103–113.

Dimitropoulos, A., & Schultz, R. T. (2007). Autistic-like symptomatology in Prader-Willi syndrome: A review of recent findings. *Current Psychiatry Reports, 9*(2), 159–164.

Dykens, E. M. (2004). Maladaptive and compulsive behavior in Prader-Willi syndrome: New insights from older adults. *American Journal on Mental Retardation, 109*(2), 142–153.

Dykens, E. M., Lee, E., & Roof, E. (2011). Prader-Willi syndrome and autism spectrum disorders: An evolving story. *Journal of Neurodevelopmental Disorders, 3*(3), 225.

Dyscerne, Noonan Syndrome Guideline Development Group. (2010). *Management of Noonan syndrome: A clinical guideline*. Retrieved from https://www.noonansyndroom.nl/activiteiten/kennisbank-noonan-syndroom/send/11-informatie-over-noonan-syndroom-engels/26-clinical-guidelines-noonan-syndrome.

Egger, J. I. M., Verhoeven, W. M. A., Groenendijk-Reijenga, R., & Kant, S. G. (2017). Phelan-McDermid syndrome due to SHANK3 mutation in an intellectually disabled adult male: Successful treatment with lithium. *BMJ Case Reports, 2017*, bcr2017220778. https://doi.org/10.1136/bcr-2017-220778

Egger, J. I. M., Zwanenburg, R. J., van Ravenswaaij-Arts, C. M. A., Kleefstra, T., & Verhoeven, W. M. A. (2016). Neuropsychological phenotype and psychopathology in seven adult patients with Phelan-McDermid syndrome: Implications for treatment strategy. *Genes, Brain and Behavior, 15*(4), 395–404.

Figura, M. G., Coppola, A., Bottitta, M., Calabrese, G., Grillo, L., Luciano, D., … Elia, M. (2014). Seizures and EEG pattern in the 22q13. 3 deletion syndrome: Clinical report of six Italian cases. *Seizure, 23*(9), 774–779.

Flores, C. G., Valcante, G., Guter, S., Zaytoun, A., Wray, E., Bell, L., … Cook, E. H. (2011). Repetitive behavior profiles: Consistency across autism spectrum disorder cohorts and divergence from Prader–Willi syndrome. *Journal of Neurodevelopmental Disorders, 3*(4), 316.

Franke, P., Leboyer, M., Hardt, J., Sohne, E., Weiffenbach, O., Biancalana, V., … Schwab, S. G. (1999). Neuropsychological profiles of FMR-1 premutation and full-mutation carrier females. *Psychiatry Research, 87*(2–3), 223–231.

Gallagher, D., Voronova, A., Zander, M. A., Cancino, G. I., Bramall, A., Krause, M. P., … Miller, F. D. (2015). Ankrd11 is a chromatin regulator involved in autism that is essential for neural development. *Developmental Cell, 32*(1), 31–42. https://doi.org/10.1016/j.devcel.2014.11.031

Goldenberg, A., Riccardi, F., Tessier, A., Pfundt, R., Busa, T., Cacciagli, P., … Philip, N. (2016). Clinical and molecular findings in 39 patients with KBG syndrome caused by deletion or mutation of ANKRD11. *American Journal of Medical Genetics,*

Part A, 170(11), 2847–2859. https://doi.org/10.1002/ajmg.a.37878

Guilmatre, A., Huguet, G., Delorme, R., & Bourgeron, T. (2014). The emerging role of SHANK genes in neuropsychiatric disorders. *Developmental Neurobiology, 74*(2), 113–122.

Hagerman, R. J., Ono, M. Y., & Hagerman, P. J. (2005). Recent advances in fragile X: A model for autism and neurodegeneration. *Current Opinion in Psychiatry, 18*(5), 490–496.

Hah, M., Lotspeich, L. J., Phillips, J. M., Torres, A. D., Cleveland, S. C., & Hallmayer, J. F. (2009). Twins with KBG syndrome and autism. *Journal of Autism and Developmental Disorders, 39*, 1744.

Hall, S. S., Hustyi, K. M., Chui, C., & Hammond, J. L. (2014). Experimental functional analysis of severe skin-picking behavior in Prader-Willi syndrome. *Research in Developmental Disabilities, 35*(10), 2284–2292.

Hernandez, R. N., Feinberg, R. L., Vaurio, R., Passanante, N. M., Thompson, R. E., & Kaufmann, W. E. (2009). Autism spectrum disorder in fragile X syndrome: a longitudinal evaluation. *American Journal of Medical Genetics, Part A, 149*(6), 1125–1137.

Herrmann, J., Pallister, P. D., Tiddy, W., & Opitz, J. M. (1975). The KBG syndrome – A syndrome of short stature, characteristic facies, mental retardation, macrodontia and skeletal anomalies. *Birth Defects Original Article Series, 11*, 7–18.

Hessl, D., Rivera, S. M., & Reiss, A. L. (2004). The neuroanatomy and neuroendocrinology of fragile X syndrome. *Mental Retardation and Developmental Disabilities Research Reviews, 10*(1), 17–24.

Ho, A. Y., & Dimitropoulos, A. (2010). Clinical management of behavioral characteristics of Prader-Willi syndrome. *Neuropsychiatric Disease and Treatment, 6*, 107.

Holder, J. L., Jr., & Quach, M. M. (2016). The spectrum of epilepsy and electroencephalographic abnormalities due to SHANK 3 loss-of-function mutations. *Epilepsia, 57*(10), 1651–1659.

Holland, A., Whittington, J., Butler, J., Webb, T., Boer, H., & Clarke, D. (2003). Behavioural phenotypes associated with specific genetic disorders: Evidence from a population-based study of people with Prader-Willi syndrome. *Psychological Medicine, 33*(1), 141–153.

Hustyi, K. M., Hammond, J. L., Rezvani, A. B., & Hall, S. S. (2013). An analysis of the topography, severity, potential sources of reinforcement, and treatments utilized for skin picking in Prader-Willi syndrome. *Research in Developmental Disabilities, 34*(9), 2890–2899.

Jauregi, J., Arias, C., Vegas, O., Alen, F., Martinez, S., Copet, P., & Thuilleaux, D. (2007). A neuropsychological assessment of frontal cognitive functions in Prader-Willi syndrome. *Journal of Intellectual Disability Research, 51*(5), 350–365.

Kim, H. J., Cho, E., Park, J. B., Im, W. Y., & Kim, H. J. (2015). A Korean family with KBG syndrome identified by ANKRD11 mutation, and phenotypic comparison of ANKRD11 mutation and 16q24.3 microdeletion. *European Journal of Medical Genetics, 58*(2), 86–94. https://doi.org/10.1016/j.ejmg.2014.11.003

Kleefstra, T., Nillesen, W. M., & Yntema, H. G. (2010 7-5-2015). Kleefstra syndrome. *GeneReviews®*.

Kolevzon, A., Angarita, B., Bush, L., Wang, A. T., Frank, Y., Yang, A., … Farrell, C. (2014). Phelan-McDermid syndrome: A review of the literature and practice parameters for medical assessment and monitoring. *Journal of Neurodevelopmental Disorders, 6*(1), 39.

Kolevzon, A., Bush, L., Wang, A. T., Halpern, D., Frank, Y., Grodberg, D., … Soorya, L. (2014). A pilot controlled trial of insulin-like growth factor-1 in children with Phelan-McDermid syndrome. *Molecular Autism, 5*(1), 54.

Lo, S. T., Siemensma, E., Collin, P., & Hokken-Koelega, A. (2013). Impaired theory of mind and symptoms of autism spectrum disorder in children with Prader-Willi syndrome. *Research in Developmental Disabilities, 34*(9), 2764–2773.

Low, K., Ashraf, T., Canham, N., Clayton-Smith, J., Deshpande, C., Donaldson, A., … Smithson, S. (2016). Clinical and genetic aspects of KBG syndrome. *American Journal of Medical Genetics, Part A, 170*(11), 2835–2846. https://doi.org/10.1002/ajmg.a.37842

Lubs, H. A. (1969). A marker X chromosome. *American Journal of Human Genetics, 21*(3), 231.

Luciani, J., De Mas, P., Depetris, D., Mignon-Ravix, C., Bottani, A., Prieur, M., … De Martinville, B. (2003). Telomeric 22q13 deletions resulting from rings, simple deletions, and translocations: Cytogenetic, molecular, and clinical analyses of 32 new observations. *Journal of Medical Genetics, 40*(9), 690–696.

Manning, K., McAllister, C. J., Ring, H. A., Finer, N., Kelly, C. L., Sylvester, K. P., … Manford, M. R. (2016). Novel insights into maladaptive behaviours in Prader–Willi syndrome: Serendipitous findings from an open trial of vagus nerve stimulation. *Journal of Intellectual Disability Research, 60*(2), 149–155.

Manning, K. E., & Holland, A. J. (2015). Puzzle pieces: Neural structure and function in Prader-Willi syndrome. *Diseases, 3*(4), 382–415. https://doi.org/10.3390/diseases3040382

Marco, E. J., & Skuse, D. H. (2006). Autism-lessons from the X chromosome. *Social Cognitive and Affective Neuroscience, 1*(3), 183–193.

Milner, K. M., Craig, E. E., Thompson, R. J., Veltman, M. W., Simon Thomas, N., Roberts, S., … Bolton, P. F. (2005). Prader-Willi syndrome: Intellectual abilities and behavioural features by genetic subtype. *Journal of Child Psychology and Psychiatry, 46*(10), 1089–1096.

Mitz, A. R., Philyaw, T. J., Boccuto, L., Shcheglovitov, A., Sarasua, S. M., Kaufmann, W. E., & Thurm, A. (2018). Identification of 22q13 genes most likely to contribute to Phelan McDermid syndrome. *European Journal of Human Genetics, 26*(3), 293. https://doi.org/10.1038/s41431-017-0042-x

Miyatake, S., Murakami, A., Okamoto, N., Sakamoto, M., Miyake, N., Saitsu, H., & Matsumoto, N. (2013). A de novo deletion at 16q24.3 involving ANKRD11 in a Japanese patient with KBG syndrome. *American Journal of Medical Genetics, Part A, 161A*(5), 1073–1077. https://doi.org/10.1002/ajmg.a.35661

Moss, J., Oliver, C., Arron, K., Burbidge, C., & Berg, K. (2009). The prevalence and phenomenology of repetitive behavior in genetic syndromes. *Journal of Autism and Developmental Disorders, 39*(4), 572–588.

Murray, N., Burgess, B., Hay, R., Colley, A., Rajagopalan, S., McGaughran, J., … Goel, H. (2017). KBG syndrome: An Australian experience. *American Journal of Medical Genetics, Part A, 173*(7), 1866–1877. https://doi.org/10.1002/ajmg.a.38121

Noonan, J. A. (1968). Hypertelorism with turner phenotype. A new syndrome with associated congenital heart disease. *American Journal of Diseases of Children, 116*(4), 373–380.

Novara, F., Rinaldi, B., Sisodiya, S. M., Coppola, A., Giglio, S., Stanzial, F., … Zuffardi, O. (2017). Haploinsufficiency for ANKRD11-flanking genes makes the difference between KBG and 16q24.3 microdeletion syndromes: 12 new cases. *European Journal of Human Genetics, 25*(6), 694–701. https://doi.org/10.1038/ejhg.2017.49

Oberman, L. M., Boccuto, L., Cascio, L., Sarasua, S., & Kaufmann, W. E. (2015). Autism spectrum disorder in Phelan-McDermid syndrome: Initial characterization and genotype-phenotype correlations. *Orphanet Journal of Rare Diseases, 10*(1), 105.

Ockeloen, C. W., Willemsen, M. H., de Munnik, S., van Bon, B. W., de Leeuw, N., Verrips, A., … Kleefstra, T. (2015). Further delineation of the KBG syndrome phenotype caused by ANKRD11 aberrations. *European Journal of Human Genetics, 23*(9), 1176–1185. https://doi.org/10.1038/ejhg.2014.253

Ogata, H., Ihara, H., Murakami, N., Gito, M., Kido, Y., & Nagai, T. (2014). Autism spectrum disorders and hyperactive/impulsive behaviors in japanese patients with Prader-Willi syndrome: A comparison between maternal uniparental disomy and deletion cases. *American Journal of Medical Genetics, Part A, 164*(9), 2180–2186.

OMIM® – Online Mendelian Inheritance in Man. (2019, 25-03-2019). Retrieved from https://www.omim.org/about.

Phelan, K., & McDermid, H. E. (2011). The 22q13.3 deletion syndrome (Phelan-McDermid syndrome). *Molecular syndromology, 2*(3–5), 186–201.

Phelan, M. C. (2008). Deletion 22q13.3 syndrome. *Orphanet Journal of Rare Diseases, 3*(1), 14.

Phelan, M. C., Rogers, R. C., & Stevenson, R. E. (1988). A de novo terminal deletion of 22q. *American Journal of Human Genetics, 43*, A118.

Philippe, A., Boddaert, N., Vaivre-Douret, L., Robel, L., Danon-Boileau, L., Malan, V., … Golse, B. (2008). Neurobehavioral profile and brain imaging study of the 22q13.3 deletion syndrome in childhood. *Pediatrics, 122*(2), e376–e382.

Raspa, M., Wheeler, A. C., & Riley, C. (2017). Public health literature review of fragile X syndrome. *Pediatrics, 139*(Suppl 3), S153–S171. https://doi.org/10.1542/peds.2016-1159C

Rice, L. J., & Einfeld, S. L. (2015). Cognitive and behavioural aspects of Prader-Willi syndrome. *Current Opinion in Psychiatry, 28*(2), 102–106. https://doi.org/10.1097/YCO.0000000000000135

Roelofs, R. L., Wingbermühle, E., Kessels, R. P., & Egger, J. I. (2019). Social cognitive training for adults with Noonan syndrome: A feasibility study. *Neuropsychiatric Disease and Treatment, 15*, 611.

Sarasua, S. M., Boccuto, L., Sharp, J. L., Dwivedi, A., Chen, C.-F., Rollins, J. D., … DuPont, B. R. (2014). Clinical and genomic evaluation of 201 patients with Phelan-McDermid syndrome. *Human Genetics, 133*(7), 847–859.

Schmidt, H., Kern, W., Giese, R., Hallschmid, M., & Enders, A. (2009). Intranasal insulin to improve developmental delay in children with 22q13 deletion syndrome: An exploratory clinical trial. *Journal of Medical Genetics, 46*(4), 217–222.

Serret, S., Thümmler, S., Dor, E., Vesperini, S., Santos, A., & Askenazy, F. (2015). Lithium as a rescue therapy for regression and catatonia features in two SHANK3 patients with autism spectrum disorder. *BMC Psychiatry, 15*(1), 107.

Sinnema, M., Einfeld, S. L., Schrander-Stumpel, C. T., Maaskant, M. A., Boer, H., & Curfs, L. M. (2011). Behavioral phenotype in adults with Prader-Willi syndrome. *Research in Developmental Disabilities, 32*(2), 604–612. https://doi.org/10.1016/j.ridd.2010.12.014

Sinnema, M., Schrander-Stumpel, C. T., Maaskant, M. A., Boer, H., & Curfs, L. M. (2012). Aging in Prader-Willi syndrome: Twelve persons over the age of 50 years. *American Journal of Medical Genetics, Part A, 158*(6), 1326–1336.

Sirmaci, A., Spiliopoulos, M., Brancati, F., Powell, E., Duman, D., Abrams, A., … Konuk, B. (2011). Mutations in ANKRD11 cause KBG syndrome, characterized by intellectual disability, skeletal malformations, and macrodontia. *American Journal of Human Genetics, 89*(2), 289–294.

Skjei, K. L., Martin, M. M., & Slavotinek, A. M. (2007). KBG syndrome: Report of twins, neurological characteristics, and delineation of diagnostic criteria. *American Journal of Medical Genetics, Part A, 143A*(3), 292–300. https://doi.org/10.1002/ajmg.a.31597

Song, D. K., Sawada, M., Yokota, S., Kuroda, K., Uenishi, H., Kanazawa, T., … Shimoda, K. (2015). Comparative analysis of autistic traits and behavioral disorders in Prader-Willi syndrome and Asperger disorder. *American Journal of Medical Genetics, Part A, 167*(1), 64–68.

Soni, S., Whittington, J., Holland, A., Webb, T., Maina, E., Boer, H., & Clarke, D. (2007). The course and outcome of psychiatric illness in people with Prader–Willi syndrome: Implications for management and

treatment. *Journal of Intellectual Disability Research, 51*(1), 32–42.

Soni, S., Whittington, J., Holland, A., Webb, T., Maina, E., Boer, H., & Clarke, D. (2008). The phenomenology and diagnosis of psychiatric illness in people with Prader-Willi syndrome. *Psychological Medicine, 38*(10), 1505–1514.

Srivastava, S., Scherrer, B., Prohl, A. K., Filip-Dhima, R., Kapur, K., Kolevzon, A., … Thurm, A. (2019). Volumetric analysis of the basal ganglia and cerebellar structures in patients with Phelan-McDermid syndrome. *Pediatric Neurology, 90*, 37–43.

Symons, F. J., Butler, M., Sanders, M., Feurer, I., & Thompson, T. (1999). Self-injurious behavior and Prader-Willi syndrome: Behavioral forms and body locations. *American Journal on Mental Retardation, 104*(3), 260–269.

Tabet, A.-C., Rolland, T., Ducloy, M., Lévy, J., Buratti, J., Mathieu, A., … Passemard, S. (2017). A framework to identify contributing genes in patients with Phelan-McDermid syndrome. *NPJ Genomic Medicine, 2*(1), 32.

Tunovic, S., Barkovich, J., Sherr, E. H., & Slavotinek, A. M. (2014). De novo ANKRD11 and KDM1A gene mutations in a male with features of KBG syndrome and Kabuki syndrome. *American Journal of Medical Genetics. Part A, 164A*(7), 1744–1749. https://doi.org/10.1002/ajmg.a.36450

Uchino, S., & Waga, C. (2015). Novel therapeutic approach for autism spectrum disorder: Focus on SHANK3. *Current Neuropharmacology, 13*(6), 786–792.

Van Der Burgt, I., Berends, E., Lommen, E., Van Beersum, S., Hamel, B., & Mariman, E. (1994). Clinical and molecular studies in a large Dutch family with Noonan syndrome. *American Journal of Medical Genetics, 53*(2), 187–191.

van Dongen, L. C., Wingbermühle, E., van der Veld, W. M., Vermeulen, K., Bos-Roubos, A. G., Ockeloen, C. W., … Egger, J. I. (2019). Exploring the behavioral and cognitive phenotype of KBG syndrome. *Genes, Brain and Behavior, 18*(4), e12553.

Veltman, M. W., Craig, E. E., & Bolton, P. F. (2005). Autism spectrum disorders in Prader–Willi and Angelman syndromes: A systematic review. *Psychiatric Genetics, 15*(4), 243–254.

Veltman, M. W., Thompson, R. J., Roberts, S. E., Thomas, N. S., Whittington, J., & Bolton, P. F. (2004). Prader-Willi syndrome: A study comparing deletion and uniparental disomy cases with reference to autism spectrum disorders. *European Child & Adolescent Psychiatry, 13*(1), 42–50.

Verhoeven, W., Tuinier, S., & Curfs, L. (2003). Prader-Willi syndrome: Cycloid psychosis in a genetic subtype? *Acta Neuropsychiatrica, 15*(1), 32–37.

Verhoeven, W., Wingbermühle, E., Egger, J., Van der Burgt, I., & Tuinier, S. (2008). Noonan syndrome: Psychological and psychiatric aspects. *American Journal of Medical Genetics, Part A, 146A*(2), 191–196. https://doi.org/10.1002/ajmg.a.32115

Verhoeven, W. M. A., Egger, J. I. M., Cohen-Snuijf, R., Kant, S. G., & de Leeuw, N. (2013). Phelan-McDermid syndrome: Clinical report of a 70-year-old woman. *American Journal of Medical Genetics, Part A, 161*(1), 158–161.

Verhoeven, W. M. A., Egger, J. I. M., Vermeulen, K., van de Warrenburg, B. P., & Kleefstra, T. (2011). Kleefstra syndrome in three adult patients: Further delineation of the behavioral and neurological phenotype shows aspects of a neurodegenerative course. *American Journal of Medical Genetics, Part A, 155A*(10), 2409–2415. https://doi.org/10.1002/ajmg.a.34186

Verhoeven, W. M. A., Egger, J. I. M., Willemsen, M. H., de Leijer, G. J., & Kleefstra, T. (2012). Phelan-McDermid syndrome in two adult brothers: Atypical bipolar disorder as its psychopathological phenotype? *Neuropsychiatric Disease and Treatment, 8*, 175.

Verhoeven, W. M. A., Kleefstra, T., & Egger, J. I. M. (2010). Behavioral phenotype in the 9q subtelomeric deletion syndrome: A report about two adult patients. *American Journal of Medical Genetics, Part B, Neuropsychiatric Genetics, 153B*(2), 536–541. https://doi.org/10.1002/ajmg.b.31015

Vermeulen, K., de Boer, A., Janzing, J. G. E., Koolen, D. A., Ockeloen, C. W., Willemsen, M. H., … Kleefstra, T. (2017). Adaptive and maladaptive functioning in Kleefstra syndrome compared to other rare genetic disorders with intellectual disabilities. *American Journal of Medical Genetics, Part A, 173*(7), 1821–1830. https://doi.org/10.1002/ajmg.a.38280

Vermeulen, K., Staal, W. G., Janzing, J. G., Buitelaar, J. K., van Bokhoven, H., Egger, J. I. M., & Kleefstra, T. (2015). From a single gene defect towards a cross species neurocognitive phenotype: The EHMT1 Disruption Example (Kleefstra Syndrome). *Austin Journal of Autism & Related Disabilities, 1*(2), 1009.

Vermeulen, K., Staal, W. G., Janzing, J. G., van Bokhoven, H., Egger, J. I. M., & Kleefstra, T. (2017). Sleep disturbance as a precursor of severe regression in Kleefstra syndrome suggests a need for firm and rapid pharmacological treatment. *Clinical Neuropharmacology, 40*(4), 185–188. https://doi.org/10.1097/WNF.0000000000000226

Vogels, A., Hert, M. D., Descheemaeker, M., Govers, V., Devriendt, K., Legius, E., … Fryns, J.-P. (2004). Psychotic disorders in Prader–Willi syndrome. *American Journal of Medical Genetics, Part A, 127*(3), 238–243.

Walz, K., Cohen, D., Neilsen, P. M., Foster, J., 2nd, Brancati, F., Demir, K., … Tekin, M. (2015). Characterization of ANKRD11 mutations in humans and mice related to KBG syndrome. *Human Genetics, 134*(2), 181–190. https://doi.org/10.1007/s00439-014-1509-2

Webb, T., Maina, E. N., Soni, S., Whittington, J., Boer, H., Clarke, D., & Holland, A. (2008). In search of the psychosis gene in people with Prader-Willi syn-

drome. *American Journal of Medical Genetics, Part A, 146*(7), 843–853.

Whittington, J., & Holland, A. (2010). Neurobehavioral phenotype in Prader-Willi syndrome. *American Journal of Medical Genetics, Part C: Seminars in Medical Genetics, 154C*(4), 438–447. https://doi.org/10.1002/ajmg.c.30283

Whittington, J., & Holland, A. (2017). Cognition in people with Prader-Willi syndrome: Insights into genetic influences on cognitive and social development. *Neuroscience & Biobehavioral Reviews, 72*, 153–167.

Whittington, J., & Holland, A. (2018). A review of psychiatric conceptions of mental and behavioural disorders in Prader-Willi syndrome. *Neuroscience & Biobehavioral Reviews, 95*, 396–405. https://doi.org/10.1016/j.neubiorev.2018.10.006

Wilson, H., Wong, A., Shaw, S., Tse, W., Stapleton, G., Phelan, M., … McDermid, H. (2003). Molecular characterisation of the 22q13 deletion syndrome supports the role of haploinsufficiency of SHANK3/PROSAP2 in the major neurological symptoms. *Journal of Medical Genetics, 40*(8), 575–584.

Wingbermühle, E., Egger, J., van der Burgt, I., & Verhoeven, W. (2009). Neuropsychological and behavioral aspects of Noonan syndrome. *Hormone Research in Pædiatrics, 72*(Suppl 2), 15–23. https://doi.org/10.1159/000243774

Wingbermühle, E., Egger, J. I. M., Verhoeven, W. M. A., van der Burgt, I., & Kessels, R. P. C. (2012). Affective functioning and social cognition in Noonan syndrome. *Psychological Medicine, 42*(2), 419–426. https://doi.org/10.1017/S0033291711001115

Wingbermuhle, E., Roelofs, R. L., van der Burgt, I., Souren, P. M., Verhoeven, W. M. A., Kessels, R. P., & Egger, J. I. M. (2012). Cognitive functioning of adults with Noonan syndrome: A case-control study. *Genes, Brain and Behavior, 11*(7), 785–793. https://doi.org/10.1111/j.1601-183X.2012.00821.x

Zwanenburg, R. J., Bocca, G., Ruiter, S. A., Dillingh, J. H., Flapper, B. C., van den Heuvel, E. R., & van Ravenswaaij-Arts, C. M. (2016). Is there an effect of intranasal insulin on development and behaviour in Phelan-McDermid syndrome? A randomized, double-blind, placebo-controlled trial. *European Journal of Human Genetics, 24*(12), 1696.

5 Effects of IQ and Adaptive Behavior on Assessment and Treatment in Dual Diagnosis

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Introduction

An intellectual disability (ID) is a developmental disorder that impacts functioning over the course of an individual's life span and is marked by the early onset of limitations in both intellectual and adaptive domains. Prevalence rates in the general population have been estimated to be approximately 1%, with significant variability noted based on factors such as age (American Psychiatric Association [APA], 2013). While the onset of developmental delays must manifest prior to the age of 18, the presentation of characteristics and features of ID depends on etiology and severity. Both genetic and environmental factors contribute to this condition, with the most common causes of the disorder being linked to chromosomal abnormalities and fetal exposure to teratogens (Walker & Johnson, 2006). Individuals with ID also present with a wide variety of comorbid conditions including developmental disorders, neurological disorders, and behavioral disorders (Johnson, Walker, Palomo-Gonzalez, & Curry, 2006). The degree of diversity in etiology and associated medical, psychiatric, and social features in this population presents numerous diagnostic and intervention planning challenges (Turygin, Matson, Adams, & Williams, 2014). Despite the frequency with which individuals with ID present with comorbid disorders, this area of research has lagged behind the study of comorbidity in other populations (Matson & Rivet, 2008). Fortunately, recent trends in the literature suggest that individuals with ID who present with a dual disorder have been receiving increased amounts of empirical attention (Bakken et al., 2010; Kozlowski, Matson, Sipes, Hattier, & Bamburg, 2011; Matson & Boisjoli, 2009; Matson & Williams, 2014; Werner & Stawski, 2012). Evidence of this trend can be seen in the study of comorbidity in the most common conditions including autism spectrum disorders (McCarthy, 2007; Tureck, Matson, Cervantes, & Konst, 2014; Wilkins & Matson, 2009), epilepsy (Arshad et al., 2011; Bhaumik, Tyrer, McGrother, & Ganghadaran, 2008; Matson, Bamburg, & Mayville, 1999; Morgan, Baxter, & Kerr, 2003), attention-deficit/hyperactivity disorder (Bradley & Isaacs, 2006; La Malfa, Lassi, Bertelli, Pallanti, & Albertini, 2008), stereotypic movement disorder (Lee, Harrington, Chang, & Conners, 2008; Matson et al., 1997), and psychiatric disorders (Brereton, Tonge, & Einfeld, 2006; Dekker & Koot, 2003; Kolaitis, 2008; Minjarez, Phillips, Feinstein, & Hardan, 2011). This chapter will focus on the various considerations and adaptations that are required when assessing individuals with ID who also present

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with co-occurring mental, neurodevelopmental, medical, and physical conditions. In doing so, we will first provide a brief review of important diagnostic considerations that have changed how ID is conceptualized.

Diagnostic Considerations

Although the diagnostic criteria have not changed substantially over the past 50 years, the way the criteria are used to arrive at diagnostic formulation for ID has evolved in accordance with advances in the field (Brown, 2007; Schalock, Luckasson, & Shogren, 2007). These advancements have resulted in two paradigm shifts in how ID is conceptualized. These advances have moved the field away from a primary descriptive model of disability to a functionality model that can be used for diagnosis, classification, and planning supports (Luckasson & Schalock, 2013).

Within the functionality model, a multidimensional framework of human functioning is used to guide clinical judgment (Luckasson & Schalock, 2015; Schalock et al., 2010). The model adopts an ecological approach to understanding human functioning that takes into consideration intellectual abilities, adaptive behavior, health, participation, and context (Luckasson & Schalock, 2013). These five dimensions are used diagnostically (see Tassé, Luckasson, & Schalock, 2016) and for classifying individuals according to their level of independent functioning (see Schalock & Luckasson, 2015). Intellectual limitations may impact an individual's reasoning, problem-solving, planning, abstract thinking, judgment, or capacity for learning. Adaptive limitations represent an inability to meet normative expectations for independence and self-sufficiency in daily living due to deficits in conceptual, social, and practical adaptive skills. The health dimension requires an understanding of an individual's physical, mental, and social well-being. The participation and context dimensions include an understanding of an individual's interaction with their environment including the performance of social activities and factors that either facilitate or inhibit social engagement, respectively.

The second shift relates to how classification occurs. Classification provides a means for describing functional levels, operationalizing the level of supports needed, considering health factors that may be of clinical importance, and evaluating legal status (Schalock & Luckasson, 2015). With this shift, the focus has changed from the historical reliance on IQ scores for classifying an individual's level of functioning to one that is based upon the individual's adaptive behavior. This change in diagnostic nomenclature has been adopted by the American Association on Intellectual and Developmental Disabilities (AAIDD; Schalock et al., 2010) and the American Psychiatric Association (APA, 2013). The World Health Organization (WHO) has also adopted a similar classification system in the *International Classification of Diseases-11th Edition* (ICD-11; WHO, 2018) that is based on the consideration of both intellectual ability and adaptive functioning for classifying individuals. While the classification system continues to be comprised of four levels (e.g., mild, moderate, severe, and profound), the *DSM-5* now uses a descriptive approach to operationalize severity levels rather than the score ranges associated with previous IQ bands. It is also relevant to note that researchers have introduced analogous bands based upon adaptive behavior levels that may be used when considering other taxonomies, i.e., *ICD-11* (Tassé et al., 2012). 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 that form the foundation for treatment planning and progress monitoring (Horn & Fuchs, 1987). Within this comprehensive view of ID as a multifaceted construct, a nuanced appreciation of the role dual disorders may play in diagnosis, and treatment is warranted. Therefore, a brief review of the literature will be followed by an introduction to the primary constructs required for the diagnoses of ID that will

highlight diagnostic and clinical implications associated with selecting and interpreting these instruments for use with individuals who have dual disorders.

Comorbidity

As noted previously, comorbidity is common among individuals with ID. Consequently, an understanding of how comorbidity impacts diagnoses and treatment is critical for any practitioner working with this population. While a comprehensive review of all forms of comorbidity is beyond the scope of the present chapter, an overview of a few fundamental clinical considerations is necessary. The central clinical issue facing practitioners relates to symptom overlap between common co-occurring conditions and ID that make differential diagnosis and treatment planning particularly challenging (Tureck et al., 2014). For example, autism spectrum disorders (ASD) have received considerable attention for its diagnostic overlap with ID (Bamburg, Cherry, Matson, & Penn, 2001; LoVullo & Matson, 2009; Matson & Shoemaker, 2009; Smith & Matson, 2010; Wilkins & Matson, 2009). As a practitioner faced with the complex task of making an initial diagnosis, the clinical presentation of social skill deficits, stereotypic behaviors, and adaptive skill deficits is common in both disorders. Differential diagnosis between these two conditions often becomes even more complex when evaluating young children whose language ability is only beginning to emerge. In these clinically ambiguous situations, practitioners must resist the temptation to succumb to the tendency to consider all abnormal behavior observed in individuals with intellectual deficits as a manifestation of their cognitive limitations, which is a bias often referred to as diagnostic overshadowing (Matson & Scior, 2004). The clinical acumen required to render sound decisions in these situations must be grounded in the empirical literature concerning dual disorders. Within this body of research, a few emerging trends need to be addressed due to their clinical relevance to the assessment and diagnosis of ID.

First, the severity of the intellectual deficit plays an important role in understanding vulnerability to other comorbid conditions. In general, a negative correlation has been observed between the severity of cognitive deficit and the prevalence of comorbidities (Minjarez et al., 2011; O'Brien & Pearson, 2004; Tureck et al., 2014). In other words, the lower the intelligence quotient (IQ), the greater the prevalence rate for co-occurring symptoms. This general observation has been noted among various comorbid disorders including ASD (Matson & Shoemaker, 2009) and psychiatric disorders (Dekker & Koot, 2003; Turygin, Matson, & Adams, 2014). Also, research has noted that children and adolescents with moderate and profound ID frequently suffer from higher prevalence rates of co-occurring physical and/or neurological handicaps that impact their language, motor, hearing, and vision (Minjarez et al., 2011). Lastly, a lower IQ has also been suggested to be a predictor of poorer prognosis and response to intervention (Ben Itzchack, Lahat, Burgin, & Zachor, 2008).

The second empirical trend relates to the relationship between comorbidity and impairment. There is a positive correlation that has been observed between the number of comorbidities and the pervasiveness of the limitations on the individual's functional independence (Dekker & Koot, 2003; Matson et al., 1999; Smith & Matson, 2010). Some researchers have attempted to explain this correlation by suggesting that having multiple comorbid disorders exacerbates functional limitations for individuals with ID by making their condition more severe (Turygin, Matson, Adams, & Williams, 2014). However, these differences are noted both in terms of the severity of core symptoms as well as the increased prevalence rates for other co-occurring problems, e.g., stereotypies and self-injury (Matson, Dempsey, & Fodstad, 2009; Matson, Rivet, Fodstad, Dempsey, & Boisjoli, 2009; Munson et al., 2008). For example, research findings have suggested that individuals with co-occurring ASD and ID have more diverse behavioral challenges and skill deficits by comparison to counterparts who present with only one of these conditions (Boucher, Bigham, Mayes, &

Muskett, 2008). Likewise, Hahn, Brady, Warren, and Fleming (2015) noted that comorbidity played a role in predicting the developmental trajectories of individuals with ID who were able to achieve greater functional independence.

Thirdly, there is preliminary research to suggest that understanding the type of comorbidity may be useful for guiding diagnosis and treatment efforts. While this line of research has typically focused on between group differences based on comorbidity, research has also included attempts to identify symptom clusters that may be more prevalent in this population (Tremblay, Richer, Lachance, & Cote, 2010; Turygin, Matson, Adams, & Williams, 2014). For example, Matson, Mayville, Lott, Bielecki, and Logan (2003) investigated group differences in adaptive behavior among individuals who were diagnosed with ID and either ASD or psychosis. In this study, the group with a comorbid diagnosis of ASD demonstrated more significant deficits in social and adaptive behavior by comparison to those who presented with comorbid psychosis. In a study by Kozlowski et al. (2011), the authors found significant correlations among psychopathology symptom clusters in a sample of individuals diagnosed with ID. Among these individuals, the most commonly occurring were mood, mania, and anxiety symptom clusters. However, not all studies have demonstrated higher prevalence rates for comorbid symptoms for individuals with dual disorders. For example, Arshad et al. (2011) study of co-occurring ID and epilepsy failed to demonstrate greater vulnerability for psychiatric symptoms.

Taken together, while this area of research remains unclear, these emergent trends underscore the importance of considering comorbid conditions as part of routine assessment practices of practitioners working with individuals who present with ID. More importantly, the impact that dual disorders have on the developmental trajectories and ultimate functional independence of these individuals underscores the importance of incorporating treatment planning efforts to address comorbidities. In reviewing available assessments, we will provide a brief overview of the primary constructs that form the basis for the diagnosis of ID followed by a discussion of the various considerations and adaptations that may be warranted when assessing individuals with dual disorders who present with language, motor, sensory, and social, emotional, or behavioral challenges.

Intellectual Functioning

While no universally agreed-upon definition for intelligence exists, the Cattell-Horn-Carroll (CHC) framework 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. Based upon continued factor analytic research, the framework has continued to evolve and command the attention of researchers and test publishers since 2000 (McGrew, 2005). 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).

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 youth and adult populations. Diagnostic and clinical implications associated with selecting and interpreting these instruments for their clinical use will also be explored.

Intellectual Assessments

Normed-referenced intelligence instruments have been utilized over the past 100 years to assist clinicians in making diagnostic decisions regarding the presence of an ID. Of the most renowned contemporary intelligence tests, the authors have categorized these assessments under three classifications when assessing a dual diagnosis of an ID with a language, physical, sensory, social-emotional, or behavioral impairment. For this review, these classifications were created solely based on the range of modifications permitted during standardized administrative practices.

The first classification grouping includes assessment tools that permit few or limited modifications to standardized administration procedures. The Wechsler Intelligence Scale for Children, Fifth Edition (WISC-V; Wechsler, 2014), the Differential Ability Scales, Second Edition (DAS-II; Elliott, 2007a), and the Kaufman Assessment Battery for Children, Second Edition (KABC-II; Kaufman & Kaufman, 2004a), are instruments that fall under this category. These measures offer clinicians limited flexibility in their discretion to use accommodations beyond options to administer portions of the test to acquire an estimation of intellectual functioning (see Table 5.1). For example, the DAS-II's Special Nonverbal Composite is helpful in assessing estimates of intellectual functioning in children with hearing impairments. By contrast, for individuals with severe orthopedic and motor impairments, verbal and diagnostic subtests may be utilized to acquire a limited sample of an individual's cognitive capacities that serve as an approximation of intellectual ability (Elliott, 2007a). When using the Full-Scale IQ Index scores for these instruments, however, no adjustment for an individual's impairment is made, and resulting intelligence estimates represent normative comparisons to same age peers.

On the opposite end of this continuum, assessment instruments including the Stanford-Binet Intelligence Scales, Fifth Edition (SB5; Roid, 2003a) and the Woodcock-Johnson IV Tests of Cognitive Abilities (WJ IV COG; Schrank, McGrew, & Mather, 2014) permit examiners to use a wide range of manualized accommodations when assessing the intellectual ability of individuals with language, physical, sensory, social-emotional, or behavioral impairments (see Table 5.1). This second classification group allows clinicians to acquire an estimate of individuals' cognitive abilities under optimal conditions. Optimal conditions are understood to represent adaptations that intentionally mitigate the impact of individual's deficit on their performance of the task. For example, an individual who presents with a physical handicap that impacts movement may be permitted to respond orally instead of transcribing answers. It is important to note that these assessment batteries have been criticized for their flexibility in allowing the use of accommodations, which may conceal deficits as well as impact their reliability and validity (Sattler, 2007).

Finally, the last classification group covers tests that were created for use in the assessment

Table 5.1 Assessment considerations for selecting intelligence measures to identify intellectual disabilities in children, adolescents, and adults with dual diagnoses

	Normative and/or clinical sample included in manual				Suggested modifications provided in manual				
Test	LI	PI	SI	SE/BI	LI	PI	SI	SE/BI	Assessment accommodation considerations
Comprehensive Test of Nonverbal Intelligence-Second Edition (CTONI-2)			✓		✓		✓		The CTONI-2 is a "language-reduced" assessment that solely requires the examinee to point to their responses (Hammill, Pearson, & Wiederholt, 2009b, p. 1)
									Nonverbal instructions can be provided to individuals with language and hearing impairments (Hammill et al., 2009b)
									For examinees who have a severe hearing impairment or who are deaf, clinicians may use "American sign language, manually coded English, aural/oral English, or signed-supported speech" (Hammill et al., 2009b, p. 9)
									No accommodations are mentioned in the examiner's manual for individuals with severe orthopedic or motor impairments (e.g., not able to point to responses)
									The normative or clinical samples do not include individuals with visual impairments
Differential Ability Scales, Second Edition (DAS-II)	✓		✓		✓				The Special Nonverbal Composite is helpful in assessing estimates of intellectual functioning in children with hearing impairments. By contrast, for individuals with severe orthopedic and motor impairments, verbal and diagnostic subtests may be utilized to acquire an estimation of intellectual functioning (Elliott, 2007b)
									Administration instructions are provided in American Sign Language for parts of the assessment battery. Also, individuals may utilize communication modalities such as simultaneous communication, cued speech, and auditory amplification (Elliott, 2007b)
									Per the administration guidelines, pointing and gesturing are permitted; however, "these types of responses on psychological tests have been reported to be ambiguous for children who are deaf and may be a source of potential error in scoring and interpretation" (Elliott, 2007b, p. 213)
									Children with severe motor impairments may be disadvantaged on subtests that require the use of manipulatives
									In the DAS-II Introductory and Techincal Handbook, general assessment considerations per subtests are provided in Tables 9.1 and 9.2 for individuals who are deaf or hard of hearing (Elliott, 2007b)
									The examiner's manual also states that minimal modifications to standardized administration procedures are recommended to ensure reliable and valid results (Elliott, 2007b)
									The normative or clinical samples do not include individuals with visual impairments

(continued)

Table 5.1 (continued)

Test	Normative and/or clinical sample included in manual				Suggested modifications provided in manual				Assessment accommodation considerations
	LI	PI	SI	SE/BI	LI	PI	SI	SE/BI	
Kaufman Assessment Battery for Children, Second Edition (KABC-II)	✓	✓	✓	✓	✓		✓		The nonverbal scale is intended for use for children with severe speech/language and hearing impairments. All nonverbal subtests can be administered in "pantomimes such as pointing, demonstrations, and facial gestures" (Kaufman & Kaufman, 2004b, p. 27)
									The examinee may present their responses in American Sign Language for all subtests (Kaufman & Kaufman, 2004b)
									The complex and lengthy directions on the KABC-II can lead to difficulties with understanding task demands, especially for children with impaired language functioning (Flanagan & Harrison, 2005)
									The normative or clinical sample of individuals with sensory impairments only includes individuals with hearing impairments (Kaufman & Kaufman, 2004b)
Leiter International Performance Scale-Third Edition (Leiter-3)	✓	✓	✓	✓	✓	✓	✓		The Leiter-3 offers nonverbal instructions as a standardized administration procedure for all subtests on this assessment (Roid, Miller, Pomplun, & Koch, 2013b)
									Manipulatives were redesigned in the Leiter-3 to accommodate individuals with physical disabilities (Roid et al., 2013b)
									The manual discusses, in detail, modifications for examinees with significant motor or communication deficits. Specific adaptations for individuals with physical or communication disabilities include "touch/scan responses, use a stop sign, design a Yes/No response system, and use of eye gaze" (Roid et al., 2013b, p. 70–71)
									A recommended accommodation for individuals with visual impairments include the use of "color cues" to help individuals separate visual images on the response cards (Roid et al., 2013b, p. 72)
									The normative or clinical samples do not include individuals with visual impairments
Stanford-Binet Intelligence Scales, Fifth Edition (SB5)	✓	✓	✓	✓	✓	✓	✓		Verbal and nonverbal formats for each factor measuring intelligence (g) allow for examiners to discern if language, visual, or motor impairment(s) are impacting intellectual functioning (Flanagan & Harrison, 2005)
									Specific adaptations for orthopedic impairments and motor skills deficits include the use of assistive technology such as keyboards, touchpad devices, and testing trays, and vocalization amplifiers can be used during administration if needed (Roid, 2003b)
									For individuals with visual impairments, magnification devices can be utilized. In addition, "portions of the verbal routing subtest and the verbal levels could be administered orally" (Roid, 2003b, p. 116)

(continued)

Table 5.1 (continued)

Test	Normative and/or clinical sample included in manual				Suggested modifications provided in manual				Assessment accommodation considerations
	LI	PI	SI	SE/BI	LI	PI	SI	SE/BI	
									Specific guidelines are discussed in the examiner's manual (see Appendix E) for providing accommodations, such as using sign language interpreters and cued speech, to individuals with hearing impairments (Roid, 2003b)
									There are no Braille or large-print editions of the SB5 available (Roid, 2003b)
									The SB5 was last modified and re-normed in 2003. Since the inception of the SB5, the diagnostic criteria for some disabilities have considerably been modified per the *Diagnostic and Statistical Manual of Mental Disorder, Fifth Edition (DSM-5)*. Thus, the normative sample may not be representative of a current population of individuals with certain disabilities
									Flexibility with administration procedures may influence the reliability and validity of this assessment (Sattler, 2007)
Test of Nonverbal Intelligence-Fourth Edition (TONI-4)	✓	✓			✓		✓		The instructions for the TONI-4 can be administered verbally or via pantomimes for individuals with language and/or hearing impairments (Brown, Sherbenov, & Johnsen, 2010b)
									The examinees provide their responses largely motor-free through means of pointing (Brown et al., 2010b)
									There were no accommodations mentioned in the examiner's manual for individuals with significant orthopedic or motor impairments (e.g., not able to point to responses) as well as visual impairments
Universal Nonverbal Intelligence Test-Second Edition (UNIT-2)	✓		✓	✓	✓		✓		This instrument is useful to rule in or out an ID diagnosis in special populations with communication or language deficits (Bracken & McCallum, 2016b)
									The UNIT-2 assessment is also recommended for individuals who are deaf or hearing impaired, due to no verbal responses required (Flanagan & Harrison, 2005)
									Responses are limited to gestures for most subtests, which may impact children with severe motor impairment (Bracken & McCallum, 2016b)
									Cube Design subtest may be difficult for individuals with severe motor impairments to complete
									Normative or clinical sample does not include individuals with visual impairments
The Wechsler Intelligence Scale for Children, Fifth Edition (WISC-V)	✓			✓					The administration and scoring manual suggest for children with limited motor skills, use verbal subtests for estimates of cognitive ability (Wechsler, Raiford, & Holdnack, 2014). By contrast, the Nonverbal Index is useful to rule in or out an ID diagnosis in special populations such as language-impaired and autism spectrum disorder with language impairment (Wechsler et al., 2014)

(continued)

Table 5.1 (continued)

Test	Normative and/or clinical sample included in manual				Suggested modifications provided in manual				Assessment accommodation considerations
	LI	PI	SI	SE/BI	LI	PI	SI	SE/BI	
									Examinees with significant language visual, hearing, and/or motor deficits may not have the ability to perform one or more subtest(s); therefore, the examiner is not able to compute all Index and Full-Scale IQ scores for this instrument (Sattler, 2007)
									Motor demands are complex on Coding Subtest of the WISC-V, which may underestimate a child with orthopedic or motor impairment's ability to process information
									Administering only the nonverbal portion of the WISC-V assessment for individuals with visual impairments may result in over or underestimating the intellectual ability of an examinee (Flanagan & Harrison, 2005)
Woodcock-Johnson IV Tests of Cognitive Abilities (WJ IV COG)	✓			✓	✓	✓	✓	✓	The WJ-IV COG provides specific guidelines (see Table 3-2 in manual) for each test, for using communication accommodations such as American Sign Language, manually coded English, signed-supported speech, and aural/oral English (Mather & Wendling, 2014, p. 50)
									For individuals with motor impairment, examiner's may allow the examinee to type responses instead of writing answers in the response booklet. In addition, some responses may be provided via pointing or orally (Mather & Wendling, 2014)
									Accommodations for individuals with hearing impairments include the use of an interpreter, use of amplification system, administer audio-recorded test orally, and use of voice recorder (Mather & Wendling, 2014, p. 47–48)
									Accommodations for individuals with visual impairments include use of "prescribed optical devices, adaptations to materials, provide appropriate light source, if needed; provide black-lined response sheets or a black felt-tip pen instead of a pencil or enlarging print, acetate to reduce glare or increase contrast between stimulus and background, may need to mask parts of a page to reduce visual clutter; and consult with visual specialist to interpret results" (Mather & Wendling, 2014, p. 49–50)
									The WJ-IV COG provides suggested subtests (see Table 3-3 in manual) to administer to individuals with either low vision or blindness (Mather & Wendling, 2014, p. 54)
									Accommodations for individuals with attention and behavioral difficulties include informing examinee of expectation, remove distractions from testing room, and provide positive reinforcement (Mather & Wendling, 2014)
									Similar to the SB5, flexibility with the use of accommodation during the administration of the WJ-IV COG may impact the reliability and validity of this instrument

Note: LI language impairment, *PI* physical impairment, *SI* sensory impairment, *SE/BI* social-emotional or behavioral impairment

of individuals with comorbid communication disorders. Instruments that fit into this category include the Universal Nonverbal Intelligence Test-Second Edition (UNIT-2; Bracken & McCallum, 2016a), the Leiter International Performance Scale-Third Edition (Leiter-3; Roid, Miller, Pomplun, & Koch, 2013a), the Comprehensive Test of Nonverbal Intelligence-Second Edition (CTONI-2; Hammill, Pearson, & Wiederholt, 2009a), and the Test of Nonverbal Intelligence-Fourth Edition (TONI-4; Brown, Sherbenov, & Johnsen, 2010a). Collectively, the test design and administration procedures are specifically designed to remove the impact of ones' impairment through the way examiner's supply instructions as well as the manner that the examinee may respond to test items. However, these instruments also produce an estimate of intellectual functioning that is based on a limited sample of an individual's cognitive capacities by comparison to the other two classification groups. Therefore, a comprehensive assessment of individuals' overall intellectual functioning, as described by the CHC model, is not able to be achieved. In the sections that follow, we will discuss assessment considerations for individuals with a dual diagnosis based on the nature of the impairment.

Assessment Considerations for Language Impairments

When clinicians begin the process of selecting an instrument to assess intellectual functioning of their client, the first step is ensuring the individual's demographic characteristics are included in the normative or clinical sample. All intelligence measures discussed in this chapter include individuals with language impairments in either the normative or clinical sample, except for the CTONI-2 (see Table 5.1). As eluded to above, there is much more discrepancy among assessments in terms of accommodations permissible. One exception is that all tests discussed have either a composite or individuals subtest scores that measure an individual's ability through nonverbal formats. In other terms, these assessments do not require examinees to provide verbal responses. The benefit of this test design gives clinicians the option of computing individuals' estimated intellectual functioning when significant language deficits are present.

As previously mentioned, the UNIT-2, Leiter-3, CTONI-2, and TONI-4 are intended for use with individuals with communication deficits. These assessments offer nonverbal instructions as a standardized administrative procedure for all subtests. Furthermore, responses given by the examinee are provided nonverbally via gesturing or pointing (Bracken & McCallum, 2016b; Brown et al., 2010b; Hammill et al., 2009b; Roid et al., 2013b). Due to the nonverbal nature of these instruments, verbal language abilities are not measured on these instruments. Further assessments measuring verbal language constructs are typically recommended for a comprehensive evaluation of an individual's intellectual and functional capacity.

The majority of modifications to standardized procedures applied during an evaluation with individuals who present with language impairments are specific to the instrument (see Table 5.1). For example, the Leiter-3's manual discusses, in detail, adaptations for examinees with significant language deficits including "touch/scan responses, use a stop sign, design a Yes/No response system, and use of eye gaze" (Roid et al., 2013b, p. 70–71). The DAS-II manual states individuals may utilize communication modalities such as simultaneous communication, cued speech, and auditory amplification throughout this assessment. The authors of the DAS-II also recommend the use of American Sign Language for directions for portions of the assessment battery (Elliott, 2007b), whereas the KABC-II allows the examinee to present their responses in American Sign Language for all subtests (Kaufman & Kaufman, 2004b). The WJ-IV COG also provides specific instructions (see Table 3-2 in manual), for using communication accommodations such as American Sign Language, manually coded English, signed-supported speech, and aural/oral English (Mather & Wendling, 2014, p. 50). Lastly, the SB5 examiner's manual states precise guidelines for utiliz-

ing accommodations, such as using sign language interpreters and cued speech (Roid, 2003b).

Assessment Considerations for Motor or Physical Impairments

Historically, intelligence tests have placed a greater emphasis on including individuals with language or sensory impairments in normative or clinical samples and offering accommodations in comparison with individuals with physical impairments. For instance, only a few assessment batteries, such as the KABC-II, Leiter-3, TONI-4, and SB5, include individuals with orthopedic, motor, or physical impairments in the normative or clinical sample. Similar to language impairments, specific motor modifications to administration and interpretation practices range across tests (see Table 5.1). As previously mentioned, the SB5, DAS-II, and WISC-V allow clinicians to compute separate verbal composite scores and/or use diagnostic subtests to acquire an estimation of intellectual functioning (Elliott, 2007b; Roid, 2003c; Wechsler et al., 2014). Furthermore, these tests have verbal and nonverbal formats for each factor measuring intelligence, which allows examiners to discern if motor impairments may be systematically influencing intellectual functioning (Flanagan & Harrison, 2005).

Specific to the CTONI-2 and TONI-4, these assessments are designed to permit examinees to provide their responses, largely motor-free, through means of pointing for all of the subtests (Brown et al., 2010b; Hammill et al., 2009b). The Leiter-3's manual allows clinician further flexibility in providing adaptations for individuals with physical disabilities including "touch/scan responses, use a stop sign, design a Yes/No response system, and use of eye gaze" (Roid et al., 2013b, p. 70–71). It is important to note that the Leiter-3 is the only assessment that provides accommodations, i.e., eye gaze, for individuals with severe orthopedic or motor impairments. In addition, the current version of this assessment modified the design of the manipulatives with the intent to let testing materials be more accessible for individuals with physical disabilities (Roid et al., 2013b). By contrast, children with severe motor impairments may be disadvantaged on certain subtests on the DAS-II, WISC-V, and UNIT-2 due to the complex manipulatives used. In addition, specific adaptations for orthopedic impairments and motor skills deficits on the SB5 include the use of assistive technology such as keyboards, touchpad devices, testing trays, and vocalization amplifiers (Roid, 2003b). For the WJ-IV COG, the examiner may allow the examinee to type responses instead of writing answers in the response booklet. Also, some responses may be provided orally or via pointing (Mather & Wendling, 2014).

Assessment Considerations for Sensory Impairments

In this section, sensory impairments will pertain strictly to visual and hearing deficiencies. Individuals with hearing deficits are more commonly represented in the normative or clinical sample of instruments compared to individuals with visual impairments. Notably, the CTONI-2, DAS-II, KABC-II, Leiter-3, UNIT-2, and SB5 include individuals with hearing impairments in their normative or clinical sample (Bracken & McCallum, 2016b; Elliott, 2007b; Hammill et al., 2009a, 2009b; Kaufman & Kaufman, 2004b; Roid, 2003b; Roid et al., 2013b). In comparison, individuals with visual deficits are not included in the normative or clinical sample of any of the normed-reference intelligence assessments discussed in this chapter (see Table 5.1).

Accommodations recommended in the examiner's manual for assessments discussed in this chapter vary based on instrument and type of sensory impairment (see Table 5.1). For individuals with hearing impairments, the CTONI-2, Leiter-3, UNIT-2, and TONI-4 can be administered with nonverbal instructions (Bracken & McCallum, 2016b; Brown et al., 2010b; Hammill et al., 2009a, 2009b; Roid et al., 2013b). Specific to the CTONI-2, clinicians may use "American sign language, manu-

ally coded English, aural/oral English, or signed supported speech" for examinees who are deaf or have a severe hearing impairment (Hammill et al., 2009b, p. 9). Also, guidelines in the SB5 are discussed in the examiner's manual (see Appendix E) for providing accommodations, such as the use of sign language interpreters and cued speech for individuals with hearing deficits (Roid, 2003b, p. 311–322). Likewise, the examiner's manual for the DAS-II provides general assessment considerations per subtest for individuals who are deaf or hard of hearing (Elliott, 2007b). The WJ- IV COG manual lists an overabundance of accommodations for individuals with sensory impairments. Specifically for individuals with hearing impairments, the clinician may use interpreters, amplification systems, administer audio-recorded test orally, and use of voice recorders (Mather & Wendling, 2014). Furthermore, Table 3-2 in the manual provides suggested accommodations per subtest when assessing an individual with a hearing impairment (Mather & Wendling, 2014, p. 50). In addition, accommodations such as American Sign Language, manually coded English, signed-supported speech, and aural/oral English may be used by the examinee.

For individuals with visual impairments, the Leiter-3 uniquely recommends the use of "color cues" to assist individuals with visual impairments in separating visual images on the response cards (Roid et al., 2013b, p. 72). The SB5 also suggests modifications including magnification devices for individuals with visual impairments as well as "portions of the verbal routing subtest and the verbal levels could be administered orally" (Roid, 2003b, p. 166). For the WJ-IV COG, accommodations for individuals with visual impairments include use of "prescribed optical devices, adaptations to materials, provide appropriate light source; if needed, provide black-lined response sheets or a black felt-tip pen instead of a pencil or enlarging print, acetate to reduce glare or increase contrast between stimulus and background, may need to mask parts of a page to reduce visual clutter; and consult with visual specialist to interpret results" (Mather & Wendling, 2014, p. 49–50). Lastly, the WJ-IV COG provides suggested subtests (see Table 3-2 in manual) to be administered to individuals with either low vision or blindness to acquire an estimated intellectual capacity (Mather & Wendling, 2014, p. 54).

Assessment Considerations for Social-Emotional or Behavioral Impairments

Assessment accommodations for individuals with social-emotional or behavioral impairments are absent from most intelligence assessments (see Table 5.1). Consistent among cognitive instruments are fundamental environmental considerations including creating a safe and comfortable workspace as well as eliminating distractions from the environment. Of all the instruments discussed in this book chapter, the WJ-IV COG is the only test to provide explicit accommodations for individuals with behavioral impairments. For instance, the manual states that the clinician may provide examinees with positive reinforcement if significant attention and/or behavioral difficulties are present (Mather & Wendling, 2014).

Adaptive Behavior

Adaptive behaviors form the foundation for personal independence and social competence. In the most simplistic terms, adaptive functioning is defined by the individual's interaction with their environment. As such, behaviors are deemed to be adaptive based upon the situational demands and cultural norms in the environment. For example, a behavior may be judged to be adaptive in the home environment but may be deemed inappropriate in the school environment. Further, the nature of the demands

placed upon an individual also changes based upon age. For example, adaptive behavior for a young child would include assisting caregiver with putting away their belongings, by comparison to an older school-age child who would be expected to use small electrical appliances independently. Therefore, adaptive behavior is understood to be a dynamic construct that is likely to be interpreted differently across situations and over time.

Theoretical conceptualizations of adaptive behavior have also evolved. This evolution has gone from a single, broadly defined domain to an empirically validated multifaceted construct that includes agreed-upon elements. Contemporary definitions are now understood to include conceptual, social, and practical adaptive skills that have been learned by an individual and are used in community settings in the service of performing daily tasks (Schalock et al., 2010; Tassé et al., 2016). This definition has given rise to new developments in standardized instruments used to measure adaptive behavior (Tassé et al., 2012). As with all psychological instruments, the available measures of adaptive behavior come with various psychometric strengths and weaknesses. However, unlike intellectual assessments, the assessment of adaptive behavior includes both direct and indirect assessments. The use of indirect assessment, e.g., asking teachers or parents to rate an individual's behavior, allows examiners a means for gathering data that may have been otherwise unavailable. For example, communication or cognitive deficits often make it difficult to obtain reliable information about symptoms from the individual. Therefore, a diagnosis is often made using objectively observable behaviors that can be reported by informants who have observed the individual in different settings. Given the dynamic nature of the construct and the susceptibility to bias due to cultural or environmental norms, the benefits of incorporating multi-rater methods cannot be overstated. When gathering information directly from the individual, there are a number of considerations and accommodations that can be used during the assessment. These considerations include repetitiveness in the standardization sample, reading items to examinees, use of an interview format, and use of communication devices or alternative means of communication (see Table 5.2).

Future Assessment Considerations for Dual Diagnosis

As society is becoming more technologically advanced, interests in creating digital versions of test instruments have gained momentum. The Pearson Corporation has been among the early pioneers in converting their assessment batteries to digital formats through the use of their commercial product, Q-interactive ®. Notably, the WISC-V is currently available for use via the Q-interactive platform (Daniel, 2012). Other assessment batteries, such as the National Institute of Health Toolbox Cognitive Battery (NIH-TCB; NIH, 2012), have utilized similar computer-based touchscreen technologies to assess cognitive abilities of individuals. It is evitable that this developing trend will have a significant impact on assessing ID in persons with accompanying language, physical, sensory, social-emotional, or behavioral impairment. More importantly, clinical studies of these above-mentioned assessments have yielded favorable results for assessing intellectual disabilities in special populations including fragile X syndrome and Down syndrome (Hessl et al., 2016) as well as autism spectrum disorder with accompanying language impairment (Raiford, Dorzidick, & Zhang, 2015). Potential administration benefits associated with utilizing computer-based technology are increased motivation and engagement, especially in children and adolescents with behavioral difficulties. Also, the use of digital versions of instruments will likely allow for ease of use for individuals with orthopedic and/or motor impairments. The authors have anticipated that assistive technology, i.e., switches, will be integrated into these programs in the near future to allow individuals with significant physical handicaps to more adequately be assessed.

Table 5.2 Assessment considerations for selecting adaptive functioning instruments to identify intellectual disabilities in children, adolescents, and adults with dual diagnoses

Test	Normative and/or clinical sample included in manual				Assessment considerations
	LI	PI	SI	SE/BI	
Adaptive Behavior Assessment System-Third Edition (ABAS-3)	✓	✓	✓	✓	For the ABAS-3, respondents who present with visual impairments or a reading disability may have items on the rating scale read to them. Questions may also be read to respondents in the form of an interview if warranted (Harrison & Oakland, 2015)
					Clinical studies suggested that this instrument lacks construct validity for individuals who are deaf or hard of hearing on adaptive domains and adaptive skills areas on this assessment (Harrison & Oakland, 2015)
Adaptive Behavior Scale-School: Second Edition (ABS-S:2)	✓	✓	✓	✓	The ABA-S:2 can be administered via an interview format to allow information to be collected by an informant who presents with a physical or sensory impairment (Lambert, Nihira, & Leland, 1993)
					The ABS-S:2 was last modified and re-normed in 1993. Therefore, the normative population may not be representative of the current population of individuals with specific disabilities (Lambert et al., 1993)
Scales of Independent Behavior-Revised (SIB-R)			✓	✓	For the social interaction and communication subtest of this instrument, respondents may rate the examinee's skills with the use of accommodations for communication such as American Sign Language and PEC boards (Bruininks, Woodcock, Weatherman, & Hill, 1996)
					The SIB-R can be administered via a structured interview, which allows for information to be acquired from respondents who present with severe visual and/motor impairments (Bruininks et al., 1996)
					Individuals with hearing impairments and behavior disorders were rated similarly on broad independent scores to a sample of individuals without disabilities when controlling for age and sex (Bruininks et al., 1996)
					Since the inception of the SIB-R, the diagnostic criteria for selected disabilities have considerably been modified per the *Diagnostic and Statistical Manual of Mental Disorder, Fifth Edition (DSM-5)*. Thus, the normative sample may not be representative of the current population of individuals with specific disabilities
Vineland Adaptive Behavior Scales-Third Edition (Vineland-3)	✓	✓	✓	✓	The Vineland-3 can be administered via an interview format, which allows for respondents to participate if he/she is not able to complete the rating scale independently due to a physical or visual impairment (Sparrow, Cicchetti, & Saulnier, 2016)
					Examinees that utilize "sign language or electronic communication aids should receive the same scores as would be given if the behaviors were performed by speaking" (Sparrow et al., 2016, p. 45)
					The Vineland-3 assesses for internalizing and externalizing emotional and behavioral concerns as well as provides a helpful guide to assist with intervention planning (Sparrow et al., 2016)
					For individuals with sensory or motor impairments, use of assistive technology for accommodating for reading and writing deficits should not be counted against examinees for items pertaining specifically to reading and writing. Refer to the manual for specific administration recommendations (Sparrow et al., 2016). Notably, permitting respondents to rate examinee's adaptive functioning with use of accommodations may result in overestimating the examinee's level of functional independence

Note: LI language impairment, *PI* physical impairment, *SI* sensory impairment, *SE/BI* social-emotional/behavioral impairment

Conclusion

As illustrated in this chapter, individuals with moderate and profound ID frequently suffer from higher occurrences of comorbid conditions. When assessing intellectual functioning of individuals with dual diagnoses of ID and co-occurring language, motor, hearing, vision, social-emotional, and/or behavioral deficits, the nuances associated with selecting an assessment battery become cumbersome due to the number of options available. For this population, practitioners are often challenged with the task of determining whether to choose an intelligence assessment that yields the most ecologically valid estimate of intellectual functioning or one that allows the use of permitted accommodations and/or administrative procedures that may mask co-occurring deficits. Clinicians that focus on choosing instruments that lead to diagnostic conclusions specific to the client, e.g., acquiring an unmodified representation of one's intellectual ability or evaluating the influence of accommodations on individual's intellectual functioning, will likely acquire useful information that can assist in effective treatment planning. Advancements in intelligence instruments, such as digitally constructed assessment batteries, will continue to play an instrumental role in mitigating the limitations of current traditional intelligence instruments when assessing individuals with comorbid conditions.

References

American Psychiatric Association. (2013). *Diagnostic and statistical manual for mental disorders* (5th ed.). Arlington, VA: American Psychiatric Publishing.

Arshad, S., Winterhalder, R., Underwood, L., Kelesidi, K., Chaplin, E., Kravariti, E., … Tsakanikos, E. (2011). Epilepsy and intellectual disability: Does epilepsy increase the likelihood of com-morbid psychopathology? *Research in Developmental Disabilities, 32*, 353–357.

Bakken, T. L., Helverschou, S. B., Eilertsen, D. E., Heggelund, T., Myrbakk, E., & Martinsen, H. (2010). Psychiatric disorders in adolescents and adults with autism and intellectual disability: A representative study in one county in Norway. *Research in Developmental Disabilities, 31*, 1669–1677.

Bamburg, J. W., Cherry, K. E., Matson, J. L., & Penn, D. (2001). Assessment of schizophrenia in persons with severe and profound mental retardation using the Diagnostic Assessment for the Severely Handicapped-II (DASH-II). *Journal of Developmental and Physical Disabilities, 13*(4), 319–331.

Ben Itzchack, E., Lahat, E., Burgin, R., & Zachor, A. D. (2008). Cognitive, behavior, and intervention outcome in young children with autism. *Research in Developmental Disabilities, 29*, 447–458.

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*, 986–995.

Boucher, J., Bigham, S., Mayes, A., & Muskett, T. (2008). Recognition and language in low functioning autism. *Journal of Autism and Developmental Disorders, 38*, 1259–1269.

Bracken, B. A., & McCallum, R. S. (2016a). *Universal nonverbal intelligence test* (2nd ed.). Austin, TX: Pro-Ed.

Bracken, B. A., & McCallum, R. S. (2016b). *Examiner's manual universal nonverbal intelligence test* (2nd ed.). Austin, TX: Pro-Ed.

Bradley, E. A., & Isaacs, B. J. (2006). Inattention, hyperactivity, and impulsivity in teenagers with intellectual disabilities, with and without autism. *Canadian Journal of Psychiatry, 55*, 598–606.

Brereton, V., Tonge, B. J., & Einfeld, S. L. (2006). Psychopathology in children and adolescents with autism compared to young people with intellectual disability. *Journal of Autism and Developmental Disorders, 36*, 863–870.

Brown, L. (2007). What is mean by intellectual and developmental disabilities? In I. Brown & M. Percy (Eds.), *A comprehensive guide to intellectual and developmental disabilities* (pp. 3–15). Baltimore, MD: Brookes.

Brown, L., Sherbenov, R. J., & Johnsen, S. K. (2010a). *Test of nonverbal intelligence* (4th ed.). Austin, TX: Pro-Ed.

Brown, L., Sherbenov, R. J., & Johnsen, S. K. (2010b). *Examiner's manual test of nonverbal intelligence* (4th ed.). Austin, TX: Pro-Ed.

Bruininks, R. H., Woodcock, R. W., Weatherman, R. F., & Hill, B. K. (1996). *Comprehensive manual scales of independent behavior-revised*. Itasca, IL: Riverside Publishing.

Daniel, M. H. (2012). *Equivalence of Q-interactive administered cognitive tasks: WISC-IV (Q-interactive Technical Report 2)*. Bloomington, MN: Pearson.

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*, 915–922.

Elliott, C. D. (2007a). *Differential ability scales* (2nd ed.). San Antonio, TX: Harcourt Assessment, Inc.

Elliott, C. D. (2007b). *Introductory and technical handbook differential ability scales* (2nd ed.). San Antonio, TX: Harcourt Assessment, Inc.

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., & Harrison, P. L. (2005). *Contemporary intellectual assessment* (2nd ed.). New York, NY: Guilford Press.

Flanagan, D. P., Ortiz, S. O., & Alfonso, V. C. (2013). *Essentials of cross-battery assessment* (3rd ed.). Hoboken, NJ: Wiley.

Hahn, L. J., Brady, N. C., Warren, S. F., & Fleming, K. K. (2015). Do children with Fragile X syndrome show declines or plateaus in adaptive behavior? *American Journal on Intellectual and Developmental Disabilities, 120*, 412–432.

Hammill, D. D., Pearson, N. A., & Wiederholt, J. L. (2009a). *Comprehensive test of nonverbal intelligence* (2nd ed.). Austin, TX: Pro-Ed Inc.

Hammill, D. D., Pearson, N. A., & Wiederholt, J. L. (2009b). *Examiner's manual comprehensive test of nonverbal intelligence* (2nd ed.). Austin, TX: Pro-Ed Inc.

Harrison, P. L., & Oakland, T. O. (2015). *Manual adaptive behavior assessment system* (3rd ed.). Torrance, CA: Western Psychological Services.

Hessl, D., Sansone, S. M., Berry-Kravis, E., Riley, K., Widaman, K. F., Abbeduto, L., … Gershon, R. C. (2016). The NIH toolbox cognitive battery for intellectual disabilities: Three preliminary studies and future directions. *Journal of Neurodevelopmental Disorders, 8*(35), 1–18.

Horn, E., & Fuchs, D. (1987). Using adaptive behavior in assessment and intervention: An overview. *The Journal of Special Education, 21*, 11–26.

Johnson, C. P., Walker, W. O., Palomo-Gonzalez, S. A., & Curry, C. J. (2006). Mental retardation: Diagnosis, management, and family support. *Current Problems in Pediatric and Adolescent Health Care, 36*, 126–165.

Kaufman, A. S., & Kaufman, N. L. (2004a). *Kaufman assessment battery for children* (2nd ed.). Bloomington, MN: Pearson.

Kaufman, A. S., & Kaufman, N. L. (2004b). *Manual Kaufman assessment battery for children* (2nd ed.). Bloomington, MN: Pearson.

Keith, T. Z., & Reynolds, M. R. (2010). Cattell–Horn–Carroll abilities and cognitive tests: What we've learned from 20 years of research. *Psychology in the Schools, 47*, 635–650.

Kolaitis, G. (2008). Young people with intellectual disabilities and mental health needs. *Current Opinion in Psychiatry, 21*, 469–473.

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*, 1610–1614.

La Malfa, G., Lassi, S., Bertelli, M., Pallanti, S., & Albertini, G. (2008). Detecting attention- deficit/hyperactivity disorder (ADHD) in adults with intellectual disability. The use of Connors' Adult ADHD Rating Scales (CAARS). *Research in Developmental Disabilities, 29*, 158–164.

Lambert, N., Nihira, K., & Leland, H. (1993). *Examiner's manual adaptive behavior scale-school* (2nd ed.). Austin, TX: Pro-Ed Inc.

Lee, L. C., Harrington, R. A., Chang, J. J., & Conners, S. L. (2008). Increased risk of injury in children with developmental disabilities. *Research in Developmental Disabilities, 29*, 247–255.

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.

Luckasson, R., & Schalock, R. L. (2013). Defining and applying a functionality approach to intellectual disability. *Journal of Intellectual Disability Research, 57*, 657–668.

Luckasson, R., & Schalock, R. L. (2015). Standards to guide the use of clinical judgment in the field of intellectual disability. *Intellectual and Developmental Disabilities, 53*, 240–251. https://doi.org/10.1352/1934-9556-53.3.240

Mather, N., & Wendling, B. J. (2014). *Examiner's manual Woodcock-Johnson IV tests of cognitive abilities*. Rolling Meadows, IL: Riverside.

Matson, 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*, 85–90.

Matson, J. L., Bamburg, J. W., & Mayville, E. A. (1999). Seizure disorder in people with intellectual disability: An analysis of difference in social functioning, adaptive functioning and maladaptive behaviors. *Journal of Intellectual Disability Research, 43*, 531–539.

Matson, J. L., & Boisjoli, J. A. (2009). An overview of developments in research on persons with intellectual disabilities. *Research in Developmental Disabilities, 30*, 587–591.

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*, 603–611.

Matson, J. L., Hamilton, M., Duncan, D., Bamburg, J., Smiroldo, B., Anderson, S., et al. (1997). Characteristics of stereotypic movement disorder and self-injurious behavior assessed with the Diagnostic Assessment for the Severely Handicapped (DASH-II). *Research in Developmental Disabilities, 18*, 457–469.

Matson, J. L., Mayville, E. A., Lott, J. D., Bielecki, J., & Logan, R. (2003). A comparison of social and adaptive functioning in person with psychosis, autism, and severe or profound mental retardation. *Journal of Developmental and Physical Disabilities, 15*, 57–65.

Matson, J. L., & Rivet, T. T. (2008). Characteristics of challenging behaviors in adults with autistic disorder, PDD-NOS, and intellectual disability. *Journal*

of Intellectual and Developmental Disability, 33, 323–329.

Matson, J. L., Rivet, T. T., Fodstad, J. C., Dempsey, T., & Boisjoli, J. A. (2009). Examination of adaptive behavior differences in adults with autism spectrum disorders and intellectual disability. *Research in Developmental Disabilities, 30*, 1317–1325.

Matson, J. L., & Shoemaker, M. (2009). Intellectual disability and its relationship to autism spectrum disorder. *Research in Developmental Disabilities, 30*, 1107–1114.

Matson, J. L., & Williams, L. W. (2014). The making of a field: The development of comorbid psychopathology research for persons with intellectual disabilities and autism. *Research in Developmental Disabilities, 35*, 234–238.

McCarthy, J. (2007). Children with autism spectrum disorders and intellectual disability. *Current Opinion in Psychiatry, 20*, 472–476.

McGrew, K. S. (2005). The Cattell-Horn-Carroll theory of cognitive abilities. In D. P. Flanagan & P. L. Harrison (Eds.), *Contemporary intellectual assessment* (2nd ed., pp. 136–181). New York, NY: Guildford Press.

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.

Minjarez, M., Phillips, J. M., Feinstein, C., & Hardan, A. Y. (2011). Psychiatric disorders in individuals with intellectual disabilities. In H. Steiner (Ed.), *Handbook of developmental psychiatry* (pp. 491–522). Singapore: World Scientific.

Morgan, C. L., Baxter, H., & Kerr, M. P. (2003). Prevalence of epilepsy and associated health service utilization and mortality among patients with intellectual disability. *American Journal on Mental Retardation, 108*, 293–300.

Munson, J., Dawson, G., Sterling, L., Beauchaine, T., Zhou, A., & Koehler, E. (2008). Evidence for latent classes of IQ in young children with autism spectrum disorder. *American Journal on Mental Retardation, 113*, 439–452.

National Institute of Health. (2012). *NIH toolbox administration manual.* Blueprint for neuroscience research.

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.

O'Brien, G., & Pearson, J. (2004). Autism and learning disability. *Autism, 8*, 125–140.

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.

Raiford, S. E., Dorzidick, L., & Zhang, O. (2015). *Q-interactive special group studies: The WISC-V and children with autism spectrum disorder and accompanying language impairment or attention-deficit/hyperactivity disorder (Q-interactive Technical Report 11).* Bloomington, MN: Pearson.

Roid, G. H. (2003a). *Stanford-Binet intelligence scales* (5th ed.). Itasca, IL: Riverside Publishing.

Roid, G. H. (2003b). *Examiner's manual Stanford-Binet intelligence scales* (5th ed.). Itasca, IL: Riverside Publishing.

Roid, G. H. (2003c). *Technical manual Stanford-Binet intelligence scales* (5th ed.). 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). *Manual Leiter international performance scale-third edition*. Wood Dale, IL: Stoelting Company.

Sattler, J. M. (2007). *Assessment of children: Cognitive foundations* (5th ed.). San Diego, CA: Jerome 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. (2015). A systematic approach to subgroup classification in intellectual disability. *Intellectual and Developmental Disabilities, 53*, 358–366.

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. https://doi.org/10.1352/1934-9556(2007)45[116:TROMRU]2.0.CO;2

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, NY: The Guilford Press.

Schrank, F. A., McGrew, K. S., & Mather, N. (2014). *Woodcock-Johnson IV tests of cognitive abilities*. Rolling Meadows, IL: Riverside.

Smith, K. R., & Matson, J. L. (2010). Psychopathology: Differences among adults with intellectually disabled, comorbid autism spectrum disorders and epilepsy. *Research in Developmental Disabilities, 31*, 743–749.

Sparrow, S. S., Cicchetti, D. V., & Saulnier, C. A. (2016). *Manual Vineland adaptive behavioral scales* (3rd ed.). Bloomington, MN: PsychCorp.

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.

Tassé, M. J., Schalock, R. L., Balboni, G., Bersani, H. A., Jr., 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*, 291–303.

Tremblay, K. N., Richer, L., Lachance, L., & Cote, A. (2010). Psychopathological manifestations of children with intellectual disabilities according to their cognitive and adaptive behavior profile. *Research in Developmental Disabilities, 31*, 57–69.

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*, 1766–1772.

Turygin, N. C., Matson, J. L., & Adams, H. L. (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*, 1802–1808.

Turygin, N. C., Matson, J. L., Adams, H. L., & Williams, L. W. (2014). Co-occurring disorder clusters in adults with mild and moderate intellectual disability in residential treatment settings. *Research in Developmental Disabilities, 35*, 3156–3161.

Walker, W., & Johnson, C. (2006). Mental retardation: Overview and diagnosis. *Pediatrics Review, 27*, 204–212.

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 Wechsler intelligence scale for children* (5th ed.). Bloomington, MN: Pearson.

Werner, S., & Stawski, M. (2012). Mental health: Knowledge, attitudes and training of professionals on dual diagnosis of intellectual disability and psychiatric disorder. *Journal of Intellectual Disability Research, 56*, 291–304.

Wilkins, J., & Matson, J. L. (2009). A comparison of social skills profiles in intellectually disabled adults with and without ASD. *Behavior Modification, 33*, 143–155.

World Health Organization. (2018). *The international classification of diseases* (11th ed.). Geneva, Switzerland: Author. Retrieved on November 21, 2018 from https://icd.who.int/browse11/l-m/en#/http%3a%2f%2fid.who.int%2ficd%2fentity%2f605267007

Service Systems

6

Anne M. Bowers

Introduction

People with intellectual and/or developmental disabilities (I/DD) and co-occurring mental health conditions (also known as dual diagnosis) have multidimensional care needs (Bouras & Holt, 2009). Meeting these needs may necessitate more extensive use of medical and specialty care (Stanton & Rutherford, 2006). However, underdeveloped service systems have not always been capable of providing appropriate support for individuals with a dual diagnosis (Lake, McMorris, & Lunsky, 2016). Amid these issues, knowledge about successful delivery models has emerged as a priority topic of research. This chapter describes evolving systems of care for people with I/DD, as well as challenges facing current service systems. A review of key service areas explores the care and support needs of this patient group. Finally, a review of previous literature identifies six aspects of highly effective service systems for dual diagnosis: providing a continuum of services and supports, using a person-centered approach, multidisciplinary provider teams, training and education, system integration, and program support and accountability.

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Transitions in Care Delivery

Contemporary service systems for the I/DD and dual diagnosis populations are only a relatively recent phenomenon (Davidson & O'Hara, 2007; Edwards & Lennox, 2002). Beginning at the turn of the twentieth century, the primary source of daily care and services for many individuals with disabilities in the USA were large, segregated residential care facilities (Lake et al., 2016; Nielsen, 2012). Historically, residential institutions provided inadequate, ad hoc service provision to people with disabilities with little regard for the respect, dignity, quality of life, and holistic needs of residents (Nielsen, 2012). The management of psychiatric disorders and challenging behaviors would also traditionally involve inhumane physical and pharmacological restraints. These types of practices carried on at extended-stay municipal hospitals and state-run institutions for decades (Davidson & O'Hara, 2007; Nielsen, 2012).

Segregated institutional care continued to be the accepted norm for people with disabilities until about the late 1970s (Nielsen, 2012). Around that time, more medical professionals and disability advocates began to oppose institutionalization as the de facto standard of treatment for disabled patients, mainly because of its proven repeated abuses and inherent infringement on human rights and personal autonomy (Bouras, 2016; Nielsen, 2012). Deinstitutionalization

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initiatives and the Independent Living Movement gave way to increases in the number of people with disabilities receiving care in the community (Davis, Barnhill, & Saeed, 2008; Hudson & Chan, 2002; Nielsen, 2012; White, Lloyd Simpson, Gonda, Ravesloot, & Coble, 2010). Now, more people with I/DD are living in noninstitutional environments of their choice (Bouras, 2016; Hudson & Chan, 2002). Accordingly, service systems for people with I/DD have predominantly transitioned away from custodial, medical-based models and toward less restrictive comprehensive care provision in community-based settings (Alvarez, 2016; Davidson & O'Hara, 2007).

Increased availability of local health, direct care, social, and vocational service options expanded opportunities for people with dual diagnosis to live and participate in their communities (Sheehan & Paschos, 2013). Still, early evaluations of deinstitutionalization outcomes found that unmet care needs persisted for most people with dual diagnosis (Bouras, 2016; Edwards & Lennox, 2002). The prevalence of mental health conditions among the I/DD population became more apparent as community-based disability service programs started to develop at scale. At the same time, the mental and behavioral health-care needs of the dual diagnosis group remained ill-addressed and unprioritized (Bouras, 2016; Edwards & Lennox, 2002). Research since then has revealed the difficulties faced by many people with disabilities attempting to access mental health care, as well as how mainstream services are not always able to support complex needs pertinent to I/DD (Hudson & Chan, 2002; Sullivan, Robertson, Daffern, & Thomas, 2013). Such issues reflect notable service system challenges affecting care for individuals with dual diagnosis.

Contemporary Challenges

In recent years, mental health services have evolved to better meet the needs of people with I/DD (Chaplin, Paschose, & O'Hara, 2010). Many advanced industrialized nations, such as the USA, the UK, Australia, and others, have achieved significant progress toward developing effective systems of community-based care and living for people with dual diagnosis (Edwards & Lennox, 2002; Sullivan et al., 2013). Despite these improvements, the long-standing division between the mental health sector and the disability sector complicates service delivery for this group. Questions about the efficacy of treatment provided through mainstream mental health services (as opposed to specialized care for dual diagnosis) also have concerned scholars in the field. More broadly, systemic barriers to appropriate community-based mental health services can impede access to necessary care (Dart, Gapen, & Morris, 2002).

(a) *System Fragmentation*

People with I/DD and co-occurring mental health conditions require a range of services from both the mental health and disability service sectors (Bouras & Holt, 2009; National Association of State Mental Health Program Directors (NASMHPD), 2004). In most cases, neither sector alone can fulfill all the needs of an individual with dual diagnosis (Dart et al., 2002; NASMHPD, 2004). Unfortunately, these systems have developed separately from one another in many western nations and remain poorly connected (Dart et al., 2002; Edwards & Lennox, 2002; Sullivan et al., 2013). One reason for this separation is that mental health service systems and disability service systems were structured to care for distinct population groups. People with disabilities can have substantially different needs and support requirements than those of the mental health group and vice versa (NASMHPD, 2004). Training for providers has been slow to account for patients that may require services from both sectors.

Long-standing fragmentation between the mental health and disability sectors complicates service provision for dual diagnosis (Chaplin et al., 2010; Sheehan & Paschos, 2013). The philosophical paradigms from which these systems operate can conflict with one another and have produced diverging treatment and training

approaches (Chaplin et al., 2010; NASMHPD, 2004). Table 6.1 presents some of the characteristics that distinguish the treatment-based paradigm of the mental health sector from the support-based paradigm of the disability sector (NASMHPD, 2004). Considering these differences explains how independent care models in separate service arenas may generate inconsistent treatment priorities and may fail to meet the global service and support needs of a person with dual diagnosis (Dart et al., 2002; Whittle, Fisher, Reppermund, Lenroot, & Trollor, 2018). Fragmented service provision also invites uncoordinated care delivery, which can lead to inefficient provision and worse health outcomes for people with disabilities (Bowers, Owen, & Heller, 2017).

Diverging mental health and disability systems are both a cause and consequence of a lack of interagency communication and collaboration (Bouras & Holt, 2009; Dart et al., 2002; Davis et al., 2008; NASMHPD, 2004). Limited intersystem communication among the broader health, social, and voluntary service sectors further compounds these challenges to care delivery (Bouras, 2016). Collaboration with disability agencies may be especially important in the mainstream mental health sector, so providers can meaningfully engage with service recipients who have I/DD (Edwards & Lennox, 2002; Mohr, 2000). Providers in the mainstream mental health sector should be prepared to treat and manage clients with dual diagnoses, even in places with highly developed disability service systems (Chaplin, 2009; Lake et al., 2016).

(b) *Mainstream and Specialized Services*

Mainstream mental health care refers to the broad scope of mental health services that are available to members of the general population who may or may not have disabilities (Lake et al., 2016). For example, mainstream services could include individual psychiatric consultation and medication follow-up, psychotherapy or counseling at a community-based mental health provider, or inpatient admission to a psychiatric unit at a

Table 6.1 Characteristics of service system paradigms[a]

Mental Health Sector: Treatment Approach	Disability Sector: Support Approach
Recovery Model: Many mental health systems support the core assumption that people with psychiatric disorders have a chronic medical condition which they can manage and successfully recover from with the appropriate clinical treatments and continuing care	*Social Model:* Disability is viewed as an inherent condition of living rather than as a medical disease. Many disability systems provide an array of services and supports to enable people with I/DD to live in community-based settings of their choice and to receive person-centered accommodations for participation in everyday living
Psychiatric Intervention: Mental health conditions are considered chronic diseases that are cyclical but manageable. Pharmaceuticals and other medical interventions are usually viewed as appropriate and necessary to achieve recovery	*Behavioral Supports:* Behavioral health issues and other challenges that may be related to I/DD are primarily addressed using applied behavioral support strategies that emphasize adaptive skill-building and harm reduction
Rehabilitation: Rehabilitation services help people regain skills, abilities, or knowledge that may have been compromised or lost due to an illness, injury, or psychiatric issue	*Habilitation:* Habilitation services help people with I/DD learn new skills. Habilitation services support individual functioning for activities of daily living
Consumers: Modern mental health systems view service recipients as "consumers" in the sense that they should be actively involved in developing and maintaining their treatment plan. The mental health consumer movement has focused on individual rights, including the right to refuse treatment	*Self-Advocates:* Individuals with I/DD are increasingly becoming active as "self-advocates," creating and leading disability advocacy efforts on behalf of themselves or others. This advocacy has focused on rights related to self-determination and autonomy, community integration, and better access to public spaces, services, and supports

[a]Modified from: National Association of State Mental Health Program Directors (NASMHPD 2004). *Serving individuals with co-occurring developmental disabilities and mental illnesses: Systems barriers and strategies for reform* (pp. 10). Alexandria, VA: NASMHPD. Available at https://www.nasmhpd.org/archive_publications

local general hospital (Lunsky & Weiss, 2012). Mainstream services also encompass targeted treatment programs, such as those for a particular demographic of patients (e.g., women, adolescents, etc.) or those focusing on using specific clinical techniques (e.g., cognitive-behavioral therapy, dialectical behavior therapy, etc.) (Lake et al., 2016).

For patients with I/DD, there is an inclination to use mainstream mental health services to the furthest possible extent (Bouras & Holt, 2009; Cumella, 2007). However, there has been a dispute regarding the aptness and adequacy of mainstream services for treating people with dual diagnosis (Lake et al., 2016). Mainstream mental health care has both advantages and limitations in terms of efficacy for these service users (Sullivan et al., 2013). Practically speaking, mainstream mental health care is more accessible because it has the capacity to serve a greater number of community members (including those with I/DD) compared to the number who could be adequately served by niche, specialized services (Lake et al., 2016). Using mainstream services may also reduce the stigmatization of patients with I/DD (Hemmings et al., 2009; Lake et al., 2016; Sullivan et al., 2013). On the other hand, providers from mainstream programs have reported being unprepared to care for clients with dual diagnosis because of a lack of professional training (Chaplin, 2009; Edwards & Lennox, 2002; Lake et al., 2016; Sheehan & Paschos, 2013). Additionally, some people with I/DD have mental and behavioral health needs that require intensive service provision and tailored treatments (Chaplin et al., 2010; Dart et al., 2002; Lake et al., 2016). Mainstream services are not always appropriately equipped to attend to these patients the way specialized services are.

Specialized mental health care refers to services developed specifically for patients with I/DD. Compared to mainstream services, specialized care for dual diagnosis typically takes a more interdisciplinary approach and elicits greater involvement from family members and caregivers (Lake et al., 2016). Specialized mental health services can provide more considerable expertise in disability and may also have seasoned multidisciplinary provider teams, intensive treatment resources, and discharge planning processes for people with complex needs (Bouras, 2016; Chaplin, 2009). Of course, the use of specialized care can have its drawbacks. In general, there are fewer specialist service options than there are mainstream options, so specialized care can be more challenging to locate and access (Birenbaum, 1999; Lake et al., 2016). In some cases, only a few specialized clinics or programs, if any, may be available in an entire service region (Davidson & O'Hara, 2007). This scarcity can lead to longer wait times for specialists or the turning away of patient referrals, resulting in unmet mental health-care needs (Birenbaum, 1999; Lake et al., 2016). Prior research has also found that specialist services are associated with increased I/DD-related stigma (Dart et al., 2002; Lake et al., 2016). Still, even though people with I/DD predominately use mainstream services, specialized care is a necessity for optimal service provision to the dual diagnosis group (Cumella, 2007; Sheehan & Paschos, 2013; Sullivan et al., 2013). Especially for individuals with complex needs, specialist mental health services will always have a place within the broader service continuum.

Given the more pervasive use of mainstream services, specialist expertise in dual diagnosis should be integrated with the generic mental health-care delivery channels that are regularly used by individuals with I/DD (Cumella, 2007). Specialists can consult with and support service providers so mainstream systems can productively address the mental health needs of clients with dual diagnosis (Edwards & Lennox, 2002). However, identifying the most effectual structure and degree of specialist integration remains a persistent and unresolved challenge (Cumella, 2007). In the meantime, mainstream programs must take the initiative to educate clinicians and staff about working with the I/DD population so these patients can receive quality mental health care in the community.

(c) *Systemic Barriers to Care*

Barriers to care for dual diagnosis can manifest from the division between the mental health and disability service sectors and the operational

differences between mainstream and specialized services (Davidson & O'Hara, 2007; Sullivan et al., 2013). Likewise, numerous systemic deficits are contributing to substandard mental health-care delivery for people with I/DD (Edwards & Lennox, 2002; Sullivan et al., 2013). In the USA, there is wide variation and inconsistency between locales in terms of the administration and delivery of public health-care services and disability programming (Bouras, 2016). The design and structure of service systems can directly obstruct or improve access. Location-specific differences in program design, service package offerings, staffing patterns, and funding resources all contribute to disparities of equitable service provision to people with I/DD (Davis et al., 2008). Some communities may have highly collaborative disability agencies, community mental health programs, and specialist programs, while others may have more siloed service delivery (Lunsky & Weiss, 2012). Depending on the location, there also may be differences in service eligibility qualifications for age, income level, and other factors (Whittle et al., 2018). Sometimes, program eligibility conditions may exclude clients with a dual diagnosis altogether (Lunsky & Weiss, 2012). Such administrative boundaries run the risk of creating artificial barriers and service gaps (Dart et al., 2002). In the USA, health services researchers have monitored how mandatory enrollment of Medicaid recipients with I/DD into systems of managed care can disrupt service access and reduce patient-experienced quality due to coverage restrictions and limited provider networks (Birenbaum, 1999; Bowers, Owen, & Heller, 2019). Until systemic barriers to care are remedied, unmet service needs in the dual diagnosis group will exist (Dart et al., 2002).

Key Service Areas

People with I/DD need community-based health-care and social services. Co-occurring mental health conditions may also require additional specialized treatment, including crisis intervention. Long-term care can be appropriate for some individuals who have severe conditions and complex health-care needs. These areas are vital hubs of service for the dual diagnosis population.

(a) *Community-Based and In-Home Services and Supports*

Individuals with dual diagnosis require a comprehensive spectrum of local and accessible community-based services to manage their mental health and lifestyle (Davidson & O'Hara, 2007; Edwards & Lennox, 2002; NASMHPD, 2004; Davis et al., 2008). Community-based services are local providers, programs, organizations, and agencies that deliver health-care and social services to community members who may or may not have disabilities (e.g., obtaining community-based primary care and specialist provider services at local doctor offices and private practice clinics). Community-based mental health services, like individual psychiatry or psychotherapy, can be useful for many people with dual diagnosis. However, these individuals can benefit from a range of other mainstream and specialized health-care and support options including physical therapy, occupational therapy, speech therapy, psychosocial rehabilitation, vocational skills training, transitional day programs, support groups, art therapy, music therapy, and animal therapy (Davis et al., 2008; Edwards & Lennox, 2002; Khasnabis et al., 2010). Additionally, support services in the community may consist of direct care workers, supported housing, supported employment, financial management resources, legal and advocacy services, family supports, as well as access to durable medical equipment, public transportation, and social and leisure activities. The provision of local, accessible services aligns with the disability sector's support-driven paradigm that seeks to enable persons with I/DD to thrive in community-based settings and participate in everyday living (NASMHPD, 2004).

Although many communities have a variety of programs available for people with dual diagnosis and their caregivers (Sullivan et al., 2013), some services may only be available on an outpatient basis depending on location, provider networks, and other factors. Today, health and social services are also increasingly being deliv-

ered directly in clients' homes (Davidson & O'Hara, 2007). In-home services are more flexible arrangements in which providers travel to a person's home to give clinical treatment and care. Mobile providers also educate and train caregivers on prevention and support techniques (Davis et al., 2008). In-home services are beneficial for people with disabilities who live in rural areas, have mobility impairments, lack access to transportation, or best receive support in their natural environments (Birenbaum, 1999; Slifkin, Hoag, Silberman, Felt-Lisk, & Popkin, 1998). The variable nature of mental health disorders and other social, environmental, and personal influences can create challenging dispositions that may lead to inconsistent service use by members of the dual diagnosis group. Such circumstances can be a barrier to receiving regular preventive, primary, and mental/behavioral health treatment. Home-based services and mobile providers may better support isolated and hard-to-reach patients who struggle to access care in the community.

(b) *Specialized Inpatient and Outpatient Programs*

At times, hospital care may be necessary for people with dual diagnosis or complex needs [10]. Inpatient care refers to acute medical services that can only be provided in a hospital setting. Short-term inpatient admission can be needed to receive urgent medical care or if patients require sophisticated assessment, preventive screening, diagnostic testing, or psychiatric observation (Lunsky & Weiss, 2012). Inpatients with I/DD in specialized psychiatric inpatient units usually require more intensive mental health supports than those in mainstream units (Alexander, Piachaud, Singh, 2001; Lunsky, Bradley, Durbin, & Koegl, 2008). Specialized inpatient programs for dual diagnosis focus on treating the patient's immediate medical issues and specific mental health conditions while also deliberately supporting needs related to disability. These services might be useful when a person needs intensive care for both medical and mental health-related concerns, such as stabilization following a mental health crisis event. Specialized inpatient care for dual diagnosis is most appropriate for patients with complex health profiles who might require skilled multi-provider clinical intervention, specialist psychiatric evaluation, and the close involvement of family and caregivers in discharge planning and care coordination. Research has found people with I/DD that have an autism spectrum disorder (ASD) or a mood disorder diagnosis are more likely to receive services from specialized inpatient programs (Alexander et al., 2001; Charlot et al., 2011; Lunsky et al., 2008). As previously discussed, mainstream inpatient services may only be appropriate for some people with dual diagnosis; those with a co-occurring psychotic disorder or addiction issues are likelier to use mainstream inpatient services (Lake et al., 2016; Lunsky et al., 2008; White, Lunsky, & Grieve, 2010). Compared to mainstream inpatient care, more considerable clinical improvement has been observed in specialized settings for complex patients with dual diagnosis (Lake et al., 2016). The better outcomes could be due to the enhanced access specialized programs have to providers with expertise in disability and adaptive mental/behavioral health interventions. These types of I/DD-focused resources are required less frequently in mainstream inpatient settings.

People with dual diagnosis may also receive care through outpatient services. In terms of the present discussion, outpatient care refers to hospital-based ambulatory patient services and affiliated care delivery channels that do not require formal hospital admission or overnight stays. Outpatient services can be provided inside hospital facilities or at off-site locations. Common examples of outpatient service sites are testing labs, freestanding dialysis units, and hospital network-affiliated medical, surgical, and mental health centers. Outpatient services can also include intensive outpatient programs (IOPs). IOPs involve patient attendance at the outpatient service site on multiple days per week (3–4 hours per visit) over a 1–2 time frame to receive structured treatment and other therapeutic care from an interdisciplinary team of providers. IOPs can target different segments of service users, such as those with particular psychiatric conditions (e.g.,

bipolar disorder, schizophrenia, etc.), histories of trauma, eating disorders, or substance use disorders. IOPs may be beneficial for patients transitioning out of inpatient care or for those who are working on managing symptoms to prevent a future mental health crisis or potential hospitalization (Davis et al., 2008).

There is growing evidence suggesting that specialized IOPs and psychotherapy interventions can lead to positive outcomes for people with I/DD (Lake et al., 2016; Vereenooghe & Langdon, 2013). Specialized IOPs for dual diagnosis can provide mental health services that pay special attention to disability-specific issues with specialist-driven treatment curriculums. These IOPs may focus on specific methods of clinical care and service delivery to the I/DD population, like psychopharmaceutical intervention, psychosocial rehabilitation, or intensive case management (Davidson & O'Hara, 2007). IOP programs can also target various diagnostic categories, such as children or adults with I/DD and co-occurring ASD, seizure disorders, or neurobehavioral emotion regulation issues (Lake et al., 2016). Specialized outpatient provider teams work closely with family members and caregivers on service plan development, medication management, and training for continued care. If more intensive specialized services are required, patients may be able to enroll in a partial hospitalization outpatient program that requires more frequent attendance (e.g., 5–6 days per week) and a longer time commitment than an IOP (5–7 hours per day) (Davidson & O'Hara, 2007).

(c) *Crisis Intervention Services*

The ongoing presence of numerous risk and vulnerability factors can contribute to the occurrence of mental health crises in people with dual diagnosis (Davis et al., 2008). A crisis involves the serious deterioration of an individual's ability to cope with everyday life (Lunsky & Weiss, 2012). For persons with dual diagnosis, such situations often present as urgent and adverse emotional or behavioral reactions which may (or may not) be precipitated by a triggering situation or traumatic event. A mental health crisis may also include emergencies that involve immediate danger, such as threats of physical violence, self-harm, or suicide. Even if the individual is not necessarily a danger to themselves or others during a crisis, outside support is typically still needed (Lunsky & Weiss, 2012). However, when crisis stabilization is required, individuals with I/DD may not always have prompt access to needed care like specialized inpatient programs, emergency interventions, and other transition services (Davis et al., 2008; Tang et al., 2008). Hospital emergency departments are frequently utilized for crisis stabilization. However, these mainstream facilities are typically not designed to treat the specific mental and behavioral health needs of the I/DD population (Colorado Department of Health Care Policy and Financing (HCPF), 2019; Tang et al., 2008).

Crisis intervention programs can provide services that help stabilize community members with I/DD during mental health crisis events (Davidson & O'Hara, 2007). These programs may be delivered in a variety of formats and can include both short-term inpatient and respite care units and multidisciplinary crisis intervention teams (Colorado HCPF, 2019; Davis et al., 2008; Owen, Bowers, Heller, Hsieh, & Gould, 2017). Crisis intervention provider teams can deliver community-based and in-home clinical intervention, assessment, care planning, service coordination, and prevention training to people with dual diagnosis and their caregivers (Owen et al., 2017). Some programs provide 24-hour mobile crisis response teams, allowing for appropriate care and consistent responses to be delivered no matter where or when a crisis event may happen within the program's jurisdiction (Davidson & O'Hara, 2007; Sullivan et al., 2013). Increasingly, crisis intervention programs also offer respite care in small, community-based home settings to stabilize patients safely without the need for hospital admission or institution-based residential care (Davis et al., 2008; Owen et al., 2017). Crisis intervention teams can help clients, family members, and caregivers create action plans for the prevention and management of future mental health crises. Studies of crisis intervention initiatives have found favorable outcomes such as the

decreased risk for rehospitalization, reduced service utilization, and improved care coordination for people with I/DD (Owen et al., 2017). When incorporated into a comprehensive community-based service continuum, crisis intervention enhances the timely provision of urgent care and extra support that enables individuals with dual diagnosis to remain living in noninstitutional settings (Owen et al., 2017).

(d) *Long-Term Care Services*

Long-term care services refer to intensive, institution-based residential services and care that provide 24-hour direct services and on-site treatment to residents of the location. Long-term care facilities provide consistent, active clinical care as well as medical testing and specialized treatment that cannot be reasonably rendered in a community-based setting (e.g., skilled nursing, care-intensive behavioral health monitoring, etc.) (Turygin, Matson, & Adams, 2014). Persons with dual diagnosis can get long-term care in a variety of places, like intermediate care facilities and psychiatric residential care centers. Services offered in these settings are most appropriate for patients with severe disabilities, highly complex needs, or significant challenges and maladaptive behaviors stemming from treatment-refractory mental/behavioral health disorders (Dart et al., 2002; Matson & Shoemaker, 2009). For persons with I/DD who are in the advanced stages of aging or have a serious illness, it is common to receive long-term care in nursing home facilities.

It is worth noting that current policy and regulatory initiatives have worked to improve the overall safety and quality of long-term care facilities in the USA (Brennan, 1998; Eremia, 2002; Gruneir & Mor, 2008; Park & Stearns, 2009). Even so, earnest efforts should be made to find community-based service alternatives before someone with dual diagnosis transfers indefinitely to an institutional care setting. Facilities providing long-term care services are usually very costly, restrictive, and often inappropriate for members of the I/DD population who can live at home and in the community with the right support and accommodations. The individual with dual diagnosis and their family members and caregivers should have active involvement in any decision-making and planning related to community living or long-term care arrangements.

Effective Service Systems for Dual Diagnosis

A review of contemporary issues and critical areas of care shapes our understanding of optimal service systems for people with dual diagnosis. Having a mental health condition generally begets higher need and utilization of care (Stanton & Rutherford, 2006), making knowledge about successful service provision a priority topic of investigation. Aspects of effective service provision for the dual diagnosis group have been well-examined in previous studies (Bouras & Holt, 2009; Dart et al., 2002; Davidson & O'Hara, 2007; Davis et al., 2008; Hudson & Chan, 2002). A synthesis of information reported within the collective literature structures the following discussion.

(a) *Continuum of Services and Supports*

Service systems are responsible for the inventory of care and support options available to recipients with dual diagnosis. A continuum of services and supports aims to seamlessly address comprehensive medical, direct support, community living, education, employment, social and leisure, and long-term care needs across a person's life span (Bowers, 2019; Dart et al., 2002). A continuum focusing on individuals with dual diagnosis will often encompass health and social service components for early identification and assessment, primary and preventive health care, specialized clinical intervention and treatment, and long-term supports and care (Dart et al., 2002; Davis et al., 2008; Khasnabis et al., 2010). Nonetheless, complex medical issues and severe mental/behavioral health conditions can impact the physical mobility, personal capacity, and participation of persons with I/DD (Matson & Shoemaker, 2009). The needs of such patient

cases may necessitate more intensive treatment in specialized inpatient programs or other long-term residential care settings (Davis et al., 2008). Many with dual diagnosis are best served by routinely receiving an individually tailored combination of community-based, outpatient, and in-home health and social services, with the option of using intermittent specialized inpatient and respite care services as necessary (Bouras, 2016; Bouras & Holt, 2009; Chaplin et al., 2010; Scior, 2016). At a minimum, mainstream mental health services used by people with dual diagnosis should be supported by specialist provider teams who can offer expert consultation in I/DD and coordination with the disability sector (Sheehan & Paschos, 2013; Sullivan et al., 2013).

Utilization of services and supports available along the continuum should be planned based on thorough assessments of the medical, daily living, social, and vocational needs of the person with dual diagnosis (Edwards & Lennox, 2002). The format of service delivery (community-based, in-home, outpatient, inpatient, mainstream or specialized, etc.) should be carefully considered to best address the identified patient needs. Services required by an individual with I/DD, including both the amount and type of care, will likely fluctuate over time. Services needed may also depend on the complexity of the person's overall health status, co-occurring conditions, changing needs with age, and degree of natural supports and family member involvement in service planning and delivery (Bowers, 2019). The multifaceted service needs of a person with dual diagnosis must be recurrently assessed to ensure continued provision of an appropriate, holistic continuum of care (Sullivan et al., 2013). Care coordination is another foundational aspect of adequate planning, organization, and management of services for people with disabilities (Bowers et al., 2017). High-need patient groups require robust and responsive delivery systems offering a spectrum of specialized care and community-based supports that are organized and accessible to users (Bowers et al., 2017; Dart et al., 2002).

(b) *Person-Centered Approach*

A person-centered care approach provides medical care and community-based services in a way that is directly informed by a person's expressed preferences, specific needs, and social circumstances (Kitson, Marshall, Bassett, & Zeitz, 2013). Unlike standardized delivery models, person-centered models situate the service recipient as a central determinant of how care can best be delivered and managed. A person-centered approach focuses on providing the appropriate types of services and treatment based on an individual's identified health service needs, medical history, and personal preferences and wishes for care (Bowers et al., 2017). Patients are considered equal partners in the creation, management, and monitoring of their care and treatment plan in these models (Bowers et al., 2017). Person-centered planning and provision also gives due consideration to the person's environmental contexts (e.g., living situation, location, etc.) and existing natural sources of support (e.g., parents, siblings, caregivers, advocates, etc.) (Hudson & Chan, 2002; Sullivan et al., 2013). Person-centered approaches actively facilitate involvement from the individual and their support sources in service planning and decision-making about current treatment and future care (Bowers et al., 2017; Cumella, 2007; Troller, 2014).

The various care needs of people with I/DD and co-occurring mental health conditions raise their likelihood of experiencing a wide range of services and treatment modalities across a variety of settings. Person-centered initiatives might be particularly beneficial in this sense, as the dual diagnosis group needs more extensive care services but may have limited agency within broader mainstream health systems (Stanton & Rutherford, 2006). The participation of service users, family members, and caregivers in person-centered approaches is essential to make sure appropriate treatment priorities get established and that services are delivered satisfactorily (Bouras, 2016). Person-centeredness can play a valuable role in service planning, delivery, and

coordination processes for people with I/DD and dual diagnosis (Bowers et al., 2017).

(c) *Multidisciplinary Provider Teams*

Service provision via a multidisciplinary team is essential for individuals with dual diagnosis (Davidson & O'Hara, 2007; Davis et al., 2008; Hudson & Chan, 2002; Troller, 2014). A multidisciplinary approach refers to when professionals with distinct discipline-specific knowledge and training come together to proactively deliver a continuum of care and forge mutually supportive relationships with one another to ensure comprehensive service provision to a person with I/DD (Davidson & O'Hara, 2007; Edwards & Lennox, 2002; Lunsky & Weiss, 2012). No single professional group has a monopoly of knowledge in dual diagnosis, which is one reason why collective input from all associated service professionals is a best practice for optimal service provision (Bouras & Holt, 2009; Hudson & Chan, 2002; Lunsky & Weiss, 2012).

A multidisciplinary approach is the preferred model of care for working with people with dual diagnosis and their families and caregivers. In this method, the skills and experience of different clinicians and health professionals are matched to the unique needs of the client (Bouras, 2016; Davis et al., 2008; Edwards & Lennox, 2002; McCallin, 2001). Identified members of the multidisciplinary provider team deliver a range of mainstream and specialized services and treatment based on a shared understanding of the person-centered care plan. Provider teams for dual diagnosis may include community psychiatrists and psychologists, mental and behavioral health practitioners, nurses, psychosocial rehabilitation providers, physical/occupational/speech therapists, in-home habilitation and respite service providers, social workers, care coordinators, and others from the broader health and disability service sectors (Bouras & Holt, 2009; Edwards & Lennox, 2002; Khasnabis et al., 2010).

One advantage of this collaborative team-based model is that it allows providers to support and build on the collective strengths of the client's formal and informal care network (Dart et al., 2002). For the dual diagnosis group, a multidisciplinary model can also be helpful for crisis prevention. Members of the provider team can act as patient advocates during urgent health events (e.g., emergency medical situations, mental health crises, etc.) or hospitalization (Owen et al., 2017). Members of the multidisciplinary provider team should regularly engage the client's caregivers in training activities to ensure consistency in the delivery of care plan directives and preventive strategies (Dart et al., 2002; Davidson & O'Hara, 2007).

(d) *Training and Education*

Effective service systems are dependent on the availability of competent mental health service providers who can successfully care for people with I/DD (Davidson & O'Hara, 2007). Thus, education and training initiatives are crucial for mental health service provision to the dual diagnosis group (Bouras & Holt, 2009; Sullivan et al., 2013; Troller, 2014). Expertise in dual diagnosis is growing, but more progress in the dissemination of knowledge about caring for these patients is needed (Edwards & Lennox, 2002). Many medical professionals receive minimal training about I/DD. Provider competency in dual diagnosis needs to be improved to enhance patient-provider communication and quality service delivery. Disability service professionals should receive training as well to better prepare for these clients who may need additional programs and forms of care. All relevant providers should obtain knowledge and experience in both the disability and mental health sectors in order to support cross-system access (Davidson & O'Hara, 2007; Edwards & Lennox, 2002; Troller, 2014). Service systems could consider incentivizing clinician education in dual diagnosis to promote provider retention and specialization in this underserved area (Davidson & O'Hara, 2007).

Disseminating educational information should not stop at the service provider level. Family members and caregivers of people with dual diagnosis also need more opportunities for education and training (Sullivan et al., 2013). As ser-

vice systems transition to community-based delivery models, direct care and support from family members and in-home caregivers is ever more crucial. These daily sources of support for people with I/DD must get training about appropriate care and prevention techniques to supplement provision from community services. Such efforts are vital so people with dual diagnosis can receive reliable continuing care regimens and remain safely living at home. Collaborative education and programming opportunities among the multiple providers, caregivers, agencies, stakeholders, and systems that provide services for these individuals should be supported and encouraged to promote responsive care delivery and integrated service provision (Colorado HCPF, 2019; Lunsky & Weiss, 2012).

(e) *System Integration*

Numerous systemic barriers have challenged comprehensive service delivery to people with I/DD and co-occurring mental health conditions. An ideal system for dual diagnosis would eliminate all barriers to receiving services to create seamless, optimized provision through identified care pathways (Dart et al., 2002; Troller, 2014). Since treating dual diagnosis requires services from multiple domains, strong coordinated linkages between disability services, mental health-care providers, and other health and social services are important for effective delivery (Colorado HCPF, 2019; Sullivan et al., 2013). Even better access to a blend of clinical expertise and resources may be possible with enhanced cross-system access and system integration (Davidson & O'Hara, 2007).

System integration is a step beyond interagency coordination and collaboration. Integrated models can enable the mental health and disability sectors to come together for streamlined provision, management, and financing of a range of mainstream and specialized services for dual diagnosis (Bowers, 2019; Lake et al., 2016). System integration has the potential to link mental health and disability services in ways that complement the strengths of each sector's approach (Davidson & O'Hara, 2007). Their combined array of services could be further integrated with the primary health-care system to promote comprehensive delivery (Edwards & Lennox, 2002). System integration also creates a centralized point of contact, planning, and coordination for service users. Enhanced service accessibility and coordination from centralized administration and streamlined delivery structures can help to reduce gaps in needed care (Bowers, 2019). System integration may be the most pragmatic way to develop a robust continuum of services and supports that are readily available and fully accessible to all community members with I/DD (Dart et al., 2002).

(f) *Program Support and Accountability*

The successful provision of services to people with dual diagnosis requires community-based agencies and programs to have adequate resources (Bouras & Holt, 2009; Davidson & O'Hara, 2007; Davis et al., 2008). In the USA, many community-based services and supports are managed by not-for-profit (NFP) organizations that do not receive direct funding. NFP program funding is often contingent upon multiple sources, including a government-allocated budget that can change from one fiscal period to the next (Dart et al., 2002). Community-based service programs for people with I/DD must have reliable financial support to pay for sufficient staffing, quality programming, and overhead costs. If the funding received is not enough to support efficient program operation, service quality will decrease to the detriment of users.

Quality services are only reliable to the extent programs are held accountable for their performance. Accountable programs are essential for service users with dual diagnosis who may engage with a plethora of providers across various care settings. Programs can demonstrate accountability by measuring their service impacts and outcomes (Bouras & Holt, 2009). This evaluation involves the deliberate collection of data and the regular assessment of service utilization and client reported experiences. Programs should routinely report the results of their internal evaluations to relevant stakeholders (Troller, 2014).

From there, programs may build credibility through regular transparent reporting and quality improvement efforts. For example, programs can implement structured grievance and appeal processes, as well as a designated ombudsman, to urge consistent delivery of quality person-centered care and support. Service delivery organizations in the USA can also partner with a member of the Association of University Centers on Disabilities (AUCD) to receive technical assistance and expert training. Members of the AUCD's nationwide network provide evidence-based resources and knowledge sharing among local agencies to support enhanced service provision to the disability community (AUCD, 2018).

Conclusion

In the aftermath of deinstitutionalization, the untreated mental health needs of people with I/DD became more apparent (Edwards & Lennox, 2002). An increasing number of people with disabilities are living and receiving services in community-based settings, but barriers to care still exist for the dual diagnosis population (Davidson & O'Hara, 2007). Long-standing divisions between the disability and mental health service sectors have allowed for the fragmented provision of care. Mainstream mental health services are not always able to deliver appropriate treatment for dual diagnosis, but specialized services can be scarce and difficult to access. Moreover, many health-care professionals have limited education in I/DD and may be unprepared to treat this patient group. Administrative obstacles imposed by health plans and broader service policy can also impede access to care. Community-based programs that lack funding and accountability will inhibit further progress in this area. More robust, knowledgeable provider networks are needed, as well as streamlined delivery structures through system integration (Bowers et al., 2019). Without access to necessary mental health-care and disability services, people with I/DD risk re-institutionalization and worse health outcomes (Bouras, 2016).

Fortunately, contemporary service systems started to become more advanced around the beginning of the twenty-first century. In many places, community members with dual diagnosis now receive a spectrum of services and supports in a variety of settings to meet their distinct needs. Flexible arrangements, like home-based care and crisis intervention, have been significant developments in service provision. Collaborative, multidisciplinary provider teams are recognized as integral to the ongoing delivery of comprehensive care for this patient group (Bouras, 2016). Providing specialized inpatient, outpatient, and long-term service options is essential for those who require intensive treatment. Facilitating the involvement of patients and their family members and caregivers also enhances the person-centeredness of care (Bowers et al., 2017). These strides in service delivery have improved care for the dual diagnosis population and lay the foundation for further innovation in the field. Nevertheless, more work remains to be done to achieve highly effective service systems for people with I/DD.

References

Alexander, R. T., Piachaud, J., & Singh, I. (2001). Two districts, two models: In-patient care in the psychiatry of learning disability. *British Journal of Developmental Disabilities, 47*, 105–110.

Alvarez, N. (2016). Large Residential Care Facilities. In *Health care for people with intellectual and developmental disabilities across the lifespan* (pp. 265–275). Cham, Switzerland: Springer.

Association of University Centers on Disabilities (AUCD). (2018). *2018 AUCD annual report*. Silver Spring, MD: AUCD. Retrieved from https://aucdannualreport.org/2018-2/

Birenbaum, A. (1999). *Disability and managed care: Problems and opportunities at the end of the century*. Westport, CT: Greenwood Publishing Group.

Bouras, N. (2016). Historical and international perspectives of services. In C. Hemmings & N. Bouras (Eds.), *Psychiatric and behavioural disorders in intellectual and developmental disabilities* (3rd ed., pp. 1–14). Cambridge: Cambridge University Press.

Bouras, N., & Holt, G. (2009). The planning and provision of psychiatric services for adults with intellectual disability. In M. Gelder, N. Andreasen, J. Lopez-Ibor, & J. Geddes (Eds.), *New Oxford textbook of psychiatry*

(2nd ed., pp. 1887–1894). Oxford: Oxford University Press.

Bowers, A. (2019). Service delivery models. In J. L. Matson (Ed.), *Handbook of intellectual disabilities: Integrating theory, research and practice* (pp. 109–119). Cham, Switzerland: Springer. https://doi.org/10.1007/978-3-030-20843-1_7

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.

Bowers, A., Owen, R., & Heller, T. (2019). Managed care experiences of Medicaid enrollees with disabilities: A qualitative analysis of consumer survey responses. *Journal of Health Care for the Poor and Underserved, 30*(3), 968–985.

Brennan, T. A. (1998). The role of regulation in quality improvement. *The Milbank Quarterly, 76*(4), 709–731.

Chaplin, E., Paschos, D., & O'Hara, J. (2010). The specialist mental health model and other services in a changing environment. In N. Bouras & G. Holt (Eds.), *Mental health services for adults with intellectual disability: Strategies and solutions* (pp. 9–22). London,UK: Psychology Press.

Chaplin, R. (2009). New research into general psychiatry services for adults with intellectual disability and mental illness. *Journal of Intellectual Disability Research, 53*, 189–199.

Charlot, L., 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.

Colorado Department of Health Care Policy and Financing (HCPF). (2019). *FY 2018–19 cross-system crisis response pilot legislative report*. Denver, CO: HCPF. Retrieved from https://www.colorado.gov/hcpf/legislator-resource-center

Cumella, S. (2007). Mental health and intellectual disabilities: The development of services. In N. Bouras & G. Holt (Eds.), *Psychiatric and behavioural disorders in intellectual and developmental disabilities* (2nd ed., pp. 353–363). Cambridge: Cambridge University Press.

Dart, L., Gapen, B., & Morris, S. (2002). Building responsive service systems. In D. Griffiths, C. Stavrakaki, & J. Summers (Eds.), *Dual diagnosis: An introduction to the mental health needs of persons with developmental disabilities* (pp. 283–324). Sudbury, ON: Habilitative Mental Health Resource Network.

Davidson, P. W., & O'Hara, J. (2007). Clinical services for people with intellectual disabilities and psychiatric or severe behaviour disorders. In N. Bouras & G. Holt (Eds.), *Psychiatric and behavioural disorders in intellectual and developmental disabilities* (2nd ed., pp. 364–387). Cambridge: Cambridge University Press.

Davis, E., Barnhill, L. J., & Saeed, S. A. (2008). Treatment models for treating patients with combined mental illness and developmental disability. *Psychiatric Quarterly, 79*(3), 205–223.

Edwards, N., & Lennox, N. G. (2002). *Not on the same page: Report submitted to Queensland health on dual diagnosis needs assessment project*. Brisbane, QLD: Developmental Disability Unit, University of Queensland. Retrieved from https://espace.library.uq.edu.au/view/UQ:84184

Eremia, A. D. (2002). When self-regulation, market forces, and private legal actions fail: Appropriate government regulation and oversight is necessary to ensure minimum standards of quality in long-term health care. *Annals of Health Law, 11*(1), 93–124.

Gruneir, A., & Mor, V. (2008). Nursing home safety: Current issues and barriers to improvement. *Annual Review of Public Health, 29*, 369–382.

Hemmings, C. P., O'Hara, J., McCarthy, J., Holt, G., Eoster, F., Costello, H., … Bouras, N. (2009). Comparison of adults with intellectual disabilities and mental health problems admitted to specialist and generic inpatient units. *British Journal of Learning Disabilities, 37*(2), 123–128.

Hudson, C., & Chan, J. (2002). Individuals with intellectual disability and mental illness: A literature review. *Australian Journal of Social Issues, 37*(1), 31–49.

Khasnabis, C., Motsch, K. H., Achu, K., Al Jubah, K., Brodtkorb, S., Chervin, P., … Goerdt, A. (2010). *Community-based rehabilitation: CBR guidelines*. Geneva, Switzerland: World Health Organization. Retrieved from https://www.ncbi.nlm.nih.gov/pubmed/26290927

Kitson, A., Marshall, A., Bassett, K., & Zeitz, K. (2013). What are the core elements of patient-centred care? A narrative review and synthesis of the literature from health policy, medicine and nursing. *Journal of Advanced Nursing, 69*(1), 4–15.

Lake, J., McMorris, C., & Lunsky, Y. (2016). Specialized and mainstream mental health services. In C. Hemmings & N. Bouras (Eds.), *Psychiatric and behavioural disorders in intellectual and developmental disabilities* (3rd ed., pp. 252–261). Cambridge: Cambridge University Press.

Lunsky, Y., Bradley, E., Durbin, J., & Koegl, C. (2008). A comparison of patients with intellectual disability receiving specialized and general services in Ontario's psychiatric hospitals. *Journal of Intellectual Disability Research, 52*, 1003–1012.

Lunsky, Y., & Weiss, J. (2012). *Dual diagnosis: An information guide*. Toronto, ON: Centre for Addiction and Mental Health. Retrieved from https://www.camh.ca/en/health-info/

Matson, J. L., & Shoemaker, M. (2009). Intellectual disability and its relationship to autism spectrum disorders. *Research in Developmental Disabilities, 30*(6), 1107–1114.

McCallin, A. (2001). Interdisciplinary practice – a matter of teamwork: An integrated literature review. *Journal of Clinical Nursing, 10*(4), 419–428.

Mohr, C. (2000). Collaboration – Together we can find the way in dual diagnosis. *Australian and New Zealand Journal of Psychiatry, 34*, A46–A46.

National Association of State Mental Health Program Directors (NASMHPD). (2004). *Serving individuals with co-occurring developmental disabilities and mental illnesses: Systems barriers and strategies for reform*. Alexandria, VA: NASMHPD. Retrieved from https://www.nasmhpd.org/archive_publications

Nielsen, K. E. (2012). *A disability history of the United States*. Boston, MA: Beacon Press.

Owen, R., Bowers, A., Heller, T., Hsieh, K., & Gould, R. (2017). The impact of support services teams: Community-based behavioral health support interventions. *Journal of Policy and Practice in Intellectual Disabilities, 14*(3), 205–213.

Park, J., & Stearns, S. C. (2009). Effects of state minimum staffing standards on nursing home staffing and quality of care. *Health Services Research, 44*(1), 56–78.

Scior, K. (2016). Service users' and carers' experiences of mental health services. In C. Hemmings & N. Bouras (Eds.), *Psychiatric and behavioural disorders in intellectual and developmental disabilities* (3rd ed., pp. 262–268). Cambridge: Cambridge University Press.

Sheehan, R., & Paschos, D. (2013). A comparison of different models to meet the mental health needs of adults with intellectual disabilities. *Advances in Mental Health and Intellectual Disabilities, 7*(3), 161–168.

Slifkin, R. T., Hoag, S. D., Silberman, P., Felt-Lisk, S., & Popkin, B. (1998). Medicaid managed care programs in rural areas: A fifty-state overview. *Health Affairs, 17*(6), 217–227.

Stanton, M. W., & Rutherford, M. K. (2006, June). The high concentration of U.S. health care expenditures: Research in action, issue 19 (Publication No. 06–0060). Rockville, MD: Agency for Healthcare Research and Quality. Retrieved from https://archive.ahrq.gov/research/findings/factsheets/costs/expriach/.

Sullivan, D., Robertson, T., Daffern, M., & Thomas, S. (2013). *Building capacity to assist adult dual disability clients' access effective mental health services*. Melbourne, VIC: Monash University Centre for Forensic Behavioural Science and the State of Victoria Department of Human Services. Retrieved from https://services.dhhs.vic.gov.au/

Tang, B., Byrne, C., Friedlander, R., McKibbin, D., Riley, M., & Thibeault, A. (2008). The other dual diagnosis: Developmental disability and mental health disorders. *BC Medical Journal, 50*, 319–324.

Trollor, J. (2014). Making mental health services accessible to people with an intellectual disability. *Australian & New Zealand Journal of Psychiatry, 48*(5), 395–398.

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.

Vereenooghe, L., & Langdon, P. E. (2013). Psychological therapies for people with intellectual disabilities: A systematic review and meta-analysis. *Research in Developmental Disabilities, 34*, 4085–4102.

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.

White, S. E., Lunsky, Y., & Grieve, C. (2010). Profiles of patients with intellectual disability and mental illness in specialized and generic units in an Ontario psychiatric hospital. *Journal of Mental Health Research, 3*, 117–131.

Whittle, E. L., Fisher, K. R., Reppermund, S., Lenroot, R., & Trollor, J. (2018). Barriers and enablers to accessing mental health services for people with intellectual disability: A scoping review. *Journal of Mental Health Research in Intellectual Disabilities, 11*(1), 69–102.

Educational Models

7

Hsu-Min Chiang

Introduction

Students with a disability in the United States are entitled to receive free and appropriate education and related services under the Individuals with Disability Education Act (IDEA). The IDEA governs how states and public agencies offer special education and related services to individuals with disabilities from birth to age 21 (Office of Special Education Programs, 2018). To be qualified for the IDEA services, a student must have a disability listed under the IDEA and have the needs of receiving special education. The disabilities listed under the IDEA include intellectual disability (ID), hearing impairment (including deafness), speech or language impairment, visual impairment (including blindness), emotional disturbance (ED), orthopedic impairment, autism, traumatic brain injury, other health impairment (including attention deficit disorder, attention deficit hyperactivity disorder), a specific learning disability, deaf-blindness, and multiple disabilities (concomitant impairments). Among these disabilities, five disabilities, autism, ED, ID, multiple disabilities, and other health impairment, are within the scope of this book. Thus, the educational models discussed in this chapter are the ones specifically for the students with the five disabilities.

Educational Placements for Students with Disabilities

The IDEA requires schools to provide an individualized education program (IEP) to each student with a disability. The special education and related services and supplementary aids to be provided to a student with a disability are required by the IDEA to be clearly stated in a student's IEP as well as the educational placement where a student with a disability is to receive these services. A continuum of alternative educational placements in the United States is available to meet the diverse needs of students with disabilities. These placements include regular classes, special classes, special schools, home instruction, and instruction in hospitals and institutions (IDEA, 2004). To the maximum extent possible, a student with a disability should be educated with his/her peers without disabilities and provided with access to the general education curriculum (IDEA, 2004). Thus, it is common to see that supplementary services (such as resource room or itinerant instruction) and other related services and supports are provided to the students who are placed in a regular classroom. Also, the opportunities to receive instruction alongside peers without disabilities are provided to the students who are placed in a

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special education classroom or special school. The IEP team makes the placement decision and determines the educational and related services to be provided to a student with a disability based on the student's needs.

Educational Models in School Settings

The interventions to address students' behavior problems and mental health disorders are considered as related support and services under the IDEA, and these are required by the IDEA to be listed in a student's IEP. Educational professionals are required by the IDEA to use scientifically based strategies and instructions for students with disabilities. To provide educational professionals evidence-based strategies, several searches were conducted in three electronic databases, ERIC, PsycINFO, and Web of Science, to find the studies that had been conducted at a school setting to address mental health and behavior problems in students with autism, ED, ID, multiple disabilities, and other health impairment. The educational models identified through the searches are presented below.

Positive Behavior Support (PBS)

PBS is specifically listed in the IDEA for schools to use to address students' behavioral needs. PBS uses the methods and strategies originated in applied behavior analysis (ABA) (e.g., functional analysis, shaping, fading, chaining, prompting, and reinforcement contingencies) to minimize a student's problem behaviors (Carr et al., 2002). Typically, PBS starts from conducting a functional analysis on a problem behavior of a target student to determine the purpose of the problem behavior (Carr, 1977). After knowing the purpose, a person-centered planning which identifies goals and intervention plans for a target student will then be started (Kincaid, 1996). PBS focuses on "fixing problem contexts, not problem behavior" (Carr et al., 2002, p. 8) in which the environment of the target student is restructured to allow changes to be happened and the support persons for the target student are to be adequately trained to facilitate changes (Knoster, Villa, & Thousand, 2000). PBS emphasizes on preventing a problem behavior to occur but not on how to deal with an occurring problem behavior (Carr et al., 2002). Common strategies that can be used to prevent the recurrence of a problem behavior include stopping reinforcing a previously reinforced response (Catania, 2013), reinforcing a desired behavior or providing a reinforcer upon no occurrence of a problem behavior (Catania, 2013), providing a student a reinforcer on a fixed or variable time schedule (Vollmer, Marcus, & Ringdahl, 1995), providing choices to a student (Dunlap et al., 1994), altering the valence of reinforcers for a problem behavior (Horner & Carr, 1997), and restructuring curricula to be functional and student centered (Dunlap, Kerndunlap, Clarke, & Robbins, 1991).

When applying PBS in school settings, PBS can include three levels of intervention: (a) primary prevention: universal strategies are applied to all the students in the school; (b) secondary prevention: selected strategies are used on the students at risk for developing problem behavior; and (c) tertiary prevention: comprehensive supports are provided to the students displaying problem behaviors (Horner, Sugai, Todd, & Lewis-Palmer, 2005). Goh and Bambara (2012) conducted a meta-analysis on the studies focusing on the tertiary prevention of the PBS in school settings and reported that the majority of these studies were conducted on elementary school students with developmental disabilities in special education classrooms. They classified these studies into four categories, antecedent-based only, consequence-based only, skills training only, and multicomponent, and found that many of the studies fell under the multicomponent category. They concluded that these studies focusing on using the PBS in schools have yielded promising results.

Art Therapy

Beh-Pajooh, Abdollahi, and Hosseinian (2018) conducted a study to examine the effectiveness of a painting therapy program on decreasing exter-

nalizing behaviors in children with ID. A total of 60 male students with ID (mean age = 12 years) from 20 special schools participated in their study. A painting therapy program was provided to the students in the intervention group. This program included 24 45-minute sessions (2 sessions per week). Students were provided with painting tools (e.g., marker, color pencil, crayon, gouache, and watercolor). In the first session, students were taught how to use the painting tools, and in the subsequent sessions, they were provided with a white sheet of paper for them to paint freely. Each session is closed with students explaining to the class their drawings. Their results indicated that students' externalizing behaviors were significantly decreased after the paining program.

Structured Physical Activity

Exercise is one of the alternative interventions with fewer disadvantages for students with ID and challenging behavior (Ogg-Groenendaal, Hermans, & Claessens, 2014). Cannella-Malone, Tullis, and Kazee (2011) conducted a study to examine the effects of systematic exercise on the challenging behavior of three students with moderate to severe developmental disabilities and an emotional behavior disorder. These students were instructed by their teachers to engage in daily exercise sessions including a 20-minute exercise routine first thing in the morning followed by a 1- to 5-minute exercise break every hour until after lunch across a variety of school settings (e.g., home classroom, art, music, gym, lunch, recess) and activities (e.g., academic and leisure). They reported that these students' challenging behavior was decreased to zero or close to zero. Prupas and Reid (2001) examined the effects of exercise frequency on stereotypic behaviors of children with developmental disabilities and found that one daily 10-minute walk/jog session in school resulted in decreased stereotypic behaviors and multiple daily exercise sessions were more effective than one single session. They also found that exercise combined with a structured classroom could yield an optimal outcome. Gencoz (1997) examined the effects of basketball training on the maladaptive behavior of 19 students with ID attending a special education school. The students received 40-minute training sessions three times a week for 7 weeks at the basketball court in their school. Gencoz reported that the students who received the basketball skills training showed a reduction in maladaptive behavior both at home and in the school. Ogg-Groenendaal et al. (2014) conducted a systematic review on the effect of exercise interventions on challenging behavior in people with ID and found that exercise could be considered as an effective treatment for people with ID and challenging behavior.

Social Stories™

Social stories are short stories written to explain social situations and provide practical social information for students with social difficulties (Gray, 1998). Kim, Blair, and Lim (2014) conducted a tablet-assisted Social Stories™ intervention study on high school students with severe ID and problem behaviors in a special school. The intervention took place in the teacher's office, and the intervention time was approximately 10 minutes per session. Students were instructed by their teachers to read the designed social stories on a tablet. After students had read their stories, teachers asked questions to students to check their understanding of the stories and provided verbal feedbacks. A token system was used to reward the target behaviors of students. The research results indicated that students' disruptive behaviors were decreased, and their academic engagement was increased.

Mindfulness-Based Approaches

In these approaches, students are taught to change the nature of their responses to problems (Singh et al., 2017). Students with ID can be taught to utilize these approaches to manage their aggressive behaviors (Singh & Jackman, 2017). Singh, Chan, Karazsia, McPherson, and Jackman (2017)

conducted a study that trained teachers to teach a mindfulness-based approach to students with ID and developmental disabilities to self-manage their aggressive behaviors. Each training session was 15- to 30-minute long prior to the beginning of the first class, and the trainings lasted for 3 weeks. The mindfulness-based approach used in their study is mediation on the soles of the feet (SoF). SoF teaches students to shift their attention from the anger-producing situation to a neutral point on the body (Singh, et al., 2017). Singh, et al. (2017) reported that this mindfulness-based approach had resulted in reducing physical and verbal aggressions in the students participated in the study.

Daily Behavior Report Cards

Behavior interventions are typically used by teachers and other education professionals to help students with ID and challenging behaviors to manage their behaviors. Daily behavior report cards (DBRCs) are one of the behavioral interventions that have been applied on students with ID and challenging behaviors. When implementing a DBRC, a specific behavior is defined and rated at least once per day, and a reward system for a target student is predetermined (Chafouleas, Riley-Tillman, Sassu, LaFrance, & Patwa, 2007). By using a DBRC, the target behavior of a target student is reviewed regularly, and the student is provided with feedback on progress by adults (Taylor & Hill, 2017). Taylor and Hill (2017) conducted a study to examine the effectiveness of using DBRCs to improve appropriate classroom behaviors of young students with ID. Each participant had a DBRC, and the DBRC was created by using these steps: (a) select target behaviors, (b) determine points for behaviors, and (c) determine how often and when students' behaviors should be evaluated. Teachers provided constant feedback to students regarding their behavior performance and communicated with students' parents regularly about their child's behaviors. Taylor and Hill (2017) reported that DBRCs resulted in positive impacts on classroom behaviors of children with ID.

Issues for Providing Effective Interventions to Students with Dual Diagnosis in School

Individuals with ID are more likely to display aggressive behaviors than those with other disabilities (Ageranioti-Belanger et al., 2012). If children with ID are not provided with effective interventions to deal with their challenging behaviors, their challenging behaviors can become more problematic and have negative impacts on their adult outcomes (Taylor & Hill, 2017). Thus, it is important for these students to receive effective interventions. However, there are issues that hinder the delivery of these interventions in school.

Few Evidence-Based Interventions Are Available for School Settings

Education professionals are required to use evidence-based strategies to help students to manage their mental health and behavior problems. However, it is always a challenge for education professionals to apply effective interventions from research to daily school practice (Burton & Chapman, 2004), and there are limited intervention studies that have been conducted in school settings. Several approaches, such as dialectical behavior therapy (Brown, Brown, & Dibiasio, 2013), psychosocial interventions (Gustafsson et al., 2009), and cognitive behavior therapy (Kellett, Matuozzo, & Kotecha, 2015), have been used in individuals with dual diagnosis, but there is a lack of research applying these approaches in a school setting. Campbell, Robertson, and Jahoda (2014) conducted a review study on the psychological therapy studies for people with ID and challenging behaviors and reported that the evidence for successful psychological interventions for students with ID and challenging behavior is thin.

Mental health problems are commonly found in people with ID (Cooper, Smiley, Morrison, Williamson, & Allan, 2007). Behavioral, cognitive behavioral, psychodynamic, and psychoeducational approaches represented the intervention techniques for addressing mental health problems in people with ID (Benson, 2004). The interven-

tions that work for people without ID would work for people with ID (Dagnan, 2007b). Although there are concerns about providing psychosocial interventions for people with ID because they have deficits in cognitive functioning and language abilities, it is generally believed that these interventions can be modified to meet the cognitive and verbal levels of these people (Hurley, 1989). However, effective psychosocial interventions have been rarely reported for people with ID (Dagnan, 2007a), and the intervention research that specifically addresses mental health problems in these people is very limited, especially those for people with severe and profound ID (Vereenooghe et al., 2018).

Not Enough Properly Trained Educational Professionals

At least 10% of individuals with ID display challenging behaviors (Emerson et al., 2001), but the available services for students with ID are often not enough to meet these students' needs (Gregori, Rispoli, Gerow, & Lory, 2018). Several factors in the school environment may account for a low number of properly trained educational professionals for students with ID and challenging behaviors. First, challenging behavior puts the educational professionals who support students with ID at a risk of harm (Lambert, Bloom, Clay, Kunnavatana, & Collins, 2014), especially those supporting adults with ID because of large body size and the severe form of challenging behaviors in these adults. Thus, some educational professionals are reluctant to intervene with students with ID and challenging behavior (Manente, Maraventano, LaRue, Delmolino, & Sloan, 2010). Second, it is common to learn that the educational professionals for students with ID are not provided with adequate training and relevant resources by their school districts (Jahr, 1998). Thus, the teachers who are interested in supporting these students may not know how to properly support these students and may be accidentally harmed by them, thus, withdraw themselves from providing further assistance to these students. Third, although the IDEA requires teachers to use evidence-based strategies to manage students' behaviors, it may not be easy for teachers to apply effective interventions from literature to daily practice (Campbell et al., 2014). Reading effective strategies from the literature can indeed give educational professionals some good ideas for how to intervene students' mental health and behavior problems. However, without proper training and guidance, educational professionals may not find effectiveness in using evidence-proven strategies on their students at a specific school setting.

Hard to Find Balance in Teaching Academic Skills and Addressing Other Needs

Teachers in the United States, including teachers of students with disabilities, are required to align their curriculum to the Common Core State Standards (CCSS) which are a set of academic standards in mathematics and English language arts (ELA)/literacy. However, this requirement has limited special education teachers in devoting their time to address students' nonacademic needs (Kanso, 2015). Murphy and Haller (2015) conducted a qualitative study to explore the experiences of teachers of English language learners and students with disabilities as they sought to align the CCSS with previously used standards and instructional approaches during the first year of the CCSS implementation. They reported that teachers of students with disabilities expressed their concerns of fulfilling the demands of the CCSS, adapting their curriculum to the CCSS, and meeting their students' non-English and mathematics needs. When teachers have the pressure to meet the State-required learning standards, they can only sacrifice the time for helping students to improve their adaptive skills.

Conclusion and Suggestions

Mental health and behavior problems are often found in people with ID, and these problems can have negative impact on these people's life and create challenges for their caregivers (Brown et al.,

2013). Students with disabilities in the United States are entitled to receive free and appropriate special education and related services. Thus, special education teachers and related educational professionals should have the knowledge and skills to address their students' mental and behavioral needs. However, there are limited evidence-based educational models for teachers to use for their students with dual diagnosis. This means teachers have only limited tools to address their students' diverse needs. To address the problem, some researchers have advocated that educational professionals should look for the interventions that have been proven to be effective in the general population (Prout & Nowak-Drabik, 2003) and in people with mental disorders without ID (National Institute for Health and Care Excellence, 2016) and adapt these on students with dual diagnosis.

More School Intervention Studies Should Be Conducted

School environments are different from clinical and community settings. The effective intervention studies conducted in other than school settings, although with proven effectiveness, may not be applied practically in a school. A student with ID and challenging behaviors and/or mental health problems may be placed in a general education classroom, inclusive classroom, or self-contained special education classroom. Each of these settings have different academic requirements, class schedules, and student populations, and these variables can have direct impacts on the intervention plans to address students' mental health and cope with their behavior problems. Thus, an intervention showing effectiveness in one setting may not be practical for another. Therefore, more intervention studies in a variety of classroom settings should be conducted to provide teachers more tools and resources to help their students with dual diagnosis.

Educational Professionals Should Be Equipped with ABA Knowledge

Having appropriately trained professionals to conduct interventions is an essential component of any educational models in school for students with disabilities (Campbell et al., 2014). Individuals with dual diagnosis may require intensive and comprehensive treatment over a long period of time (Brown et al., 2013). However, there are limited educational professionals who have received adequate training to help students manage their behaviors and cope their mental issues. Many educational professionals who provide support to students with dual diagnosis do not have appropriate training (Ito, Kurita, & Shiiya, 1999). The shortage of trained educational professionals makes it difficult for schools to provide treatment for students with dual diagnosis (Manente et al., 2010). Nonetheless, every student with dual diagnosis still needs help. The effectiveness of applying ABA strategies on challenging behaviors of students with ID has been documented in the literature (Harvey, Boer, Meyer, & Evans, 2009; Hassiotis et al., 2011; Luiselli, 2009). Functional behavior assessment based interventions can effectively reduce students' problem behaviors and increase their use of appropriate behaviors (Goh & Bambara, 2012). Thus, trainings should be provided to educational professionals supporting students with challenging behaviors to acquire the knowledge of ABA and the skills of applying these strategies to manage students' behaviors. Ongoing support should also be provided to these educational professionals.

Embed Behavioral Interventions into Academic Instruction

It is very common to learn that teachers do not have enough time to address both students' academic and nonacademic needs. However, teachers have the pressure from their State to align their curriculum with the State standards but at the same time feel the urgent need to deal with students' mental health and behavior problems. Thus, a solution that may overcome the difficulty is that teachers can try to embed the behavioral/mental intervention into academic instruction. For example, teachers can teach mathematics (e.g., number sense, calculation, and subtraction) and introduce a token system to students in a

mathematics lesson. Students can learn the required mathematics knowledge and at the same time learn that if they display good behaviors, they can earn points and exchange these points to something they like. Teachers can discuss ways to manage anger mood while teaching ELA. Using thematic instruction may make it easier to integrate multiple subjects learning into one coherent learning experience. Thematic instruction uses themes to integrate multiple subjects (e.g., language arts, mathematics, science, social studies) instruction (Erwin, Hines, & Curtis, 1992). This type of instruction allows students to learn new knowledge as a coherent whole experience (Funderstanding.com, 2018). Thematic instruction has been used in teaching students with and without disabilities since the 1960s (Erwin et al., 1992). Previous studies have reported the effectiveness of using thematic instruction to foster the development of literacy skills (Gelzheiser, Hallgren-Flynn, Connors, & Scanlon, 2014), mathematical abilities (Henderson & Landesman, 1995), and socioemotional skills (Pataki, Metz, & Pakulski, 2014).

References

Ageranioti-Belanger, S., Brunet, S., D'Anjou, G., Tellier, G., Boivin, J., & Gauthier, M. (2012). Behaviour disorders in children with an intellectual disability. *Paediatrics and Child Health, 17*, 84–88. https://doi.org/10.1093/pch/17.2.84

Beh-Pajooh, A., Abdollahi, A., & Hosseinian, S. (2018). The effectiveness of painting therapy program for the treatment of externalizing behaviors in children with intellectual disability. *Vulnerable Children and Youth Studies, 13*, 221–227. https://doi.org/10.1080/17450128.2018.1428779

Benson, B. A. (2004). Psychological interventions for people with intellectual disability and mental health problems. *Current Opinion in Psychiatry, 17*, 353–357. https://doi.org/10.1097/01.yco.0000139969.14695.dc

Brown, J. F., Brown, M. Z., & Dibiasio, P. (2013). Treating individuals with intellectual disabilities and challenging behaviors with adapted dialectical behavior therapy. *Journal of Mental Health Research in Intellectual Disabilities, 6*, 280–303. https://doi.org/10.1080/19315864.2012.700684

Burton, M., & Chapman, M. J. (2004). Problems of evidence based practice in community based services. *Journal of Intellectual Disabilities, 8*, 56–70.

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*, 172–188. https://doi.org/10.1111/j.1365-2788.2012.01646.x

Cannella-Malone, H. I., Tullis, C. A., & Kazee, A. R. (2011). Using antecedent exercise to decrease challenging behavior in boys with developmental disabilities and an emotional disorder. *Journal of Positive Behavior Interventions, 13*, 230–239. https://doi.org/10.1177/1098300711406122

Carr, E. G. (1977). The motivation of self-injurious behavior: A review of some hypotheses. *Psychological Bulletin, 84*, 800–816.

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

Catania, A. C. (2013). *Learning* (5th ed.). New York, NY: Sloan Publishing, Cornwall-on-Hudson.

Chafouleas, S. M., Riley-Tillman, T. C., Sassu, K. A., LaFrance, M. J., & Patwa, S. S. (2007). Daily behavior report cards: An investigation of the consistency of on-task data across raters and methods. *Journal of Positive Behavior Interventions, 9*, 30–37. https://doi.org/10.1177/10983007070090010401

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. https://doi.org/10.1192/bjp.bp.106.022483

Dagnan, D. (2007a). Psychosocial interventions. In N. Bouras & G. Holt (Eds.), *Psychiatric and behavioural disorders in intellectual and developmental disabilities* (pp. 330–338). Cambridge: Cambridge University Press.

Dagnan, D. (2007b). Psychosocial interventions for people with intellectual disabilities and mental ill-health. *Current Opinion in Psychiatry, 20*, 456–460. https://doi.org/10.1097/YCO.0b013e3282ab9963

Dunlap, G., de Perczel, M., Clarke, S., Wilson, D., Wright, S., White, R., & Gomez, A. (1994). Choice making and proactive behavioral support for students with emotional and behavioral challenges. *Journal of Applied Behavior Analysis, 27*, 505–518.

Dunlap, G., Kerndunlap, L., Clarke, S., & Robbins, F. R. (1991). Functional assessment, curricular revision, and severe behavior problems. *Journal of Applied Behavior Analysis, 24*, 387–397. https://doi.org/10.1901/jaba.1991.24-387

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

Erwin, B., Hines, C., & Curtis, C. (1992). Thematic units: A Scottish approach to literature-based instruction. *Reading Horizons, 33*, 108–120.

Funderstanding.com. (2018). *Thematic instruction*. Retrieved from https://www.funderstanding.com/educators/thematic-instruction/.

Gelzheiser, L., Hallgren-Flynn, L., Connors, M., & Scanlon, D. (2014). Reading thematically related texts to develop knowledge and comprehension. *Reading Teacher, 68*, 53–63. https://doi.org/10.1002/trtr.1271

Gencoz, F. (1997). The effects of basketball training on the maladaptive behaviors of trainable mentally retarded children. *Research in Developmental Disabilities, 18*, 1–10. https://doi.org/10.1016/s0891-4222(96)00029-7

Goh, A. E., & Bambara, L. M. (2012). Individualized positive behavior support in school settings: A meta-analysis. *Remedial and Special Education, 33*, 271–286. https://doi.org/10.1177/0741932510383990

Gray, C. (1998). Social Stories and comic strip conversations with students with Asperger syndrome and high-functioning autism. In E. Schopler, G. B. Mesibov, & L. J. Kunce (Eds.), *Asperger syndrome or high-functioning autism?* (pp. 167–198). New York: Plenum.

Gregori, E., Rispoli, M., Gerow, S., & Lory, C. (2018). Treatment of self-injurious behavior in adults with intellectual and developmental disabilities: A systematic review. *Journal of Developmental and Physical Disabilities, 30*, 111–139. https://doi.org/10.1007/s10882-017-9568-7

Gustafsson, C., Ojehagen, A., Hansson, L., Sandlund, M., Nystrom, M., Glad, J., … Fredriksson, M. (2009). Effects of psychosocial interventions for people with intellectual disabilities and mental health problems. *Research on Social Work Practice, 19*, 281–290. https://doi.org/10.1177/1049731508329403

Harvey, S. T., Boer, D., Meyer, L. H., & Evans, I. M. (2009). Updating a meta-analysis of intervention research with challenging behaviour: Treatment validity and standards of practice. *Journal of Intellectual and Developmental Disability, 34*, 67–80. https://doi.org/10.1080/13668250802690922

Hassiotis, A., Canagasabey, A., Robotham, D., Marston, L., Romeo, R., & King, M. (2011). Applied behaviour analysis and standard treatment in intellectual disability: 2-year outcomes. *British Journal of Psychiatry, 198*, 490–491. https://doi.org/10.1192/bjp.bp.109.076646

Henderson, R. W., & Landesman, E. M. (1995). Effects of thematically integrated mathematics instruction on students of mexican descent. *Journal of Educational Research, 88*, 290–300.

Horner, R. H., & Carr, E. G. (1997). Behavioral support for students with severe disabilities: Functional assessment and comprehensive intervention. *Journal of Special Education, 31*, 84–104. https://doi.org/10.1177/002246699703100108

Horner, R. H., Sugai, G., Todd, A. W., & Lewis-Palmer, T. (2005). Schoolwide positive behavior support. In L. M. Bambara & L. Kern (Eds.), *Individualized supports for students with problem behaviors: Designing positive behavior plans* (pp. 359–390). New York, NY: Guilford.

Hurley, A. D. (1989). Individual psychotherapy with mentally-retarded individuals: A review and call for research. *Research in Developmental Disabilities, 10*, 261–275. https://doi.org/10.1016/0891-4222(89)90015-2

IDEA. (2004). *Individuals with disabilities education improvement act of 2004*, 20 U.S.C., § 1400 et seq.

Ito, H., Kurita, H., & Shiiya, J. (1999). Burnout among direct-care staff members of facilities for persons with mental retardation in Japan. *Mental Retardation, 37*, 477–481.

Jahr, E. (1998). Current issues in staff training. *Research in Developmental Disabilities, 19*, 73–87. https://doi.org/10.1016/s0891-4222(97)00030-9

Kanso, H. (2015). *Common core: What's right for special education students?* Retrieved from https://www.cbsnews.com/news/common-core-whats-right-for-special-educations-students/.

Kellett, S., Matuozzo, H., & Kotecha, C. (2015). Effectiveness of cognitive-behaviour therapy for hoarding disorder in people with mild intellectual disabilities. *Research in Developmental Disabilities, 47*, 385–392. https://doi.org/10.1016/j.ridd.2015.09.021

Kim, M. S., Blair, K. S. C., & Lim, K. W. (2014). Using tablet assisted Social Stories (TM) to improve classroom behavior for adolescents with intellectual disabilities. *Research in Developmental Disabilities, 35*, 2241–2251. https://doi.org/10.1016/j.ridd.2014.05.011

Kincaid, D. (1996). Person-centered planning. In L. K. Koegel, R. L. Koegel, & G. Dunlap (Eds.), *Positive behavior support*. Baltimore: Brookes.

Knoster, T. P., Villa, R. A., & Thousand, J. S. (2000). A framework for thinking about systems change. In R. A. Villa & J. S. Thousand (Eds.), *Restructuring for caring and effective education* (pp. 93–128). Baltimore: Brookes.

Lambert, J. M., Bloom, S. E., Clay, C. J., Kunnavatana, S. S., & Collins, S. D. (2014). Training residential staff and supervisors to conduct traditional functional analyses. *Research in Developmental Disabilities, 35*, 1757–1765. https://doi.org/10.1016/j.ridd.2014.02.014

Luiselli, J. K. (2009). Behavior support of people with intellectual and developmental disabilities: Contemporary research applications. *Journal of Developmental and Physical Disabilities, 21*, 441–442. https://doi.org/10.1007/s10882-009-9162-8

Manente, C. J., Maraventano, J. C., LaRue, R. H., Delmolino, L., & Sloan, D. (2010). Effective behavioral intervention for adults on the autism spectrum: Best practices in functional assessment and treatment. *The Behavior Analyst Today, 11*, 36–48.

Murphy, A. F., & Haller, E. (2015). Teachers' perceptions of the implementation of the literacy Common Core State Standards for English language learners and students with disabilities. *Journal of Research in Childhood Education, 29*, 510–527. https://doi.org/10.1080/02568543.2015.1073200

National Institute for Health and Care Excellence. (2016). *Mental health problems in people with learning disabilities: Prevention, assessment and management*. Reetrieved from https://www.nice.org.uk/guidance/ng54?unlid=942277916201713131247

Office of Special Education Programs. (2018). *About IDEA*. Retrieved from https://sites.ed.gov/idea/about-idea/.

Ogg-Groenendaal, M., Hermans, H., & Claessens, B. (2014). A systematic review on the effect of exercise interventions on challenging behavior for people with intellectual disabilities. *Research in Developmental Disabilities, 35*, 1507–1517. https://doi.org/10.1016/j.ridd.2014.04.003

Pataki, K. W., Metz, A. E., & Pakulski, L. (2014). The effect of thematically related play on engagement in storybook reading in children with hearing loss. *Journal of Early Childhood Literacy, 14*, 240–264.

Prout, H. T., & Nowak-Drabik, K. M. (2003). Psychotherapy with persons who have mental retardation: An evaluation of effectiveness. *American Journal on Mental Retardation, 108*, 82–93. https://doi.org/10.1352/0895-8017(2003)108<0082:pwpwhm>2.0.co;2

Prupas, A., & Reid, G. (2001). Effects of exercise frequency on stereotypic behaviors of children with developmental disabilities. *Education and Training in Mental Retardation and Developmental Disabilities, 36*, 196–206.

Singh, N. N., Chan, J., Karazsia, B. T., McPherson, C. L., & Jackman, M. M. (2017). Tele-health training of teachers to teach a mindfulness-based procedure for self-management of aggressive behavior to students with intellectual and developmental disabilities. *International Journal of Developmental Disabilities, 63*, 195–203. https://doi.org/10.1080/20473869.2016.1277841

Singh, N. N., & Jackman, M. M. (2017). Teaching mindfulness to individuals with intellectual and developmental disabilities and their caregivers. In D. McCown, D. K. Reibel, & M. S. Micozzi (Eds.), *Resources for teaching mindfulness: A cross-cultural and international handbook*. New York, NY: Springer.

Singh, N. N., Lancioni, G. E., Myers, R. E., Karazsia, B. T., Courtney, T. M., & Nugent, K. (2017). A mindfulness-based intervention for self-management of verbal and physical aggression by adolescents with Prader-Willi syndrome. *Developmental Neurorehabilitation, 20*, 253–260. https://doi.org/10.3109/17518423.2016.1141436

Taylor, J. C., & Hill, D. (2017). Using daily behavior report cards during extended school year services for young students with intellectual and developmental disabilities. *Education and Treatment of Children, 40*, 525–546.

Vereenooghe, L., Flynn, S., Hastings, R. P., Adams, D., Chauhan, U., Cooper, S. A., … Waite, J. (2018). Interventions for mental health problems in children and adults with severe intellectual disabilities: A systematic review. *BMJ Open, 8*, e021911. https://doi.org/10.1136/bmjopen-2018-021911

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*, 15–26. https://doi.org/10.1901/jaba.1995.28-15

8 Risk Factors for Dual Disorders in Individuals with Intellectual Disabilities

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Intellectual disabilities (ID) are defined by the *Diagnostic and Statistical Manual of Mental Disorders* (5th ed.; DSM-5; American Psychiatric Association, 2013) as a developmental disability consisting of impaired intellectual and adaptive functioning, manifested though three specific domains (social, conceptual, and practical). Individuals with intellectual disabilities are at significant risk for the development and severity of mental health problems. Research demonstrates that both children and adults with intellectual disabilities exhibit higher rates of behavioral problems, emotional disturbance, and psychiatric disorders than individuals without intellectual disabilities (Harris, 2006; Matson & Frame, 1985). The prevalence of individuals with co-occurring psychopathology and ID varies, with estimates ranging from 7% to 97% (Cooper, Smiley, Morrison, Williamson, & Allan, 2007). This discrepancy in prevalence rates is likely due to varying assessment methods, samples, and diagnostic criteria (Cooper et al., 2007; Dykens, 2000; Maulik, Mascarenhas, Mathers, Dua, & Saxena, 2011). Further, comorbid psychological disorders may be more difficult to detect in individuals with ID, particularly those in the moderate to profound range, due to impairments in communication and cognitive functioning (Axmon, Björne, Nylander, & Ahlström, 2017; Deb, Thomas, & Bright, 2001; Matson, Smiroldo, & Bamburg, 1998). Psychopathology may also be difficult to identify in individuals with ID due to difficulties in assessing symptoms that may present differently in individuals with ID (Moss, 2001).

There are a number of biological and environmental factors that contribute to the comorbidity of psychological disorders in individuals with intellectual disabilities. The literature illuminates many of the risk factors associated with comorbid psychopathology in individuals with ID. Specific biological factors including genetic syndromes, gender, and chromosomal abnormalities are associated with an individual's susceptibility to specific comorbidities. Environmental risk factors associated with psychopathology in individuals with ID include socioeconomic status, job satisfaction, and exposure to stressful life events. This chapter will explore the many risk factors associated with comorbid psychological disorders and intellectual disabilities. Additionally, discussion will include examination of the many complexities and issues that undermine continued research on risk factors for psychopathology within this population.

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Severity of Intellectual Disability

The variation of adaptive and cognitive functioning among individuals with intellectual disabilities led to the hierarchical diagnostic system, which allows clinicians to distinguish the level of severity into four categories: mild, moderate, severe, and profound. Research demonstrates that the severity of intellectual disability serves as a risk factor for the development of psychopathology. Specifically, there are key differences in both the likelihood of developing a psychological disorder as well as type of psychological disorder in individuals with different levels of cognitive impairment (Eisenhower, Baker, & Blacher, 2005). The majority of research studies found that children and adults with ID are at a higher risk of developing a psychological disorder than the general population (Dekker & Koot, 2003; Einfeld, Ellis, & Emerson, 2011; Emerson, 2003). Further, individuals with mild/moderate ID are at a higher risk of developing psychopathology than those with severe/profound ID (Axmon et al., 2017; Koskentausta, Iivanainen, & Almqvist, 2007; Tsiouris, Kim, Brown, & Cohen, 2011). However, several studies found little to no differences in the prevalence of psychological disorders based on the level of cognitive impairment (Einfeld et al., 2011). The disparities in observed rates of psychopathology based on the degree of intellectual impairment are likely due to limited research in the area. Despite conflicting research findings regarding the level of intellectual impairment, a large body of evidence supports the differences in the types of co-occurring psychiatric diagnoses present in individuals with ID and the differences in the risk factors that contribute to the development of psychopathology.

The majority of individuals with intellectual impairments are in the mild range of functioning. Mild cognitive impairment is conceptualized in the DSM-5 as individuals who exhibit difficulties in communication, daily living tasks, and cognitive/intellectual abilities that require minimal levels of support (American Psychiatric Association, 2013). Individuals with mild ID are more likely to exert greater control and independent decision-making than more impaired individuals. Further, individuals with mild ID are at an increased likelihood to have independent living situations, financial independence and control, and manage their own health. Research indicates that individuals with mild impairments are at greater risk for developing psychopathology than those with more severe ID. For example, individuals with mild intellectual disabilities are more likely to experience mood disorders than individuals with more severe cognitive impairments (Hartley & MacLean, 2009; Hurley, 2006; McGillivray & McCabe, 2007). The increased likelihood of individuals with mild cognitive impairments to experience depressive symptoms may be due to a greater capacity to experience maladaptive cognitive processes than more severely impaired individuals. One of the hallmark symptoms of major depressive disorder is the presence of cognitive distortions that negatively impact functioning (Gotlib & Joormann, 2010). For example, maladaptive thoughts can lead to depressed mood and negative affect. Individuals with mild ID possess a greater cognitive capacity, which is necessary for negative thoughts to occur. Maladaptive cognitions include dysfunctional self-statements that result in experiencing depressive symptoms, such as feelings of worthlessness and lowered self-esteem. Research demonstrates that individuals with ID who engage in negative social comparison tend to develop poor self-esteem and depressive symptoms on par with the general population (Dagnan & Sandhu, 1999). This is especially likely in individuals who lack protective factors such as feelings of belonging and adequate social support. Individuals with intellectual disabilities are less likely to acquire social roles that contribute to self-worth (Dagnan & Sandhu, 1999; Linville, 1987). For example, individuals without intellectual disabilities are more likely to develop a wide variety of complex social roles that contribute to their self-aspect, such as being a parent, spouse, lawyer, community leader, nurse, runner, and many more. These roles generally fall within specific categories, such as relationships with others, careers, specific activities, and personality traits (Linville, 1987). Individuals with ID are less likely to develop a

large variety of social roles due to restrictions placed on them by society or their own limitations. The restricted opportunities for role acquisition by individuals with mild cognitive impairments often lead to increased negative social comparisons, which further increases negative affect. For example, McGillivray and McCabe (2007) found that individuals with mild intellectual disabilities were at increased risk of developing depressive symptomology due to social comparisons. As with the general population of individuals without intellectual disabilities, individuals with mid/moderate ID demonstrated increased negative evaluation of themselves in comparison to others, which in turn negatively impacted their self-esteem, identity, and sense of self-worth. Mild ID individuals who experience negative thoughts, such as negative evaluations of the self, biased interpretations of events, attentional biases to adverse and harmful information, and destructive attitudes about one's own situation and experiences, are more likely to understand and process these maladaptive cognitions.

In addition to the increased presence and recognition of maladaptive cognitions, individuals with mild ID are more likely to experience stressful social interactions (compared to their nondisabled counterparts). Negative social experiences include isolation, rejection, and criticism from others and are associated with increased depressive symptoms. Individuals with mild ID are at a greater risk for experiencing negative social interactions due to engagement in maladaptive thought patterns and limited social skills (Hartley & MacLean, 2009). Heiman (2001) found that adolescents with mild ID who attended specialized schools for students with disabilities endorsed significantly more symptoms of depression and feelings of loneliness than those who attended a school with typically developing peers. Heiman posited that this may be due to the fact that students with mild ID who are enrolled in traditional academic settings were more likely to experience opportunities for social interaction with their peers inside and outside of school. Research demonstrates that individuals with mild ID are more likely to feel lonely and isolated due to difficulties initiating and maintaining relationships with others. Gilmore and Cuskelly (2014) suggested that stigmatization and negative attitudes toward individuals with intellectual disabilities create barriers to opportunities for social engagement. Social isolation further exacerbates deficits in social skills, as cognitively impaired individuals are less likely to experience interactions with individuals who model appropriate behavior. Research suggests that difficulties in behavior regulation in children with mild and moderate ID also contribute to increased social isolation and rejection (compared to their typically developing peers). Externalizing behaviors, such as aggression, poor emotional regulation, and disruptive behavior, is common in children with mild ID, and these behaviors are often associated with rejection and criticism from typically developing peers (Bellanti & Bierman, 2000; Einfeld et al., 2006; Taggart, Taylor, & McCrum-Gardner, 2010).

Compared to individuals with mild ID, individuals with moderate ID have more obvious delays in adaptive functioning, cognitive abilities, and social communication and require greater levels of support (American Psychiatric Association, 2013). Research indicates that individuals with moderate ID tend to experience psychological disorders at rates comparable to the general population (Dekker & Koot, 2003; Turygin, Matson, & Adams, 2014). For instance, children with moderate ID often exhibit aggressive and destructive behaviors, such as self-injurious behaviors (Handley, Adams, Simkiss, & Oliver, 2013). This is a significant concern, as these behaviors are often attributable to changes in the environment or experiences of pain, but individuals with moderate ID may have trouble communicating these experiences in an appropriate manner. Further, self-injurious behaviors in individuals with moderate ID are often persistent and can have long-term consequences (Tureck, Matson, & Beighley, 2013). In regard to autism spectrum disorder (ASD), there is some research that indicates ASD is more common in individuals with moderate ID than those with mild ID (Turygin et al., 2014). However, the current research on co-occurring ID and autism is limited,

likely attributed to the fact that consideration of ASD alongside a diagnosis of ID has only recently become accepted practice (Matson & Shoemaker, 2009).

The dearth in research on the comorbidity of psychiatric diagnoses in individuals with ID is particularly true of individuals with moderate intellectual disabilities. The few studies that have examined comorbid psychopathology and intellectual disability generally do not differentiate individuals with moderate ID from those with mild ID. To address this lack of research, Dekker and Koot (2003) evaluated the experiences of children with moderate ID compared to typically developing children. The authors found that children with moderate ID compared to typically developing peers experienced similar types and prevalence of psychological disorders, including disruptive, mood, and anxiety disorders. Some research indicates that individuals with moderate ID possess some of the same risk factors as those with mild ID, such as intense feelings of loneliness due to social exclusion and isolation (Gilmore & Cuskelly, 2014). Research in risk factors specific to individuals with moderate ID separate from those with mild ID is lacking. As the presentation of moderate ID differs significantly from other severity levels, it is likely that these individuals may experience psychological symptoms in a unique way. However, limitations in language and communication create further problems in assessment, and differing presentations may be incorrectly attributed to others.

Similar to moderate ID, severe and profound IDs are often considered as a singular presentation in research on risk factors for the development of psychological disorders. Individuals with severe ID demonstrate noticeably deficient communication abilities and limited language and literacy skills. Further, these individuals require supervision and support for activities of daily living (American Psychiatric Association, 2013). Individuals with profound ID possess extremely limited communication and language abilities and visuospatial skills and exhibit increased dependency on others for every aspect of daily living (American Psychiatric Association, 2013). In all likelihood, research has failed to differentiate individuals with severe and profound ID due to the combined incidence rate of less than 1% of the general population (Maulik et al., 2011).

Research on mental illness in individuals with severe/profound ID indicates that these individuals are at a greater risk for developing challenging behavior than their less severely impaired counterparts. Challenging behaviors are defined as harmful and disruptive behaviors that are typically observed in individuals with ID, including self-injurious behavior (SIB), destructive behavior, pica, and aggression (Matson & Minshawi, 2007). Challenging behavior problems are evident across the life span in individuals with severe/profound impairment (Handley et al., 2013; Tureck et al., 2013; Witwer & Lecavalier, 2008). One risk factor for the occurrence of SIB, aggression, and destructive behaviors is comorbid diagnosis of autism spectrum disorder (ASD), which is the most prevalent co-occurring disorder in individuals with intellectual disabilities (Handley et al., 2013; LoVullo & Matson, 2009). Further, individuals with severe/profound ID and ASD exhibit challenging behavior at a rate roughly four times higher than individuals without comorbid ASD (Bradley, Summers, Wood, & Bryson, 2004). Despite this, research is mixed on identifying factors that contribute to the disparities in comorbid mental illness in individuals with ASD and severe/profound ID. Tureck et al. (2013) found that self-injurious behavior was more common in individuals with severe ID and ASD than individuals with severe ID alone. Further, they posited that the association is likely due to the fact that SIB is a commonly observed stereotypic behavior in cognitively impaired individuals with ASD. Similarly, Oliver, Petty, Ruddick, and Bacarese-Hamilton (2012) found that repetitive or ritualistic behaviors served as a risk factor for the presence of challenging behavior in children with severe ID. Although the study did not examine ASD as a risk factor for developing challenging behaviors in children with severe ID, the authors noted that the occurrence of repetitive behaviors may suggest a diagnosis of comorbid ASD. Several studies found a link between the presence of stereotypic behaviors and self-injurious behaviors in individuals with

severe or profound impairments (Davies & Oliver, 2016; Furniss & Biswas, 2012). Research also has indicated that individuals with severe/profound ID are at a higher risk of developing SIBs irrespective of a comorbid ASD diagnosis due to deficits in overall functioning (Furniss & Biswas, 2012). Individuals with severe/profound ID are also at increased risk of developing challenging behaviors due to health problems that cause physical pain. For example, individuals with ID who exhibit SIB may do so in response to experiencing pain. The SIB may serve as a method of alleviating pain, as the behavior can be a distractor from the sensory experience of pain (Handley et al., 2013).

Aggression and destructive behaviors are also commonly seen in individuals with severe/profound ID for several possible reasons. As with SIB, individuals with severe/profound ID often demonstrate restrictive and repetitive behavior, which serves as a risk factor for exhibiting aggressive and destructive behavior (Davies & Oliver, 2016; Lundqvist, 2013). Several environmental variables have been linked to increased aggression and destructive behavior. For example, adolescents with ID who lacked continuity in maternal care or whose parents employed corporal punishment were significantly more likely to exhibit behavior problems (Chadwick, Kusel, & Cuddy, 2008). Dysfunctional patterns of sleep also have served as a risk factor for adults with severe/profound ID in the exhibition of externalizing behaviors. Lundqvist (2013), for example, found that adults with severe/profound ID who slept poorly were more likely to engage in aggressive or destructive behaviors than those with adequate sleep. Additionally, children with ID who experienced sleep problems were more likely to demonstrate challenging behaviors, as well as symptoms of anxiety, than their counterparts who had adequate sleep (Rzepecka, McKenzie, McClure, & Murphy, 2011).

Individuals with severe/profound ID are at increased risk for exhibiting pica, the ingestion of inedible items. Pica often leads to an array of negative health outcomes, such as intestinal parasites and the need for surgical intervention (Ali, 2001). Ashworth, Martin, and Hirdes (2008) found a relationship between the level of cognitive impairment and pica in individuals with ID, but the risk decreased when the cognitive impairment reached its highest levels. Further, this study found a negative relationship between the probability of pica and the presence of higher adaptive functioning and daily living skills. Research has identified specific biological mechanisms that place individuals with ID at greater risk for pica. Specifically, ID individuals with iron and zinc deficiencies are more likely to exhibit pica (Ali, 2001; Matson, Belva, Hattier, & Matson, 2011; Swift, Paquette, Davison, & Saeed, 1999).

Overall, research on the correlates of ID severity and co-occurring psychopathology is in its infancy. Future research should examine specific differences between ID severity levels, rather than clustering levels using inconsistent inclusion criteria. However, research is hindered due to the majority of individuals with ID being diagnosed with mild cognitive impairments; thus, conducting studies with sufficient sample sizes of individuals with moderate, severe, and profound ID is challenging.

Behavioral Phenotypes and Genetic Markers

Intellectual disabilities are often linked to genetic abnormalities, and that may play an important role in the development of psychopathology for individuals with ID. In particular, recent findings substantiate that individuals with ID are susceptible to biological risk factors that increase the likelihood they will experience comorbid psychological problems. Behavioral phenotypes are the distinct, observable characteristics that tend to occur at a higher rate in individuals with a particular genetic syndrome. A substantial amount of evidence has found specific behavioral phenotypes of genetic syndromes commonly associated with ID serve as risk factors for psychiatric diagnoses and specific psychological symptoms. For the purposes of this chapter, discussion of phenotypic expression is highlighted through the use of examples, as there are several hundreds of genetic

syndromes that commonly co-occur with ID, and an exhaustive discussion of these syndromes would defeat the purpose of this chapter.

One of the most common genetic syndrome comorbidities in individuals with intellectual disabilities is Down syndrome, which is defined by errant cell division that results in the presence of 47 chromosomes. Individuals with comorbid ID and Down syndrome typically exhibit fewer behavior problems than their typically developing peers (Dykens & Kasari, 1997; Eisenhower et al., 2005). This is generally attributed to evidence that individuals with Down syndrome often possess more adaptive skills and behaviors in comparison to individuals with other genetic syndromes (Di Nuovo & Buono, 2011). However, research supports that individuals with Down syndrome are more likely to exhibit noncompliance and stubbornness than those with ID alone (Coe et al., 1999; Patti & Tsiouris, 2006). Ritualistic, repetitive, and compulsive behaviors may also contribute to the behavioral phenotype of children with Down syndrome (Evans & Gray, 2000; Patti & Tsiouris, 2006). Further, there is some evidence to support that children with Down syndrome may demonstrate maladaptive behaviors due to an interaction between a behavioral phenotype of Down syndrome and environmental factors, such as family discord and parental psychopathology (Gath, 1990).

Another example of gene-environment interaction is evidenced by phenotypes in Smith-Magenis syndrome, which is a developmental disorder characterized by the deletion of a part of chromosome 17. Individuals with Smith-Magenis syndrome have ID and demonstrate a predisposition to maladaptive and disruptive behaviors, including self-injury, aggression, impulsivity, hyperactivity, and noncompliance (Dykens, 2000). Further, one study conducted with individuals diagnosed with Smith-Magenis syndrome found that environmental factors (adult attention) socially reinforced self-injury and disruptive outbursts and proposed that individuals with Smith-Magenis syndrome possess a genetic predisposition to seek social interaction as reinforcement for problem behaviors, demonstrating the risk of these phenotypes for the presence of psychological disturbance in individuals with ID (Taylor & Oliver, 2008).

The research on Prader-Willi syndrome (PWS) further supports the role that genetic syndromes play in the risk of developing psychopathology. Clarke (1998) found that individuals with PWS demonstrated a higher occurrence of psychotic symptoms (e.g., irritability, withdrawal, stereotypical behaviors, inappropriate speech, etc.) than could not reliably be attributed to the presence of intellectual disability alone. This finding was also supported by Boer et al. (2002), in which the authors postulated a genetic predisposition for affective disorders and psychosis in individuals with uniparental disomies, mutations, or deletions on chromosome 15. Verhoeven, Tuinier, and Curfs (2003) further provided evidence of a psychopathological phenotype found in individuals with PWS, finding that individuals with uniparental disomy met the criteria for psychosis, with symptoms including auditory hallucinations and paranoid ideation.

Research on behavioral phenotypes for genetic syndromes that co-occur with intellectual disabilities is currently in the early stages. Research is also limited for genetic syndromes that are characteristically rare. Despite this, it is important to consider behavioral phenotypes when assessing the risk of comorbid psychopathology. This is especially important for early intervention and treatment considerations.

Gender Differences

Gender serves as a risk factor for the development of psychopathology in individuals with ID. Similar to the general population of individuals, there are specific observed differences for risk between males and females with respect to diagnoses (Bangasser & Valentino, 2014; Lai, Lombardo, Auyeung, Chakrabarti, & Baron-Cohen, 2015; McCabe, Lansing, Garland, & Hough, 2002; Zahn-Waxler, Shirtcliff, & Marceau, 2008). Within the general population, women tend to develop internalizing disorders (such as depression and anxiety) at greater rates, while men are more likely to suffer from

externalizing disorders, such as attention deficit/hyperactivity disorder and antisocial personality disorder (Angst et al., 2002; Boyd et al., 2015; Weissman et al., 1996). There are several theories surrounding diagnostic and gender differences in typically developing individuals, including neurobiological and genetic differences and societal influences such as gender roles and societal expectations (Bangasser & Valentino, 2014; Chester et al., 2013; Lunsky, 2003). Although gender is rarely acknowledged in risk assessment for psychopathology in individuals with ID, it is an important consideration. However, research has identified several differences in psychopathology for men and women with intellectual disabilities and how these differences serve as risk factors for the development of psychopathology.

Similar to typically developing women, women with ID tend to experience affective disorders at higher levels than men with ID. In women with ID, mood disorders generally occur at rates 2–3 times higher than observed in men (Chester et al., 2013; Lunsky, 2003; Lunsky, Bradley, Gracey, Durbin, & Koegl, 2009). Although gender is a risk factor for experiencing psychological symptoms, it is important to note that environmental factors may serve to increase or decrease this likelihood. For example, social support decreases the risk that women with ID will experience mood disorders. Social support includes the perception that the individual is cared for, understood, and supported by friends, family, and others that contribute to their well-being and can include emotional and instrumental support. A lack of perceived social support is associated with depressive symptoms for women with ID (Lunsky, 2003; Lunsky & Canrinus, 2005). This relationship is similar to that found in the general population; however, the lack of social support tends to be associated with more depressive symptoms in women with ID than men with ID. Research findings indicate that women with ID are at a greater risk for developing comorbid mood disorders due to increased susceptibility to physical and sexual abuse/trauma (Barger, Wacker, Macy, & Parish, 2009; Eastgate, Van Driel, Lennox, & Scheermeyer, 2011; Lunsky, 2003). In particular, women with ID are more prone to be physically and/or sexually abused than typically developing women. Additionally, perpetrators of abuse appear to target females with ID at a higher rate than the general population without ID, as these women are apt to be vulnerable to manipulation, uneducated on sexual assault, are isolated without proper social supports in place, and are unlikely to report their abuse. Further, women with ID are more dependent on others, lack appropriate sex education, and may experience greater difficulties communicating about sexual assault than typically developing women (Eastgate et al., 2011; Sobsey, 2006; Strickler, 2001). This risk is further compounded by the existence of several barriers to services for individuals with ID and a history of abuse. These barriers include restricted access to financial resources for treatment, inability to arrange services for oneself, and the absence of clinicians with relevant training in the assessment and treatment of individuals with ID and experiences of abuse. The lack of resources for assessing and treating posttraumatic stress symptoms in women with ID is exacerbated by their difficulties understanding and interpreting traumatic experiences, especially sexual assault. As with the general population of women without ID, women with ID experience expectations to conform to biased societal standards on the role of women, including specific behaviors and identities (e.g., caretaking, motherhood, and being sociable). Further, when roles are not met or are incongruent with an individual's identity, it may lead to an increase in experiences of negative emotionality (Kreiser & White, 2014; Lunsky et al., 2009).

Males with intellectual disabilities also experience greater symptoms of maladjustment that is linked to their gender. For example, men with ID are at greater risk for developing substance abuse problems than women with ID. Research demonstrates that the prevalence of substance abuse issues in individuals with ID is approximately 2 times the non-ID population, and men with ID are 11 times more likely to develop substance abuse problems than women with ID (Chaplin, Gilvarry, & Tsakanikos, 2011; Glazier & Kling, 2013). The reasoning behind males with ID being

at increased risk is inconclusive, but studies have demonstrated evidence for several theories, including inadequate methods of coping, (Didden, Embregts, van der Toorn, & Laarhoven, 2009), increased engagement in risky behaviors (including exposure to alcohol use) for adolescent males (Reis, Wetzel, & Häßler, 2017), and living independently (Taggart, McLaughlin, Quinn, & Milligan, 2006). In addition to substance use problems, men with ID demonstrate a higher risk of developing co-occurring personality disorders, but research is limited at this time (Tsakanikos, Bouras, Sturmey, & Holt, 2006). Similar to the general population, males with ID are at a greater risk of exhibiting comorbid autism spectrum disorder (Matson & Shoemaker, 2009; Tsakanikos et al., 2006). This increased risk may be associated with phenotypic differences between males and females (e.g., higher presence of repetitive stereotyped behaviors and fixated interests), increased severity of ID, and biases that result in the disproportionate diagnosis of autism in females (Bryson, Bradley, Thompson, & Wainwright, 2008; Loomes, Hull, & Mandy, 2017; Werling & Geschwind, 2013). As in the general population of males without ID, males with ID are also at an increased risk for being diagnosed with an externalizing disorder such as attention deficit/hyperactivity disorder and oppositional defiant disorder (Witwer & Lecavalier, 2008). The risk for increased disruptive behavior in males with ID is associated with several causes, such as lower IQ, slowed development of behavioral regulation, and deficits in inhibition (Dekker & Koot, 2003; Koolhof, Loeber, Wei, Pardini, & D'escury, 2007). However, one distinct difference in individuals with ID and co-occurring disruptive behavior is that males tend to exhibit significantly less aggressive behaviors than females with ID (Cooper et al., 2009; Lundqvist, 2013). Regardless of these differences, there is ample research to support the conclusion that males with ID are at a heightened risk for developing disruptive behavior disorders.

Although the literature concerning gender differences in co-occurring psychopathology and ID has grown over the past few years, the research is limited. Future research should more fully evaluate the relationship between gender and comorbid psychopathology in individuals with intellectual disabilities.

Socioeconomic Status

Socioeconomic status is a well-established predictor for numerous negative health outcomes in typically developing individuals, as well as those with intellectual impairments. Negative health outcomes include physical health as well as psychological maladjustment (Miech, Caspi, Moffitt, Wright, & Silva, 1999). Low-socioeconomic status serves both as a risk factor and a moderator for the presence and severity of mental illness. The conditions associated with living in poverty perpetuate a negative cycle in which mental illness thrives due to increased stress, discrimination, social exclusion, and violence as well as decreased educational and occupational opportunities (Dohrenwend et al., 1992; Lund et al., 2011). The social selection theory posits that individuals may be genetically predisposed to their respective socioeconomic status; that is, healthy and able-bodied individuals are more likely to achieve or maintain a high SES, whereas unhealthy or disabled individuals experience more difficulties climbing out of poverty or will experience declines in income due to their impairments (Dohrenwend et al., 1992; Lund et al., 2010; Miech et al., 1999). Further, research indicates that individuals with intellectual disabilities are more likely to be impoverished and that poverty continues to be a risk factor for the development of psychological symptoms (Emerson & Hatton, 2007; Graham, 2005).

Like their adult counterparts, children with intellectual disabilities are at greater risk for living in an impoverished environment than their typically developing peers (Emerson, 2013; Leonard et al., 2005; Leonard & Wen, 2002). As with the general population, intellectually disabled individuals with fewer financial resources are more likely to experience negative outcomes resulting from restricted access to resources such as healthy food, adequate shelter, and sufficient

medical care. These depleted or absent resources further exacerbate the likelihood of impoverished individuals with developmental disabilities will experience mental illness. Family stress associated with poverty also is a risk factor for the development of psychopathology as children with ID require greater support and care than non-ID children. These supports are associated with diminished resources for parents and family members in terms of time, money, and other resources (Emerson, 2007). Further, parents of children with ID report increased stress, poorer health, and higher levels of caregiving burden (McIntyre, Blacher, & Baker, 2002; Olsson & Hwang, 2001). Parental stress often negatively impacts children due to diminished family functioning and coercive parenting. Consequently, children with ID are more likely to exhibit disruptive and noncompliant behavior, which further perpetuates the cycle of stress and dysfunction for caregivers. Thus, families who experience a significant burden due to the lack of financial resources may further place their children with ID at risk of psychological maladjustment.

Evidence also supports the fact that socioeconomic disadvantage in families of children with ID increases the risk of deficient parenting practices. There is a substantial amount of evidence throughout the literature that negative parenting practices, such as harsh, coercive, and inconsistent punishment, low warmth, and poor monitoring and parental involvement, increase the likelihood of both externalizing and internalizing symptoms in typically developing children (Barry, Frick, & Grafeman, 2008). Dysfunctional parenting practices are often the result of the contextual experiences of impoverished individuals that contribute to increased psychological problems such as depression, anxiety, and stress. In addition, research indicates that low SES parents are more likely to engage in harsh punishment practices, such as the use of physical discipline, and are less likely to engage in monitoring behaviors (Kotchick & Forehand, 2002). Further, the presence of parent psychopathology is common in parents of children with intellectual disabilities, due to the significant demands that caring for a child with ID places on caregivers. Experiences of psychological distress due to both parenting a child with ID and the stressful experiences of poverty further place parents in a position to develop negative punishment practices and coping skills and demonstrate poor displays of warmth and nurturance to their child (Emerson, 2004). Thus, these adverse experiences with dysfunctional parenting practices further place children with ID and low SES at increased risk of comorbid psychological disorders.

Socioeconomic disadvantage is also associated with negative physical health outcomes in both typically developing individuals and those with ID. These health outcomes include increased cardiovascular problems, poor oral health, and obesity (Chen, Martin, & Matthews, 2006; Johnson et al., 2011; Phelan, Link, & Tehranifar, 2010). A number of factors are associated with poor physical health in low SES individuals with ID including inadequate nutrition, lack of exercise, and limited access to preventative medical care. Physical health problems can lead to poor psychological health and the potential for development of comorbid mental illness in individuals with ID (Crocker, Prokić, Morin, & Reyes, 2014; Sutherland, Couch, & Iacono, 2002). This presence of mental illness in individuals with ID is especially problematic as these individuals require ongoing specialized psychological, medical, and psychopharmacological treatment, but are less likely to receive it due to barriers such as lack of financial resources, transportation, and access to services.

Although there is a large body of research dedicated to socioeconomic status and the risk it poses for the development of mental illness, this area is less established for individuals with ID. Future research should examine the relationships between poverty and negative medical and mental health outcomes in individuals with ID. Further, identifying services and resources that may buffer the impact of poverty in individuals with ID is sorely needed.

Life Events and Experiences of Trauma

Adverse life events and traumatic experiences can have a profound impact on an individual's physical and mental health. These events encompass a large variety of negative experiences, such as experiences of abuse, death of a loved one, serious illness or injury, experiencing a natural disaster, and being the victim of crime. Further, negative life events can serve as a catalyst for the development of short- and long-term mental health problems in the general population. These include depression, anxiety, disruptive behaviors, psychosis, and traumatic stress symptoms (Keller, Neale, & Kendler, 2007; Read, Bentall, & Fosse, 2009; Tiet et al., 2001; Zetterqvist, Lundh, & Svedin, 2013). Individuals with ID are more likely to experience adverse life events than the general population without intellectual disabilities (Eastgate et al., 2011; Strickler, 2001). Research on the impact of adversity and trauma on individuals with intellectual disabilities illuminates why adverse events serve as a significant risk factor in the development of psychological symptoms.

Although typically developing individuals often experience psychological problems as the result of trauma and stressful life events, there are several key differences in the impact of adversity and traumatic experience in individuals with ID. First, individuals with ID often have experiences that predispose them to the experience of stressful life events, such as living in a residential hospital, requiring extensive support for activities of daily living, and being separated from family and loved ones. Additionally, research demonstrates that both children and adults with ID experience negative life events at a greater frequency than the general population without ID (Hatton & Emerson, 2004; Hulbert-Williams & Hastings, 2008). For example, individuals with ID who reside in an assisted living facility often are physically restrained by staff, which can cause psychological distress; this rarely is experienced by individuals without intellectual disabilities (Hulbert-Williams et al., 2014). A study examining the impact of life events and traumatic experiences in individuals with ID found that 75% of participants experienced at least one negative life event at some point in their life and 50% of participants experienced at least one negative life event within the past 12 months (Martorell et al., 2009). Additionally, of these individuals who reported experiencing at least one traumatic life event, 34% of participants had a psychological disorder as classified by the ICD-10. This finding further supports that increased adverse and traumatic experiences leave individuals with ID at increased risk of mental health problems, as they exhibit deficiencies in appropriate coping mechanisms due to their diminished cognitive abilities.

As previously discussed, individuals with intellectual disabilities and a low-socioeconomic status are placed at a greater risk of developing psychological problems (Taggart et al., 2010). This is further compounded by the negative life events and trauma that co-occur with having a lower SES, such as living in a dangerous neighborhood with a high rate of crime, exposure to violence, and food insecurity and malnourishment. The combined experiences of low SES and negative life events heighten the risk of psychopathology in individuals with ID (Emerson, 2013; Hatton & Emerson, 2004). For example, Owen et al. (2004) found that adults with ID who live in a residential facility and experienced negative life events within the past year were more likely to exhibit aggressive or destructive behavior. In addition, many of these individuals were more likely to experience affective symptoms such as repetitive behavior, decreased energy, and irritability. Further, adolescents with ID who experience negative life events, such as abuse, removal from their household, and having contact with police, are more likely to demonstrate emotional and behavioral problems than their peers without intellectual disabilities (Taggart et al., 2010). Individuals with ID who experience traumatic events are at significant risk for experiencing PTS symptoms; however, these individuals often go undiagnosed and therefore remain untreated. One reason for PTS symptoms often go undetected is due in part to heavy reliance on informant reports (Wigham, Hatton, & Taylor,

2011). An overreliance on informant reports allows for errors in assessment and misattribution of symptoms. Often, informants are unaware of adverse life experiences in individuals with ID, or may possess motivation to suppress this information (e.g., in the case that the informant is also the perpetrator of abuse of an individual with ID). Accurate detection and assessment of trauma symptoms in individuals with ID is further hindered by the lack of appropriate assessment (Mevissen, Didden, & de Jongh, 2016; Wigham & Emerson, 2015).

Social factors also contribute to the role of adverse life events as a risk factor for the development of psychiatric illness in individuals with intellectual disabilities. Primarily, individuals with ID are more likely to have limited social supports in place. A large body of the research literature indicates the importance of social support on physical and mental health and the moderating role that social support plays in the development and severity of associated trauma symptoms (Dumont & Provost, 1999; Guay, Billette, & Marchand, 2006; Ozbay et al., 2007; Panagioti, Gooding, Taylor, & Tarrier, 2014). However, individuals with intellectual disabilities who experience traumatic stressors and stressful life events are less likely to have those supports in place to act as a buffer between traumatic experiences and psychological distress. As previously discussed in this chapter, individuals with ID demonstrate unique barriers that cause difficulties in establishing healthy interpersonal relationships with others (Gilmore & Cuskelly, 2014; Lunsky, 2003). Additionally, individuals with ID and low levels of social support who experience stressful life events are more likely to exhibit disruptive, destructive, and/or self-injurious behaviors, especially those with more severe levels of ID (Scott & Havercamp, 2014). This is troublesome when considering the fact that many individuals with genetic syndromes are genetically predisposed to behavioral phenotypes that render the establishment and maintenance of supportive relationships more difficult. Such is the case in individuals with fragile X syndrome, who experience deficits in social cognition such as issues with emotion perception, facial recognition, and attention switching (Cornish et al., 2005; Hall, Lightbody, Huffman, Lazzeroni, & Reiss, 2009). A genetic predisposition to impairments in social functioning further places individuals with intellectual disabilities at risk in the instance of traumatic or adverse life events.

Individuals with ID are also placed at a greater risk of developing psychopathology due to increased risks of experiencing abuse, neglect, and maltreatment by others. They are often considered "invisible victims," as their traumatic experiences are frequently unreported, disregarded, or unpursued by law enforcement. In general, both adults and children with intellectual disabilities are more vulnerable to sexual, physical, and emotional abuse and being a victim of crime than the general population without ID (Fisher, Baird, Currey, & Hodapp, 2016; Horner-Johnson & Drum, 2006; McCabe, Cummins, & Reid, 1994). This increased victimization is likely due to individuals with ID being easier targets for manipulation, and less likely to report instances of abuse as a result of limitations in communication, and difficulties recalling previous events. Further, individuals with ID may be reluctant to report instances of abuse due to previous bad experiences, such as being disregarded (Carmody, 1991). Lack of agency and control may also contribute to increased experiences of abuse and maltreatment for individuals with severe/profound ID who require greater care and support. One study conducted by Reiter, Bryen, and Shachar (2007) found that adolescents with developmental disabilities were more likely to experience physical, sexual, and emotional abuse than their nondisabled peers. In addition, the adolescents with disabilities were more likely to be physically attacked or forced to do something against their will. Interestingly, they found that disabled students were more likely to be a victim of theft, usually by a relative or similar-aged peer at home or in an assisted living facility, and they were less likely to report these incidents of theft. Reiter and colleagues also found that disabled adolescents reported increased incidents where they were forced to touch someone sexually and that they generally knew the perpetrator of such incidents (e.g., relative or peer). The increased

vulnerability to incidents of maltreatment, neglect, and abuse of individuals with ID further contributes to the increased risk that these individuals will experience psychological symptoms as the result of adverse life events.

The impact of adverse or traumatic experiences in individuals with ID is associated with greater risk of demonstrating several psychological symptoms. For instance, a study conducted by Owen et al. (2004) found that adults with ID in a residential treatment facility who experienced negative life events within the past 12 months were more likely to exhibit aggressive and/or destructive behaviors. In addition, many of these individuals were at a greater likelihood to experience symptoms of an affective/neurotic disorder, such as repetitive behaviors, decreased energy, and irritability. Research also supports that adolescents with ID who experience negative life events, such as abuse (e.g., physical, sexual, or emotional); removal from their household; and having contact with police are more likely to demonstrate emotional and behavioral problems than their peers (Taggart et al., 2010). There is also research that supports experiences of post-traumatic stress disorder as the result of traumatic experiences in individuals with ID (Turk, Robbins, & Woodhead, 2005; Wigham et al., 2011). These psychological symptoms emphasize the importance of conducting further research on the level of risk that adverse life events and trauma have on individuals with intellectual disabilities. However, it is worth noting that there are several variables that complicate research in this area. For instance, research on traumatic experiences and adverse events in individuals with ID is primarily from the prospective of informants, rather than the individual (Wigham et al., 2011). Further, individuals with ID may not possess the intellectual capacity to attribute their psychological distress to adverse events or the ability to communicate these experiences to others. Individuals with ID who experience psychopathology as the result of traumatic stress may have their experiences missed and unrecognized by clinicians who incorrectly attribute their presenting symptomology to other factors. Thus, disparities in the assessment and treatment of individuals with ID and psychological symptoms as the result of adverse life events should focus on improved methods of assessment that are sensitive to the cognitive abilities of individuals with ID.

Job Satisfaction

Vocational employment for people with intellectual disabilities has grown over the last several decades. Many attribute this increase in gainful employment for individuals with ID to legislative measures enacted in the USA, such as the Rehabilitation Act of 1973 and the Americans with Disabilities Act (ADA) of 1990. These federal laws helped to decrease the many barriers to employment that individuals with ID faced, such as discrimination based on their disability and inaccessibility in the workplace. According to the US Bureau of Labor Statistics (BLS), 18.7% of individuals with a disability were employed in the USA in 2017, and of that group, only 32% of people with a disability had a full-time job. Further, individuals with disabilities were much more likely to be unemployed than those without a disability and had a higher overall unemployment rate (9.2% and 4.2%, respectively), demonstrating that barriers to employment still remain for individuals with disabilities (US Bureau of Labor Statistics [BLS], 2017).

Despite continued obstacles to employment, individuals with ID are able to enjoy employment in a variety of settings. This includes occupations that provide more inclusive settings for individuals with ID to work alongside those without intellectual disabilities (i.e., supported employment), as well as occupations that are considered sheltered, and segregate individuals with disabilities while providing greater supervision. Some evidence in the literature supports the idea that the type of work environment can affect job satisfaction for individuals with ID. For example, a study conducted by Griffin, Rosenberg, Cheyney, and Greenberg (1996) found that individuals with ID who were employed in sheltered employment setting reported decreased self-esteem and lower levels of job satisfaction than ID workers

employed within a supported employment setting. The authors postulated that sheltered work environments likely offer less opportunities for independence and growth, as they are generally comprised of greater amounts of supervision and decreased variety in the types of work activities. Evidence within the literature demonstrates that job satisfaction is much higher for individuals with ID when the duties of a job are congruent with an employee's skills and preferences (Kocman & Weber, 2018). This is unsurprising, as duties that may be considered boring or below one's skill level can elicit feelings of frustration. Less independence and limited prospects for development as an employee are likely to contribute to negative psychological symptoms, such as symptoms of anxiety and depression (Colligan & Higgins, 2006; Faragher, Cass, & Cooper, 2013). Further, sheltered environments offer less opportunities for social interaction and opportunities to form interpersonal relationships with coworkers, particularly those without disabilities (Migliore, Mank, Grossi, & Rogan, 2007). As previously discussed in this chapter, opportunities for social interaction are incredibly important for individuals with ID, as they model appropriate social behavior and aid in the development of social skills. Decreased opportunities for socialization can further increase risk of psychopathology, as it can contribute to feelings of isolation and loneliness. However, there is research that supports increased social interaction in supported work environments may not always have a positive impact on employees with intellectual disabilities. For instance, a study of workers with ID conducted by Petrovski and Gleeson (1997) found that some workers in integrated work settings experienced discomfort and feelings of loneliness due to stigma surrounding their intellectual disability. These feelings further decreased perceived job satisfaction, as workers felt like they were "different" from their coworkers and unable to fit in.

Evidence within the research literature supports that meaningful employment provides several positive outcomes for people with intellectual disabilities. This includes increased self-esteem, enhanced quality of life, and greater psychological well-being (Jahoda, Kemp, Riddell, & Banks, 2008). However, lack of job satisfaction can serve as a risk factor for the development and presentation of psychological symptoms in persons with intellectual disabilities. The impact of job satisfaction on individuals with ID is a relatively new area of research, and research on this subject is scarce. However, there is evidence within the literature confirming that individuals with ID experience poor levels of satisfaction with their employment situation, and similar to the general population without ID, this lack of satisfaction can have several negative consequences on an individual's mental health. Future studies should also seek to examine gender differences in job satisfaction and mental health, as this area is also severely limited. There is a considerable demand for research in this area, as people with intellectual disabilities continue to enter the workforce and face difficulties that increase the chance of experiencing psychological distress.

Problem-Solving Ability

Problem-solving ability is a cognitive process in which an individual evaluates potential mechanisms in order to overcome obstacles. These skills are an important factor in fostering resilience. There is a substantial body of evidence within the literature that supports the assertion that poor or maladaptive problem-solving skills are linked to negative outcomes in one's psychological well-being. Given that individuals with intellectual disabilities generally exhibit poor problem-solving skills, it is unsurprising that deficient problem-solving abilities can serve as a risk factor in the development of psychopathology for individuals with ID.

In general, individuals with ID often demonstrate poor problem-solving abilities. This is especially the case for social problem-solving skills required to navigate interpersonal relationships (Wehmeyer & Kelchner, 1994). As discussed earlier in this chapter, individuals with ID often experience difficulties maintaining interpersonal relationships with others. Further, these

individuals are often the targets of bullying and social exclusion and are less likely to experience supportive relationships that have a buffering effect on negative experiences. Deficits in social cognition and deficient problem-solving skills further increase the risk of experiencing psychological symptoms for individuals with ID, as their predisposition to increased negative social experiences provides more opportunities to employ inappropriate or maladaptive social problem-solving skills (McGillivray & McCabe, 2007).

The absence of efficient problem-solving abilities is often exhibited in individuals with anxiety and depression (Marx, Williams, & Claridge, 1992). Research in depression for individuals with intellectual disabilities has also replicated this finding (Hartley & MacLean, 2009). However, further research is necessary to determine the distinct relationship between deficits in social problem-solving skills and depressive symptoms for individuals with ID. In particular, future studies should delineate whether the deficits in social cognition and problem-solving abilities in individuals with ID foster depressive and anxiety symptoms, or if these problem-solving skills further decline in the presence of depression and/or anxiety.

Conclusion and Future Directions

The evidence for risk factors in the development of psychopathology for individuals with intellectual disabilities is similar to the general population of individuals without intellectual disabilities. However, there are many differences for both the presentation and symptoms of psychological disorders in individuals with ID. These differences occur for a variety of reasons, including social and biological factors.

In general, the research on correlates in the level of severity of ID, and co-occurring psychopathology is in its infancy. Studies that illuminate some of the key differences in the presentation and types of mental health problems are limited in several ways, including lack of consensus on diagnosis, difficulties in assessment, and the presentation of symptoms that are distinctly different from the general population without ID. These issues highlight the importance and need for future studies to examine specific differences between severity levels, rather than clustering levels of ID into groups with inconsistent inclusion criteria. However, it is important to note that this research is hindered by the fact that a majority of individuals with ID are diagnosed with mild ID; thus, conducting studies with sufficient sample sizes of individuals with moderate, severe, and profound ID is challenging and cumbersome.

Studies on genetic factors associated with risk of psychological symptoms are also important and necessary in the prevention of dual disorders. Phenotypes associated with specific syndromes shed light on comorbidities, and may even serve as important clues in the assessment and treatment of psychological disorders in individuals with ID. Because this area of research is fairly new, studies in the future should examine behavioral phenotypes and their contribution to experiences of mental illness in individuals with ID.

As with the general population of individuals without intellectual disabilities, there are many facets of gender that are associated with the risk of dual diagnosis of disorders in individuals with ID. These differences include specific behavioral phenotypes associated with gender, gender roles imposed by the society, and increased opportunity for victimization. It is important to note that most of the research within this area is extremely limited, and the studies that have examined gender differences to date demonstrate issues in methodology, including small sample sizes, absence of segregation based on severity level of ID, and the application of controls for comorbid genetic syndromes. These issues further press the importance for future studies to examine the risk of gender in the development of psychological symptoms for individuals with ID.

The research literature demonstrates a great amount of evidence for socioeconomic status as a risk factor in the development of psychopathology. However, this area is limited to studies mostly on SES in the general population of individuals without ID. Individuals with ID are often

from a lower SES, and thus are at increased risk of developing comorbid psychological disorders. Further, these individuals are less equipped to address the many psychosocial stressors associated with a lower SES, and as a result often demonstrate maladaptive coping behaviors. Individuals with ID who are low SES may also experience greater barriers to assessment and treatment of physical and mental health problems, which further increases their risk of exhibiting a psychological disorder. This demonstrates the importance of educating low SES individuals with ID on social supports and services that they can take advantage of in order to improve health outcomes. This may include patient education on government-subsidized services or educating caretakers on services available for individuals with ID. Utilization of services designed for low SES individuals can decrease the burden and stress placed on those with ID and a low SES.

Individuals with ID tend to undergo adverse life events and traumatic experiences at rates much higher than those without ID. In general, individuals with ID are at an increased risk of exposure to adverse and traumatic events, much higher than the general population. This increased risk is often linked to specific characteristics of individuals with ID, such as lack of agency or cognitive functioning that leaves them vulnerable to victimization and experiences of trauma. Individuals with ID are also at an increased risk of experiencing adverse life events due to the increased likelihood of individuals with ID having a low SES and the several types of negative life experiences that often occur within the context of low SES households. Social factors, such as limited social support and lack of social relationships, also contribute to the risk of experiencing trauma and adverse life events in individuals with ID, as these factors often assist in the development of appropriate coping skills and resilience. Further, the risk of adversity is also complicated by the lack of appropriate assessment measures for PTSD designed specifically for use in ID populations. Assessment also tends to rely on informant reports, which may cause symptoms to go undetected or misattributed to other causes. As such, individuals with ID experience psychological distress as the result of adversity, but these symptoms often go undetected, or are misattributed to other causes.

Lack of job satisfaction is also a risk factor in the dual diagnosis of psychopathology in individuals with intellectual disabilities. While society has made progress toward inclusive employment for individuals with ID, many of these individuals continue to experience discrimination that can result in the lack of satisfaction at work. Further, individuals with ID who work within environments that limit their opportunity for occupational growth can be at a greater risk of experiencing symptoms of psychological distress. Employment duties that aren't congruent with an individual's skills may also contribute to feelings of job dissatisfaction.

Individuals with ID may also be placed at a greater risk for psychopathology when their work environment has decreased opportunities for socialization with others, as it can foster feelings of isolation and loneliness. While there is a large body of research for the impact of job dissatisfaction on mental health, and the risk it places on the development of psychopathology in the general population, this area of research is very limited within the ID population. Thus, further research is necessary to assess the risk of job dissatisfaction on psychological disorders in individuals with ID.

Research indicates that individuals with ID exhibit decreased problem-solving abilities in comparison to those without ID. This places individuals with ID at an increased risk of experiencing psychiatric symptoms, as poor or maladaptive problem-solving skills can cause negative mental health outcomes. In particular, a lack of social problem-solving skills necessary in the formation and maintenance of interpersonal relationships can serve as a risk factor for the development of psychological symptoms, as impaired relationships can cause significant distress. This risk can be further increased by decreased opportunities for the modeling of appropriate social skills and social interactions.

As with the research literature on dual diagnosis in general, the research literature on the risk factors for dual diagnosis in individuals with ID is

relatively young. However, the gaps within the literature should be addressed, as the presence of psychopathology in individuals with ID is much higher than in the population of individuals without ID. Further, identification of risk factors is an important step in determining appropriate procedures for both assessment and treatment of comorbid psychopathology. Thus, future research should seek to identify the many risk factors that contribute to dual diagnosis in individuals with ID.

References

Ali, Z. (2001). Pica in people with intellectual disability: A literature review of aetiology, epidemiology and complications. *Journal of Intellectual and Developmental Disability, 26*(3), 205–215.

American Psychiatric Association. (2013). *Diagnostic and statistical manual of mental disorders* (5th ed.). Arlington, VA: American Psychiatric Publishing.

Angst, J., Gamma, A., Gastpar, M., Lépine, J. P., Mendlewicz, J., & Tylee, A. (2002). Gender differences in depression. *European Archives of Psychiatry and Clinical Neuroscience, 252*(5), 201–209.

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*(3), 176–190.

Axmon, A., Björne, P., Nylander, L., & Ahlström, G. (2017). Psychiatric diagnoses in relation to severity of intellectual disability and challenging behaviors: A register study among older people. *Aging & Mental Health, 22*(10), 1344–1350.

Bangasser, D. A., & Valentino, R. J. (2014). Sex differences in stress-related psychiatric disorders: Neurobiological perspectives. *Frontiers in Neuroendocrinology, 35*(3), 303–319.

Barger, E., Wacker, J., Macy, R., & Parish, S. (2009). Sexual assault prevention for women with intellectual disabilities: A critical review of the evidence. *Intellectual and Developmental Disabilities, 47*(4), 249–262.

Barry, C. T., Frick, P. J., & Grafeman, S. J. (2008). Child versus parent reports of parenting practices: Implications for the conceptualization of child behavioral and emotional problems. *Assessment, 15*(3), 294–303.

Bellanti, C. J., & Bierman, K. L. (2000). Disentangling the impact of low cognitive ability and inattention on social behavior and peer relationships. *Journal of Clinical Child Psychology, 29*(1), 66–75.

Boer, H., Holland, A., Whittington, J., Butler, J., Webb, T., & Clarke, D. (2002). Psychotic illness in people with Prader Willi syndrome due to chromosome 15 maternal uniparental disomy. *The Lancet, 359*(9301), 135–136.

Boyd, A., Van de Velde, S., Vilagut, G., De Graaf, R., Florescu, S., Alonso, J., … Investigators, E. U.-W. M. H. (2015). Gender differences in mental disorders and suicidality in Europe: Results from a large cross-sectional population-based study. *Journal of Affective Disorders, 173*, 245–254.

Bradley, E. A., Summers, J. A., Wood, H. L., & Bryson, S. E. (2004). Comparing rates of psychiatric and behavior disorders in adolescents and young adults with severe intellectual disability with and without autism. *Journal of Autism and Developmental Disorders, 34*(2), 151–161.

Bryson, S. E., Bradley, E. A., Thompson, A., & Wainwright, A. (2008). Prevalence of autism among adolescents with intellectual disabilities. *The Canadian Journal of Psychiatry, 53*(7), 449–459.

Carmody, M. (1991). Invisible victims: Sexual assault of people with an intellectual disability. *Australia and New Zealand Journal of Developmental Disabilities, 17*(2), 229–236.

Chadwick, O., Kusel, Y., & Cuddy, M. (2008). Factors associated with the risk of behavior problems in adolescents with severe intellectual disabilities. *Journal of Intellectual Disability Research, 52*(10), 864–876.

Chaplin, E., Gilvarry, C., & Tsakanikos, E. (2011). Recreational substance use patterns and comorbid psychopathology in adults with intellectual disability. *Research in Developmental Disabilities, 32*(6), 2981–2986.

Chen, E., Martin, A. D., & Matthews, K. A. (2006). Understanding health disparities: The role of race and socioeconomic status in children's health. *American Journal of Public Health, 96*(4), 702–708.

Chester, R., Chaplin, E., Tsakanikos, E., McCarthy, J., Bouras, N., & Craig, T. (2013). Gender differences in self-reported symptoms of depression and anxiety in adults with intellectual disabilities. *Advances in Mental Health and Intellectual Disabilities, 7*(4), 191–200.

Clarke, D. (1998). Prader–Willi syndrome and psychotic symptoms: 2. A preliminary study of prevalence using the psychopathology assessment schedule for adults with developmental disability checklist. *Journal of Intellectual Disability Research, 42*(6), 451–454.

Coe, D. A., Matson, J. L., Russell, D. W., Slifer, K. J., Capone, G. T., Baglio, C., & Stallings, S. (1999). Behavior problems of children with Down syndrome and life events. *Journal of Autism and Developmental Disorders, 29*(2), 149–156.

Colligan, T. W., & Higgins, E. M. (2006). Workplace stress: Etiology and consequences. *Journal of Workplace Behavioral Health, 21*(2), 89–97.

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.

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.

Cornish, K., Kogan, C., Turk, J., Manly, T., James, N., Mills, A., & Dalton, A. (2005). The emerging fragile X premutation phenotype: Evidence from the domain of social cognition. *Brain and Cognition, 57*(1), 53–60.

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.

Dagnan, D., & Sandhu, S. (1999). Social comparison, self-esteem and depression in people with intellectual disability. *Journal of Intellectual Disability Research, 43*(5), 372–379.

Davies, L. E., & Oliver, C. (2016). Self-injury, aggression and destruction in children with severe intellectual disability: Incidence, persistence and novel, predictive behavioral risk markers. *Research in Developmental Disabilities, 49*, 291–301.

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. II: Child and family predictors. *Journal of the American Academy of Child & Adolescent Psychiatry, 42*(8), 923–931.

Di Nuovo, S., & Buono, S. (2011). Behavioral phenotypes of genetic syndromes with intellectual disability: Comparison of adaptive profiles. *Psychiatry Research, 189*(3), 440–445.

Didden, R., Embregts, P., van der Toorn, M., & Laarhoven, N. (2009). Substance abuse, coping strategies, adaptive skills and behavior and emotional problems in clients with mild to borderline intellectual disability admitted to a treatment facility: A pilot study. *Research in Developmental Disabilities, 30*(5), 927–932.

Dohrenwend, B. P., Levav, I., Shrout, P. E., Schwartz, S., Naveh, G., Link, B. G., … Stueve, A. (1992). Socioeconomic status and psychiatric disorders: The causation-selection issue. *Science, 255*(5047), 946–952.

Dumont, M., & Provost, M. A. (1999). Resilience in adolescents: Protective role of social support, coping strategies, self-esteem, and social activities on experience of stress and depression. *Journal of Youth and Adolescence, 28*(3), 343–363.

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.

Dykens, E. M., & Kasari, C. (1997). Maladaptive behavior in children with Prader-Willi syndrome, Down syndrome, and non-specific mental retardation. *American Journal on Mental Retardation, 102*(3), 228–237.

Eastgate, G., Van Driel, M. L., Lennox, N., & Scheermeyer, E. (2011). Women with intellectual disabilities: A study of sexuality, sexual abuse and protection skills. *Australian Family Physician, 40*(4), 226.

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.

Einfeld, S. L., Piccinin, A. M., Mackinnon, A., Hofer, S. M., Taffe, J., Gray, K. M., … Tonge, B. J. (2006). Psychopathology in young people with intellectual disability. *JAMA, 296*(16), 1981–1989.

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*(9), 657–671.

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.

Emerson, E. (2004). Poverty and children with intellectual disabilities in the world's richer countries. *Journal of Intellectual and Developmental Disability, 29*(4), 319–338.

Emerson, E. (2007). Poverty and people with intellectual disabilities. *Mental Retardation and Developmental Disabilities Research Reviews, 13*(2), 107–113.

Emerson, E. (2013). Commentary: Childhood exposure to environmental adversity and the well-being of people with intellectual disabilities. *Journal of Intellectual Disability Research, 57*(7), 589–600.

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.

Evans, D. W., & Gray, F. L. (2000). Compulsive-like behavior in individuals with Down syndrome: Its relation to mental age level, adaptive and maladaptive behavior. *Child Development, 71*(2), 288–300.

Faragher, E. B., Cass, M., & Cooper, C. L. (2013). The relationship between job satisfaction and health: A meta-analysis. In C. L. Cooper (Ed.), *From stress to wellbeing* (Vol. 1, pp. 254–271). Berlin: Springer.

Fisher, M. H., Baird, J. V., Currey, A. D., & Hodapp, R. M. (2016). Victimisation and social vulnerability of adults with intellectual disability: A review of research extending beyond Wilson and Brewer. *Australian Psychologist, 51*(2), 114–127.

Furniss, F., & Biswas, A. B. (2012). Recent research on aetiology, development and phenomenology of self-injurious behaviour in people with intellectual disabilities: A systematic review and implications for treatment. *Journal of Intellectual Disability Research, 56*(5), 453–475.

Gath, A. (1990). Down syndrome children and their families. *American Journal of Medical Genetics, 37*(S7), 314–316.

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.

Glazier, R. E., & Kling, R. N. (2013). Recent trends in substance abuse among persons with disabilities compared to that of persons without disabilities. *Disability and Health Journal, 6*(2), 107–115.

Gotlib, I. H., & Joormann, J. (2010). Cognition and depression: Current status and future directions. *Annual Review of Clinical Psychology, 6*, 285–312.

Graham, H. (2005). Intellectual disabilities and socioeconomic inequalities in health: An overview of research. *Journal of Applied Research in Intellectual Disabilities, 18*(2), 101–111.

Griffin, D. K., Rosenberg, H., Cheyney, W., & Greenberg, B. (1996). A comparison of self esteem and job satisfaction of adults with mild mental retardation in sheltered workshops and supported employment. *Education and Training in Mental Retardation and Developmental Disabilities, 31*(2), 142–150.

Guay, S., Billette, V., & Marchand, A. (2006). Exploring the links between posttraumatic stress disorder and social support: Processes and potential research avenues. *Journal of Traumatic Stress: Official Publication of The International Society for Traumatic Stress Studies, 19*(3), 327–338.

Hall, S. S., Lightbody, A. A., Huffman, L. C., Lazzeroni, L. C., & Reiss, A. L. (2009). Physiological correlates of social avoidance behavior in children and adolescents with fragile X syndrome. *Journal of the American Academy of Child & Adolescent Psychiatry, 48*(3), 320–329.

Handley, L., Adams, D., Simkiss, D., & Oliver, C. (2013). Self-injurious, aggressive and destructive behaviour in young children with a moderate to profound intellectual disability. *Paediatrics and Child Health, 23*, 322–324.

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

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.

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.

Heiman, T. (2001). Depressive mood in students with mild intellectual disability: Students' reports and teachers' evaluations. *Journal of Intellectual Disability Research, 45*(6), 526–534.

Horner-Johnson, W., & Drum, C. E. (2006). Prevalence of maltreatment of people with intellectual disabilities: A review of recently published research. *Mental Retardation and Developmental Disabilities Research Reviews, 12*(1), 57–69.

Hulbert-Williams, L., Hastings, R., Owen, D. M., Burns, L., Day, J., Mulligan, J., & Noone, S. J. (2014). Exposure to life events as a risk factor for psychological problems in adults with intellectual disabilities: A longitudinal design. *Journal of Intellectual Disability Research, 58*(1), 48–60.

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.

Hurley, A. D. (2006). Mood disorders in intellectual disability. *Current Opinion in Psychiatry, 19*(5), 465–469.

Jahoda, A., Kemp, J., Riddell, S., & Banks, P. (2008). Feelings about work: A review of the socio-emotional impact of supported employment on people with intellectual disabilities. *Journal of Applied Research in Intellectual Disabilities, 21*(1), 1–18.

Johnson, N. W., Warnakulasuriya, S., Gupta, P. C., Dimba, E., Chindia, M., Otoh, E. C., … Kowalski, L. (2011). Global oral health inequalities in incidence and outcomes for oral cancer: Causes and solutions. *Advances in Dental Research, 23*(2), 237–246.

Keller, M. C., Neale, M. C., & Kendler, K. S. (2007). Association of different adverse life events with distinct patterns of depressive symptoms. *American Journal of Psychiatry, 164*(10), 1521–1529.

Kocman, A., & Weber, G. (2018). Job satisfaction, quality of work life and work motivation in employees with intellectual disability: A systematic review. *Journal of Applied Research in Intellectual Disabilities, 31*(1), 1–22.

Koolhof, R., Loeber, R., Wei, E. H., Pardini, D., & D'escury, A. C. (2007). Inhibition deficits of serious delinquent boys of low intelligence. *Criminal Behaviour and Mental Health, 17*(5), 274–292.

Koskentausta, T., Iivanainen, M., & Almqvist, F. (2007). Risk factors for psychiatric disturbance in children with intellectual disability. *Journal of Intellectual Disability Research, 51*(1), 43–53.

Kotchick, B. A., & Forehand, R. (2002). Putting parenting in perspective: A discussion of the contextual factors that shape parenting practices. *Journal of Child and Family Studies, 11*(3), 255–269.

Kreiser, N. L., & White, S. W. (2014). ASD in females: Are we overstating the gender difference in diagnosis? *Clinical Child and Family Psychology Review, 17*(1), 67–84.

Lai, M. C., Lombardo, M. V., Auyeung, B., Chakrabarti, B., & Baron-Cohen, S. (2015). Sex/gender differences and autism: Setting the scene for future research. *Journal of the American Academy of Child & Adolescent Psychiatry, 54*(1), 11–24.

Leonard, H., Petterson, B., De Klerk, N., Zubrick, S. R., Glasson, E., Sanders, R., & Bower, C. (2005). Association of sociodemographic characteristics of children with intellectual disability in Western Australia. *Social Science & Medicine, 60*(7), 1499–1513.

Leonard, H., & Wen, X. (2002). The epidemiology of mental retardation: Challenges and opportunities in the new millennium. *Mental Retardation and Developmental Disabilities Research Reviews, 8*(3), 117–134.

Linville, P. (1987). Self-complexity as a cognitive buffer against stress-related illness and depression. *Journal of Personality and Social Psychology, 52*, 663–676.

Loomes, R., Hull, L., & Mandy, W. P. L. (2017). What is the male-to-female ratio in autism spectrum disorder? A systematic review and meta-analysis. *Journal of the American Academy of Child & Adolescent Psychiatry, 56*(6), 466–474.

LoVullo, S. V., & Matson, J. L. (2009). Comorbid psychopathology in adults with autism spectrum disorders and intellectual disabilities. *Research in Developmental Disabilities, 30*(6), 1288–1296.

Lund, C., Breen, A., Flisher, A. J., Kakuma, R., Corrigall, J., Joska, J. A., … Patel, V. (2010). Poverty and common mental disorders in low and middle income countries: A systematic review. *Social Science & Medicine, 71*(3), 517–528.

Lund, C., De Silva, M., Plagerson, S., Cooper, S., Chisholm, D., Das, J., … Patel, V. (2011). Poverty and mental disorders: Breaking the cycle in low-income and middle-income countries. *The Lancet, 378*(9801), 1502–1514.

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.

Lunsky, Y. (2003). Depressive symptoms in intellectual disability: Does gender play a role? *Journal of Intellectual Disability Research, 47*(6), 417–427.

Lunsky, Y., Bradley, E. A., Gracey, C. D., Durbin, J., & Koegl, C. (2009). Gender differences in psychiatric diagnoses among inpatients with and without intellectual disabilities. *American Journal on Intellectual and Developmental Disabilities, 114*(1), 52–60.

Lunsky, Y., & Canrinus, M. (2005). Gender issues, mental retardation and depression. In P. Sturmey (Ed.), *Mood disorders in people with mental retardation* (pp. 113–130). Kingston, NY: NADD Press.

Martorell, A., Tsakanikos, E., Pereda, A., Gutiérrez-Recacha, P., Bouras, N., & Ayuso-Mateos, J. L. (2009). Mental health in adults with mild and moderate intellectual disabilities: The role of recent life events and traumatic experiences across the life span. *The Journal of Nervous and Mental Disease, 197*(3), 182–186.

Marx, E. M., Williams, J. M., & Claridge, G. C. (1992). Depression and social problem solving. *Journal of Abnormal Psychology, 101*(1), 78–86. https://doi.org/10.1037//0021-843x.101.1.78

Matson, J., & Frame, C. (1985). *Psychopathology among mentally retarded children and adolescents*. Beverly Hills, CA: Sage.

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*(4), 1459–1464.

Matson, J. L., & Minshawi, N. F. (2007). Functional assessment of challenging behavior: Toward a strategy for applied settings. *Research in Developmental Disabilities, 28*(4), 353–361.

Matson, J. L., & Shoemaker, M. (2009). Intellectual disability and its relationship to autism spectrum disorders. *Research in Developmental Disabilities, 30*(6), 1107–1114.

Matson, J. L., Smiroldo, B. B., & Bamburg, J. W. (1998). The relationship of social skills to psychopathology for individuals with severe or profound mental retardation. *Journal of Intellectual Disabilities Research, 23*(2), 137–145.

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.

McCabe, K. M., Lansing, A. E., Garland, A. N. N., & Hough, R. (2002). Gender differences in psychopathology, functional impairment, and familial risk factors among adjudicated delinquents. *Journal of the American Academy of Child & Adolescent Psychiatry, 41*(7), 860–867.

McCabe, M. P., Cummins, R. A., & Reid, S. B. (1994). An empirical study of the sexual abuse of people with intellectual disability. *Sexuality and Disability, 12*(4), 297–306.

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*(1), 59–70.

McIntyre, L. L., Blacher, J., & Baker, B. L. (2002). Behaviour/mental health problems in young adults with intellectual disability: The impact on families. *Journal of Intellectual Disability Research, 46*(3), 239–249.

Mevissen, L., Didden, R., & de Jongh, A. (2016). Assessment and treatment of PTSD in people with intellectual disabilities. In C. R. Martin, V. R. Preedy, & V. B. Patel (Eds.), *Comprehensive guide to post-traumatic stress disorders* (pp. 281–299). Cham: Springer.

Miech, R. A., Caspi, A., Moffitt, T. E., Wright, B. R. E., & Silva, P. A. (1999). Low socioeconomic status and mental disorders: A longitudinal study of selection and causation during young adulthood. *American Journal of Sociology, 104*(4), 1096–1131.

Migliore, A., Mank, D., Grossi, T., & Rogan, P. (2007). Integrated employment or sheltered workshops: Preferences of adults with intellectual disabilities, their families, and staff. *Journal of Vocational Rehabilitation, 26*(1), 5–19.

Moss, S. (2001). Psychiatric disorders in adults with mental retardation. *International Review of Research in Mental Retardation, 24*, 211–243.

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.

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.

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), 35–40.

Panagioti, M., Gooding, P. A., Taylor, P. J., & Tarrier, N. (2014). Perceived social support buffers the impact of PTSD symptoms on suicidal behavior: Implications into suicide resilience research. *Comprehensive Psychiatry, 55*(1), 104–112.

Patti, P. J., & Tsiouris, J. A. (2006). Psychopathology in adults with down syndrome: Clinical findings from an outpatient clinic. *International Journal on Disability and Human Development, 5*(4), 357–364.

Petrovski, P., & Gleeson, G. (1997). The relationship between job satisfaction and psychological health in people with an intellectual disability in competitive employment. *Journal of Intellectual and Developmental Disability, 22*(3), 199–211.

Phelan, J. C., Link, B. G., & Tehranifar, P. (2010). Social conditions as fundamental causes of health inequalities: Theory, evidence, and policy implications. *Journal of Health and Social Behavior, 51*(S), S28–S40.

Read, J., Bentall, R. P., & Fosse, R. (2009). Time to abandon the bio-bio-bio model of psychosis: Exploring the epigenetic and psychological mechanisms by which adverse life events lead to psychotic symptoms. *Epidemiology and Psychiatric Sciences, 18*(4), 299–310.

Reis, O., Wetzel, B., & Häßler, 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.

Reiter, S., Bryen, D. N., & Shachar, I. (2007). Adolescents with intellectual disabilities as victims of abuse. *Journal of Intellectual Disabilities, 11*(4), 371–387.

Rzepecka, H., McKenzie, K., McClure, I., & Murphy, S. (2011). Sleep, anxiety and challenging behaviour in children with intellectual disability and/or autism spectrum disorder. *Research in Developmental Disabilities, 32*(6), 2758–2766.

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.

Sobsey, D. (2006). Violence & disability. In W. M. Nehring (Ed.), *Health promotion for persons with intellectual/developmental disabilities: The state of scientific evidence*. Washington, DC: American Association on Mental Retardation.

Strickler, H. L. (2001). Interaction between family violence and mental retardation. *Mental Retardation, 39*(6), 461–471.

Sutherland, G., Couch, M. A., & Iacono, T. (2002). Health issues for adults with developmental disability. *Research in Developmental Disabilities, 23*(6), 422–445.

Swift, I., Paquette, D., Davison, K., & Saeed, H. (1999). Pica and trace metal deficiencies in adults with developmental disabilities. *The British Journal of Development Disabilities, 45*(89), 111–117.

Taggart, L., McLaughlin, D., Quinn, B., & Milligan, V. (2006). An exploration of substance misuse in people with intellectual disabilities. *Journal of Intellectual Disability Research, 50*(8), 588–597.

Taggart, L., Taylor, D., & McCrum-Gardner, E. (2010). Individual, life events, family and socio-economic factors associated with young people with intellectual disability and with and without behavioural/emotional problems. *Journal of Intellectual Disabilities, 14*(4), 267–288.

Taylor, L., & Oliver, C. (2008). The behavioural phenotype of Smith–Magenis syndrome: Evidence for a gene–environment interaction. *Journal of Intellectual Disability Research, 52*(10), 830–841.

Tiet, Q. Q., Bird, H. R., Hoven, C. W., Moore, R., Wu, P., Wicks, J., … Cohen, P. (2001). Relationship between specific adverse life events and psychiatric disorders. *Journal of Abnormal Child Psychology, 29*(2), 153–164.

Tsakanikos, E., Bouras, N., Sturmey, P., & Holt, G. (2006). Psychiatric co-morbidity and gender differences in intellectual disability. *Journal of Intellectual Disability Research, 50*(8), 582–587.

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.

Tureck, K., Matson, J. L., & Beighley, J. S. (2013). An investigation of self-injurious behaviors in adults with severe intellectual disabilities. *Research in Developmental Disabilities, 34*(9), 2469–2474.

Turk, J., Robbins, I., & Woodhead, M. (2005). Post-traumatic stress disorder in young people with intellectual disability. *Journal of Intellectual Disability Research, 49*(11), 872–875.

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.

U.S. Bureau of Labor Statistics (BLS). (2017). *Persons with a disability: Labor force characteristics – 2017*. U.S. Department of Labor (DOL), USDL-18-1028. www.bls.gov/news.release/pdf/disabl.pdf.

Verhoeven, W. M. A., Tuinier, S., & Curfs, L. M. G. (2003). Prader-Willi syndrome: The psychopatho-

logical phenotype in uniparental disomy. *Journal of Medical Genetics, 40*(10), e112.

Wehmeyer, M. L., & Kelchner, K. (1994). Interpersonal cognitive problem-solving skills of individuals with mental retardation. *Education and Training in Mental Retardation and Developmental Disabilities, 29*(4), 265–278.

Weissman, M. M., Bland, R. C., Canino, G. J., Faravelli, C., Greenwald, S., Hwu, H. G., … Lépine, J. P. (1996). Cross-national epidemiology of major depression and bipolar disorder. *JAMA, 276*(4), 293–299.

Werling, D. M., & Geschwind, D. H. (2013). Sex differences in autism spectrum disorders. *Current Opinion in Neurology, 26*(2), 146.

Wigham, S., & Emerson, E. (2015). Trauma and life events in adults with intellectual disability. *Current Developmental Disorders Reports, 2*(2), 93–99.

Wigham, S., Hatton, C., & Taylor, J. L. (2011). The effects of traumatizing life events on people with intellectual disabilities: A systematic review. *Journal of Mental Health Research in Intellectual Disabilities, 4*(1), 19–39.

Witwer, A. N., & Lecavalier, L. (2008). Psychopathology in children with intellectual disability: Risk markers and correlates. *Journal of Mental Health Research in Intellectual Disabilities, 1*(2), 75–96.

Zahn-Waxler, C., Shirtcliff, E. A., & Marceau, K. (2008). Disorders of childhood and adolescence: Gender and psychopathology. *Annual Review of Clinical Psychology, 4*, 275–303.

Zetterqvist, M., Lundh, L. G., & Svedin, C. G. (2013). A comparison of adolescents engaging in self-injurious behaviors with and without suicidal intent: Self-reported experiences of adverse life events and trauma symptoms. *Journal of Youth and Adolescence, 42*(8), 1257–1272.

9 Interviewing and Report Writing for Persons with Dual Diagnosis

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Introduction

The purpose of this chapter is to discuss critical elements of interviewing and report writing when conducting assessments for individuals with intellectual and developmental disabilities (IDDs). A primary focus is on comorbid concerns and diagnoses. The first section of this chapter covers considerations for interviewing individuals with IDDs as well as parents, caregivers, or staff members. The focus is on semi-structured interviews, as information on structured interviews and measures are discussed in subsequent chapters in this volume. The second section discusses report writing and critical elements of the report to inform care.

Theoretical approach to assessment is a complex and nuanced concept. However, for the purposes of this chapter and given the population under consideration, one model that is commonly used to gather client information in clinical assessment of individuals with intellectual disabilities as well as in educational environments is the Review, Interview, Observe, and Test (RIOT) model (retrieved from Wright, 2010). The RIOT model guides collection of information from multiple sources. The first of these sources includes a review of existing information from records regarding the client. This may include any medical history, past psychological evaluations, records of any accommodations or services received, and behavioral data records, among other resources. Next, the interview step involves gathering information directly from multiple sources, including the client themselves, caregivers such as parents or direct care staff, and any other person involved in the client's care. Interview approaches include structured, semi-structured, or open-ended interviews. This step is of primary interest in this chapter. In addition to the review of records and interviews, direct observation of the client is necessary. This may occur in a variety of contexts, though ideally observation in a more natural environment (e.g., home, school, day programming) may yield more relevant data. Observations consist of structured observations, such as data collection challenging behaviors, which may include data such as frequency, duration, topography, antecedents, and consequences, or less structured, such as general notes regarding the individual's behavior and affect. Lastly, formal testing with standardized measures is typically included. While tests relevant to an intellectual disability (ID) diagnosis, such as intelligence and adaptive measures, are likely included, diagnosis of comorbid concerns

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would include a broader range of measures. Clinicians should be deliberate in selecting measures appropriate for the developmental level of the individual.

All of these components of assessment are necessary; however, this chapter focuses primarily on the interview section, as well as how to integrate all of the information gathered into a comprehensive report that answers the referral question, provides relevant information, creates a record of the assessment for future providers, and provides and informs treatment recommendations.

Ethics and Consent

Prior to collecting collateral and historical information about a client, the clinician must consider the ethical implications of contacting outside providers and family members, as well as what information is necessary and appropriate to include in a report. This should be discussed with the client and/or guardian, and specific consent forms and releases of information should be signed by the client and/or guardian to ensure they consent to the clinician contacting family members, teachers, or outpatient providers (Cameron & Shepel, 1981).

For individuals with ID, their legal and mental capacity is relevant to the consent process. The legal capacity of the individual indicates "a person's power or responsibility to act within the framework of the legal system" while mental capacity is "the decision-making skills of a person" (McSherry, 2015). Mental capacity includes understanding and appreciation, in addition to reasoning and choice (Grisso, Grisso, & Appelbaum, 1998). These considerations determine whether the individual can consent to the assessment or whether a legal guardian must provide consent. Clinicians should be aware of the right to self-determination, referring to one's right to act as their primary causal agent by exerting control over their lives and acting on their own will (Loman, Vatland, Strickland-Cohen, Horner, & Walker, 2010). There is a great deal of controversy over this concept as self-determination is often refused due to disability status rather than individual consideration of each case (Skarstad, 2018).

When preparing each section of a report, overall considerations involve the ethical responsibility of the clinician, including beneficence, fidelity and responsibility, and respect for people's rights and dignity (American Psychological Association, 2002). The terminology in a written report should always uphold the dignity of the individual, and stigmatizing language should be avoided.

Interviewing

Clinical Skills and Rapport

In a psychological interview setting, therapeutic skills are crucial to effectively work with a client. This includes skills such as active listening, empathy, collaborative attitude, creating a client/clinician alliance, goal consensus, positive regard, and collecting feedback (Hilsenroth & Cromer, 2007).

When interviewing an informant, the clinician should consider the wording of the questions and the informant's emotional state (Adams & Boyd, 2010). The clinician must ensure that the informant is aware of and understands the purpose of the interview and overall assessment. When explaining the purpose of the interview, the interviewer should use "language that is reasonable and understandable to the person being assessed" (American Psychological Association, 2002). For individuals with intellectual disability, the structure of the questions and the content or context of the questions being asked may need to be modified depending on the individual's receptive language abilities. Therefore, when explaining the purpose of the interview to the client, simple and succinct language should be used (Adams & Boyd, 2010; Fisher, 2003; Perry, 2004).

Informants

Before conducting an interview, the assessor should identify the informant(s) that will be contributing to the assessment. Informants may include the individual with an intellectual dis-

ability being assessed, depending on their expressive and receptive communication skills, as well as family members, caregivers, staff, key support persons chosen by the individual, and other professionals. Informants should be chosen carefully, and the clinician should consider the informant's role in the individual's life. Information and symptomology collected should also be evaluated within the context of the informant's role. In particular, the assessor should consider how the informant's role may influence reports of the individual's abilities, needs, and overall lifestyle. This provides an understanding of the type of key information they are able to contribute to the evaluation (i.e., teachers can offer a school setting perspective, non-immediate family members offer outside perspective of family unit, and possible behaviors when the individual is away from primary caregivers; Carnaby, 2007).

During the assessment process, data collection from interviews is largely influenced by reports of the caregiver as the informant. Using the caregiver as the informant in the interview process often depends on the individual's cognitive ability (Charlot, Deutsch, Hunt, Fletcher, & Mcllvane, 2007). However, according to Emerson, Hatton, Dickson, Gone, and Caine (2012), if interviewing is inclusive and individuals are involved in self-report, they are more likely to receive treatment more quickly, are more successfully supported, and show higher rates of client satisfaction. Therefore, before conducting the interview, the interviewer should always consider the individual's level of functioning, communication skills, and verbal ability to determine and maximize the extent to which the client can participate in the interview process (Adams & Boyd, 2010; Fisher, 2003; Perry, 2004).

Once the degree to which the individual is able to participate in the interview is determined, the interviewer should accommodate the individual's needs and adapt the interview wherever possible. While some individuals with intellectual disability may have limited ability to describe internal states of feelings and emotions, maximizing the opportunities for individuals to self-report can increase quality of information being reported and decrease the reliance on behavioral observations. Adjustment of the interview could include adapting the interview location, changing the order of assessment procedures, and adjusting the question style/format. For example, interviewing in a confidential and familiar place may be helpful in building rapport and making the individual feel comfortable (Emerson et al., 2012; Groth-Marnat, 2003; Perry, 2004). In addition, when asking individuals with suspected or known intellectual disability questions, the interviewer should speak direct, in short segments, and use simple wording to account for difficulties with receptive and expressive language abilities (Adams & Boyd, 2010; Emerson et al., 2012; Perry, 2004). Using close-ended questions (e.g., yes/no, multiple choice, and either/or formats) may increase responsiveness and consume less time. However, open-ended questions allow the individual to answer freely, which may yield new information, but can decrease responsiveness due to complexity and be more time consuming. It is important that the clinician weighs the advantages and disadvantages of each question style/format and appropriately adapt the interview to obtain necessary information for the assessment (Perry, 2004).

It is also important to consider that individuals with intellectual disability may have a tendency to comply with the preconceptions of others, a bias called acquiescence, which can interfere with reliable and accurate reporting. To help protect against acquiescence, interviewers should avoid leading and suggestive questions (e.g., "You're upset today, aren't you?"; "You feel happy today, right?"), and focus on more open-ended and close-ended questions that challenge the tendency of response bias (e.g., Are you sure?; Emerson et al., 2012; Finlay & Lyons, 2002; Perry, 2004).

If the client is unable to participate in an interview independently, respect should be demonstrated by having the individual participate in questions or activities that are meaningful to the individual (Carnaby, 2007). Conducting a joint interview with both the individual and their caregiver or a key informant of their choice promotes autonomy and self-advocacy due to the continued

inclusion of the individual (Arksey, 1996). For example, although having an additional informant present can be helpful, any informants other than the individual being assessed should refrain for answering questions unless necessary. This example of autonomy and promotion of self-advocacy demonstrates ethical responsibilities, which should always be considered when working with individuals with intellectual disability (Adams & Boyd, 2010; Carnaby, 2007; Emerson et al., 2012; Perry, 2004).

In addition to having the individual join in preferred parts of the interview, the dyadic interviewing technique can also be considered. In dyadic interviewing, the interview process combines both individual and joint interviews with the client and a key informant chosen by the client (Caldwell, 2014). After building rapport, the individual meets one-on-one with the interviewer, then with both the interviewer and key informant, and once again one-on-one with the interviewer for the last interview. Dyadic interviewing is a developing interview approach that enables individuals with intellectual disability to respond and self-advocate for their own perspective, while still involving a key support informant of their choice (Caldwell, 2014). The alternating individual and joint interview promotes autonomy as the individual can influence and re-visit their narrative following the joint interview. Due to multiple informants, this approach allows for comparison and cross-checking of the report while maintaining self-advocacy and rapport with the interviewer (Bandura, 2000, 2001; Caldwell, 2014; Eisikovits & Koren, 2010).

While information collected from informants is essential to the evaluation process, the clinician's judgment and interpretation of the information is also incorporated in the final report. For instance, report bias should always be considered when interviewing informants. Report bias occurs when the informant's ability to objectively report information is affected by their role in the individual's life, and consequently may alter their perceptions of the individual's abilities, needs, and overall lifestyle (Kripke, 2017). This could include family members and staff members differing in their perceptions of an individual's skill level. Externalizing behavior could be under-reported or over-reported due to the informant's tolerance level or how the behavior displays across settings (Adams & Boyd, 2010; Carnaby, 2007). For example, aggressive and non-compliant behaviors may be less tolerated in a day treatment facility than an individual's home, or an academic teacher may not report as many deficits in self-help and adaptive skills as a caregiver who is present throughout the day. In addition, due to deficits in an individual's ability to appraise situations, communicate, and tendency to acquiesce, report bias should be taken into account when considering self-reports (Adams & Boyd, 2010; Emerson, Felce, & Stancliffe, 2013). Key information gathered from informants can be used to guide additional areas of an assessment (e.g., testing, behavioral observations; (Carnaby, 2007; Groth-Marnat, 2003; Vertue & Haig, 2008). Although information gathered from multiple informants is important for the evaluation process, the clinician's interpretation of this information based on clinical judgment and integration of findings across sources is also included in the report.

Referral Questions

The goal of the assessment is to answer the referral question by conducting a comprehensive psychological evaluation and providing treatment recommendations for support. For individuals with intellectual disability, a referral can originate from a variety of sources. Considering the referral source helps clinicians form a theoretical framework when preparing for the interview and overall assessment (Adams & Boyd, 2010; Vertue & Haig, 2008; Wilcox & Schroeder, 2015). Common referral sources include family members, caregivers, staff, educational systems, legal systems, case managers, medical doctors, or other professionals. Self-referrals also occur but are less common, as individuals are more often referred by their caregiver, family members, or general practitioners (Bouras, Cowley, Holt,

Newton, & Sturmey, 2003; Carnaby, 2007; Edelstein & Glenwick, 1997; Oliver, Miller, & Skillman, 2005; Tsakanikos, Sturmey, Costello, Holt, & Bouras, 2007). The reason for the referral may also vary by the settings in which the source interacts with the individual. For example, family members are more likely to refer issues regarding socialization, while inpatient and outpatient treatment facilities are more likely to refer due to externalizing behavior. In general, externalizing behaviors (e.g., aggression, inattention, social impairments) are listed as reasons for referral more often than internalizing behaviors (e.g., depression, anxiety; Charlot, Doucette, & Mezzacappa, 1993).

Due to a high number of referrals related to externalizing behaviors, it is important to consider referral sources that are unfamiliar with mental illness (e.g., family members, case managers), as they may have difficulty recognizing internalizing symptoms (Adams & Boyd, 2010; Borthwick-Duffy & Eyman, 1990; Bruininks, Hill, & Morreau, 1988; Oliver, Leimkuhl, & Skillman, 2003), particularly since individuals with intellectual disability experience similar levels of anxiety and depression as typically developing individuals (Deb, Thomas, & Bright, 2001; Einfeld & Tonge, 2007; Emerson, 2003). Although a referral is primarily used as the starting point from which a clinician forms their initial hypothesis, it should not be the only factor influencing case conceptualization. The referral should be used as a guide when collecting additional client information and creating a framework for the interview (Adams & Boyd, 2010; Vertue & Haig, 2008; Wilcox & Schroeder, 2015).

Interview questions targeting both past and current concerns provides not only a history of symptomatology, but also creates a timeline of symptoms. This timeline ultimately aids the clinician in answering the referral question and informs the diagnostic process. When asking questions that address current concerns, the interviewer should consider the age of the client and compare current presentation of symptoms and severity to the diagnostic criteria.

Interview Structure

A semi-structured interview format is useful when answering referral questions and working with individuals with intellectual disability. Semi-structured interviews collect key information that cannot be obtained by a test or behavioral observation. In addition, semi-structured interviews are flexible and allow the clinician to assess how questions are answered. The questions are determined by the assessor and can vary in specificity and content in order to match the case of the client being assessed. Interview questions assess the individual's overall functioning by targeting a variety of areas (e.g., activity level, physical health, developmental history, symptomatology, personal history, and overall emotional well-being; Carnaby, 2007). To increase the reliability and validity of identifying intellectual disability, questions should utilize behavioral descriptions of DSM-5 criteria and symptomatology and be gathered from a variety of focused and targeted questions (Charlot et al., 2007; Charlot & Beasley, 2013; Groth-Marnat, 2003).

The reliability and validity of the interview can decrease due to response bias of the interviewer and vary in structure case-to-case (Groth-Marnat, 2003). Since informants often report a biased interpretation of behaviors rather than specific behavioral observations, flexibility allows interviewers to further probe and clarify their responses, thus obtaining a more accurate description of symptoms (Charlot et al., 2007). Overall, the information obtained from the interview influences the selection of assessment tools and ultimately builds a framework for answering the referral questions and writing a report (Carnaby, 2007; Groth-Marnat, 2003; Vertue & Haig, 2008).

Developmental History

For individuals with IDDs, a client's developmental history is particularly relevant. Early skills such as a child saying their first word, taking their first step, and becoming toilet trained

are all considered developmental milestones. A clinician should assess whether milestones have been delayed, are abnormal (e.g., regression of skills), or have been missed.

Due to the role of delayed or missed developmental milestones as a defining feature of ID and global developmental delay (GDD), it can be beneficial to gather additional information about adaptive functioning development when collecting overall developmental history (American Psychiatric Association, 2013). Adaptive functioning develops in early childhood as a child's motor skills, sensory perception, communication, and social skills collectively develop. In late childhood and adolescence, higher order conceptual skills develop leading to greater adaptive competence. Adults with developmentally appropriate adaptive functioning are able to live in community settings by satisfying expectations of their social environment (Gligorović & Buha Đurović, 2014). Individuals with lower adaptive functioning may not possess certain higher order abilities, which may impede their ability to live independently. This suggests that it is imperative to assess individuals' current developmental functioning and history so that specific interventions can be implemented in order to improve overall outcomes.

Medical History

Gathering relevant medical history can also serve to help the clinician understand medical comorbidities that may have affected the individual's emotional functioning. Collecting a detailed medical history allows a more detailed picture of an individual's functioning and provides information that may help with diagnosis. Individuals with intellectual disability often experience higher rates of medical conditions than the general population (e.g., epilepsy, cardiovascular disease; Beange, McElduff, & Baker, 1995; Hollins, Attard, von Fraunhofer, McGuigan, & Sedgwick, 1998; Matthews, Weston, Baxter, Felce, & Kerr, 2008). Fragile X syndrome, down syndrome, fetal alcohol syndrome, and specific infections during pregnancy are known causes of ID (Center for Disease Control and Prevention, 2015). Other conditions found to be causal factors for ID include Rett syndrome, Prader-Willi syndrome, Lesch-Nyhan syndrome, adrenoleukodystrophy, neurofibromatosis, and tuberous sclerosis. Congenital factors such as lead poisoning and prenatal exposure to substances, infections including rubella, genital type II herpes, and syphilis, and metabolic factors including neonatal hypothyroidism can all increase risk for ID (Katz & Lazcano-Ponce, 2008).

Information regarding medical history should be discussed during the interview and subsequently included in the report, as it has implications for understanding symptomology and comorbid psychopathology. For example, medical conditions causing physical discomfort may increase rates of challenging behavior, while medications that the individual may be prescribed may have side effects that include changes in mood or emotional functioning.

Psychiatric History

Studies have also found that ID commonly co-occurs with psychiatric disorders. The prevalence of intellectual disability is 1–3%, and comorbidity of psychopathology is three to four times higher individuals with ID compared to the typically developing population (Borthwick-Duffy & Eyman, 1990; Dekker, Koot, van der Ende, & Verhulst, 2002). Some common comorbid psychiatric disorders of intellectual disability include problem behavior, affective disorder, autism spectrum disorder, psychotic disorder, and anxiety disorder (Cooper, Smiley, Morrison, Williamson, & Allan, 2007; Deb et al., 2001; Emerson, 2003; Matson & Shoemaker, 2011).

Einfeld, Ellis, and Emerson (2011) have found that rates of psychological disorders in children and adolescents with ID range from 30% to 50%. Relatedly, Morgan, Leonard, Bourke, and Jablensky (2008) found that 31.7% of individuals with ID had a comorbid disorder. Individuals with ID and a comorbid psychiatric disorder less frequently had severe or profound ID and more frequently had mild or borderline ID (Axmon,

Björne, Nylander, & Ahlström, 2018; Holden & Gitlesen, 2004; Morgan et al., 2008; Nettelbladt, Göth, Bogren, & Mattisson, 2009). Holden and Gitlesen (2004) further discuss that in their sample, almost all symptoms, including nonverbal symptoms, were more often present in individuals with moderate ID. They note that it can be difficult to diagnose comorbid psychiatric disorders in individuals with severe and profound ID. Their study utilized the Mini PAS-ADD (Moss, 2002), which is used to assess psychiatric disorders in individuals with ID. It is comprised of 43 items, half of which are not easily demonstrated, or cannot be demonstrated at all, by those with severe and profound ID as some items require an extensive behavioral repertoire and some require language. Even when assessing nonverbal symptoms, clinicians should be aware that some behaviors may be indicative of multiple disorders, such as sudden vocal or motor responses, which may be associated with anxiety but also may be associated with a stereotypic movement disorder (Matson, Smiroldo, Hamilton, & Baglio, 1997). Overall, previous diagnoses of comorbidities should be assessed in the interview and included in the report to help inform clinical impressions from the assessment.

Family History

Several familial risk factors exist for intellectual disability and should be examined as a part of an overall family history assessment. For example, increased risk for having a child with ID is associated with mothers who have asthma (57–62%) and renal and urinary conditions (58–68%) and mothers who have diabetes (56–62%) (Langridge et al., 2013; Leonard, de Klerk, Bourke, & Bower, 2006). Clinicians should collect a comprehensive family history including known diagnoses of family members, family background, living arrangements, and family stressors (Kishore, Udipi, & Seshadri, 2019; Lichtenberg, Mather, Kaufman, & Kafuman, 2004).

By collecting the client's family history, existing risk factors may be revealed and aid in case conceptualization. If any background information is collected separate from the interview (e.g., requested records, questionnaires) and potential familial risk factors are mentioned, the clinician should follow-up and adapt the interview questions to probe for relevant information (Lichtenberg et al., 2004). Any family history that contributed to the diagnostic impression should be included in the report (Groth-Marnat, 2003).

Report Writing

Although the structure and content of reports vary across assessment types and referral concerns, a few central goals are common. Lichtenberg et al. (2004) describe several principles and four central purposes emerged: (1) answer the referral question; (2) describe the person; (3) interpret and integrate the data; (4) provide comprehensive and appropriate recommendations for treatment. These primary goals should guide the report writing process.

Clinicians should avoid fragmented, stereotyped, and vaguely written reports that can be easily replicated and generalized to other clients. Instead, clinicians must ensure reports are integrative and individualized. For example, data from the assessment (e.g., test results, behavioral observations, self-report, or caregiver-report) should be presented with consideration to the individual client and the overall diagnostic impressions (Groth-Marnat, 2003). Although there is some flexibility regarding the content and structure of the report based on the type of evaluation (e.g., neuropsychological, educational, vocational, developmental disabilities, forensic, emotional functioning), important components include identifying information, referral and background information, behavioral observations, interpretation of measure results, diagnostic impression, and recommendations (Schneider, Lichtenberger, Mather, & Kaufman, 2018). The interview and measure components of assessment have been discussed previously in this chapter or will be considered later in this book, and therefore will not be covered in detail in this section.

Identifying Information and Referral Concern

When writing a comprehensive psychological report, the critical elements of identifying client information and the reason for referral are listed at the beginning. The identifying client information includes the following: client's name, date of birth, age, grade or occupation, dates of testing and of report, as well as the examiner and, if applicable, supervisor (Schneider et al., 2018).

The next component of the report is the description of the referral concern. Re-stating the referral question in the beginning of the report creates a narrative for why the evaluation was necessary or requested. Generally, this paragraph should begin with a brief statement that includes identifying information and a description of the reason for referral (e.g., intellectual evaluation, psychoeducational assessment; Groth-Marnat, 2003; Schneider et al., 2018). As mentioned earlier in this chapter, common reasons for referral for individuals with intellectual disability are due to the presence of externalizing behaviors (e.g., aggression, non-compliance), and the specific behavior concern may vary from source to source (e.g., parents, school, case manager; de Ruiter, Dekker, Verhulst, & Koot, 2007; Edelstein & Glenwick, 2001).

Background Information

Previously mentioned in this chapter, the collection of the client's developmental, medical, psychiatric, and family history is essential when assessing for IDDs. Relevant background information, typically obtained during the interview portion of the assessment, should be reported. Information that contributed to interpretation of test scores and overall case conceptualization must be included, and personal information that is irrelevant to the evaluation excluded (Lichtenberg et al., 2004). Clinicians should write background information in a concise manner, presenting the information as a narrative that is easy to follow and understand (Groth-Marnat, 2003).

Behavioral Observation

When assessing individuals with ID who potentially have a dual diagnosis, behavioral observation is used in addition to other assessment tools. Individuals with ID may not be able to describe or may lack insight into psychological symptoms, and caregivers may misinterpret or not report certain symptoms. Objective observation by a clinician trained in behavioral observation and differential diagnosis is an important component of the assessment.

When including the behavioral observations in the report, clinicians should review their observations and group them into meaningful categories. Rather than merely reporting the exact behaviors, the clinician should include interpretation of these categories of behaviors (Schneider et al., 2018). This interpretation is meant to clarify why these observations were important and what they may indicate for the individual's difficulties and potential diagnoses.

Additional information that may be relevant to include in the behavior observation section include physical appearance (e.g., grooming, stature), rapport establishment and maintenance, speech (e.g., intonation, volume), and social communication (e.g., reciprocal conversation, quality of responses). Additionally, observations of the individual's response to testing procedures (e.g., persistence with difficult items, response to feedback, problem-solving), attention, activity level, temperament, and affect are important to consider. Unusual mannerisms or behavior is also particularly relevant in this population, as these types of behaviors may provide insight into comorbid concerns (Schneider et al., 2018).

Objective measures of behavior may also be helpful. Several assessment measures include behavioral observation components, such as the Woodcock Johnson Tests of Achievement (Schrank, McGrew, Mather, Wendling, & LaForte, 2014). Although not always necessary, observation measures may help to assess a particular area or ensure comprehensive observations or accurate behavioral probes. The clinician should also record and interpret the impact of behavior on the assessment. For example, if the

individual demonstrates high levels of inattention or hyperactivity that impeded their ability to complete assessment measures (e.g., intelligence tests), this information should be included in the report and the clinician should specify whether the results of testing are considered valid taking these behaviors into consideration. In individuals with ID, physical difficulties (i.e., limited motor movement, vision or hearing impairments) or problems attending the stimuli may be prevalent and may impact their performance during the assessment.

To assess comorbidities in individuals with ID, specific behavioral probes may help to provide insight into symptomology. For example, when assessing for specific phobias or separation anxiety, it is possible for clinicians to present the individual with anxiety-provoking stimuli and directly observe behavioral responses such as avoidance; this is known as a behavioral avoidance test (BAT; Dadds, Rapee, & Barrett, 1994; Hagopian & Jennett, 2008). Conversely, assessing for generalized anxiety disorder (GAD) in individuals with ID may require more naturalistic observation by both the clinician and caregivers. Behavioral symptoms of concern for GAD involve avoidant behavior and negative emotional expression which may be observed through facial expression, vocalization, motor movements, and posture. As mentioned previously, caution should be taken when examining behavioral symptoms of anxiety in individuals with ID as they may be indicative of other disorders commonly associated with ID (e.g., motor responses that are associated with stereotypic movement disorder, behavioral avoidance of non-preferred activities that may not provoke anxiety; Hagopian & Jennett, 2008; Matson et al., 1997). Further distinctions should be made with regard to these behaviors which may be done through a functional behavioral assessment (FBA; Hagopian & Jennett, 2008). Objective findings from these observations should be added to a behavioral observation report to provide evidence to support the diagnostic impressions.

While behavioral observations are often conducted in testing environments (e.g., hospital, clinic, office), observations of the individuals in naturalistic settings such as their homes or communities may also be helpful in assessing difficulties. In addition to determining which behaviors are to be observed, the clinician should also decide how these behaviors will be recorded in advance. For example, if the clinician is assessing challenging behaviors in the context of a group home, the frequency, duration, severity, and topography would be important information to collect. Information regarding the antecedents and consequences of each behavior would also inform the clinician's interpretation of the function of the behaviors and would inform the treatment recommendations (Cooper, Heron, & Heward, 2014).

Diagnostic Impressions and Recommendations

Finally, the report concludes with the diagnostic impression and recommendations. This section should not only provide the diagnosis but also include sufficient evidence to support the diagnosis. The diagnostic code should be recorded in this section as providers such as insurance agencies require this code, in addition to the diagnostic label, to provide services. Additionally, the report should include a description of other diagnoses that were considered and not given, with evidence for why these were being considered as differential diagnoses and why the individual does not meet criteria for them based on the results of the assessment. Clinical impression and explanations for any inconsistencies in data should also be provided to detail the clinician's rationale for the results of the assessment (Schneider et al., 2018).

An ethical consideration in providing diagnostic impressions is whether the clinician is qualified to make a diagnosis. If an examiner suspects a diagnosis but does not have sufficient training and expertise in that area, a referral to a specialist would be appropriate (Schneider et al., 2018). It is still important to include all of the information typically described in a report, so the report can serve as a record for other professionals who may conduct their own assessments.

The final section of the report outlines the recommendations for treatment, supports, and services. They are targeted recommendations to address the referral problems and the clinical impressions of what difficulties are causing, contributing, or maintaining these difficulties (Hass & Carriere, 2014). Treatment recommendations should be reasonable, client-centered, and supported by information and data obtained during the assessment and included in the report. This allows readers to easily understand the reasoning behind each recommendation suggested (Groth-Marnat, 2003). Additionally, the clinician's suggestions for treatment should be informed by evidence-based practices to address the difficulties the client is experiencing. Clinicians should consider the number, complexity, and specificity of the recommendations (Schneider et al., 2018). Recommendations should be practical and special consideration should be made for any barriers to services and the individual's current environment (Groth-Marnat, 2003). For example, if a client lives in an area that has limited access to services or long waiting lists for treatment, clinicians should consider incorporating supplemental recommendations.

Recommendations should be written with the reader of the report in mind, with language that is simple and easy to comprehend (Groth-Marnat, 2003; Lichtenberg et al., 2004). For individuals diagnosed with ID, it is imperative that the clinician prioritize the client and caregivers' understanding of treatment recommendations, while supporting the client's decision-making role and their right to self-determination (Krahn, Hammond, & Turner, 2006).

Conclusion

The interview and report writing processes are crucial aspects of all assessments but have particular importance in the assessment of psychiatric comorbidities for individual with ID. The previous chapters in the volume outlined some of the common comorbidities associated with ID, and while the purpose of the assessment may vary depending on the client and their specific needs, an overall goal is to improve outcomes. Training in effective interviewing techniques for this population and their caregivers is crucial as if differs from how interviews are conducted in typically developing populations. Individual outcomes can be greatly influenced by clinical assessments as correct diagnosis enables access to services. Therefore, the importance of assessment and treatment of comorbidities within this population is imperative to maximizing functioning.

References

Adams, Z. W., & Boyd, S. E. (2010). Ethical challenges in the treatment of individuals with intellectual disabilities. *Ethics & Behavior, 20*(6), 407–418.

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

American Psychological Association. (2002). *Ethical principles of psychologists and code of conduct*. Retrieved November 26, 2016, from.

Arksey, H. (1996). Collecting data through joint interviews. *Social Research Update, Winter*(15).

Axmon, A., Björne, P., Nylander, L., & Ahlström, G. (2018). Psychiatric diagnoses in relation to severity of intellectual disability and challenging behaviors: A register study among older people. *Aging & Mental Health, 22*(10), 1344–1350.

Bandura, A. (2000). Exercise of human agency through collective efficacy. *Current Directions in Psychological Science, 9*(3), 75–78.

Bandura, A. (2001). Social cognitive theory: An agentic perspective. *Annual Review of Psychology, 52*, 1–26.

Beange, H., McElduff, A., & Baker, W. (1995). Medical disorders of adults with mental retardation: A population study. *American Journal on Mental Retardation, 99*(6), 595–604.

Borthwick-Duffy, S. A., & Eyman, R. K. (1990). Who are the dually diagnosed? *American Journal of Mental Retardation: AJMR, 94*(6), 586–595.

Bouras, N., Cowley, A., Holt, G., Newton, J. T., & Sturmey, P. (2003). Referral trends of people with intellectual disabilities and psychiatric disorders. *Journal of Intellectual Disability Research, 47*(6), 439–446.

Bruininks, R. H., Hill, B. K., & Morreau, L. E. (1988). Prevalence and implications of maladaptive behaviors and dual diagnosis in residential and other service programs. In J. A. Stark, F. J. Menolascino, M. H. Albarelli, & V. C. Gray (Eds.), *Mental retardation and mental health: Classification, diagnosis, treatment, services* (pp. 3–29). New York, NY: Springer New York.

Caldwell, K. (2014). Dyadic interviewing: A technique valuing interdependence in interviews with individuals with intellectual disabilities. *Qualitative Research, 14*(4), 488–507.

Cameron, R., & Shepel, L. (1981). Strategies for preserving the confidentiality of psychological reports. *Canadian Psychology/Psychologie Canadienne, 22*(2), 191–193.

Carnaby, S. (2007). Developing good practice in the clinical assessment of people with profound intellectual disabilities and multiple impairment. *Journal of Policy and Practice in Intellectual Disabilities, 4*(2), 88–96.

Center for Disease Control and Prevention. (2015, April 15). *Facts about fragile X syndrome*. Retrieved September 9, 2016, from Fragile X Syndrome (FXS) website: https://www.cdc.gov/ncbddd/fxs/facts.html

Charlot, L., & Beasley, J. (2013). Intellectual disabilities and mental health: United States–based research. *Journal of Mental Health Research in Intellectual Disabilities, 6*(2), 74–105.

Charlot, L., Deutsch, C., Hunt, A., Fletcher, K., & Mcllvane, 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.

Charlot, L., Doucette, A. C., & Mezzacappa, E. (1993). Affective symptoms of institutionalized adults with mental retardation. *American Journal of Mental Retardation: AJMR, 98*(3), 408–416.

Cooper, J. O., Heron, T. E., & Heward, W. L. (2014). *Applied behavior analysis* (2nd ed.). Harlow, UK: Pearson Education.

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.

Dadds, M. R., Rapee, R. M., & Barrett, P. M. (1994). Behavioral observation. In T. H. Ollendick, N. J. King, & W. Yule (Eds.), *International handbook of phobic and anxiety disorders in children and adolescents* (pp. 349–364). Boston, MA: Springer.

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.

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. *Journal of Child Psychology and Psychiatry, 43*(8), 1087–1098.

Edelstein, T. M., & Glenwick, D. S. (1997). Referral reasons for psychological services for adults with mental retardation. *Research in Developmental Disabilities, 18*(1), 45–59.

Edelstein, T. M., & Glenwick, D. S. (2001). Direct-care workers' attributions of psychopathology in adults with mental retardation. *Mental Retardation, 39*(5), 368–378.

Einfeld, S. L., & Tonge, B. J. (2007). Population prevalence of psychopathology in children and adolescents with intellectual disability: II. Epidemiological findings. *Journal of Intellectual Disability Research, 40*(2), 99–109.

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 & Developmental Disability, 36*(2), 137–143.

Eisikovits, Z., & Koren, C. (2010). Approaches to and outcomes of dyadic interview analysis. *Qualitative Health Research, 20*(12), 1642–1655.

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.

Emerson, E., Felce, D., & Stancliffe, R. J. (2013). Issues concerning self-report data and population-based data sets involving people with intellectual disabilities. *Intellectual and Developmental Disabilities, 51*(5), 333–348.

Emerson, E., Hatton, C., Dickson, K., Gone, R., & Caine, A. (2012). *Clinical psychology and people with intellectual disabilities*. Chichester, UK: Wiley.

Finlay, W. M. L., & Lyons, E. (2002). Acquiescence in interviews with people who have mental retardation. *Mental Retardation, 40*(1), 14–29.

Fisher, C. B. (2003). Goodness-of-fit ethic for informed consent to research involving adults with mental retardation and developmental disabilities. *Mental Retardation & Developmental Disabilities Research Reviews, 9*(1), 27–31.

Gligorović, M., & Buha Đurović, N. (2014). Inhibitory control and adaptive behaviour in children with mild intellectual disability: Inhibitory control and adaptive behaviour. *Journal of Intellectual Disability Research, 58*(3), 233–242.

Grisso, T., Grisso, A., & Appelbaum, P. S. (1998). *Assessing competence to consent to treatment: A guide for physicians and other health professionals*. New York, NY/Oxford, UK: Oxford University Press.

Groth-Marnat, G. (2003). *Handbook of psychological assessment*. Hoboken, NJ: Wiley.

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*(5), 467–483.

Hass, M., & Carriere, J. A. (2014). *Writing useful, accessible, and legally defensible psychoeducational reports* (1st ed.). Hoboken, NJ: Wiley.

Hilsenroth, M. J., & Cromer, T. D. (2007). Clinician interventions related to alliance during the initial interview

and psychological assessment. *Psychotherapy (Chicago, Ill.), 44*(2), 205–218.

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.

Hollins, S., Attard, M. T., von Fraunhofer, N., McGuigan, S., & Sedgwick, P. (1998). Mortality in people with learning disability: Risks, causes, and death certification findings in London. *Developmental Medicine & Child Neurology, 40*(1), 50–56.

Katz, G., & Lazcano-Ponce, E. (2008). Intellectual disability: Definition, etiological factors, classification, diagnosis, treatment and prognosis. *Salud Pública de México, 50*, s132–s141.

Kishore, M. T., Udipi, G. A., & Seshadri, S. P. (2019). Clinical practice guidelines for assessment and management of intellectual disability. *Indian Journal of Psychiatry, 61*, 194–210.

Krahn, G. L., Hammond, L., & Turner, A. (2006). A cascade of disparities: Health and health care access for people with intellectual disabilities. *Mental Retardation and Developmental Disabilities Research Reviews, 12*, 70–82.

Kripke, C. (2017). Patients with disabilities: Avoiding unconscious bias when discussing goals of care. *American Family Physician, 96*(3), 192–195.

Langridge, A. T., Glasson, E. J., Nassar, N., Jacoby, P., Pennell, C., Hagan, R., … Stanley, F. J. (2013). Maternal conditions and perinatal characteristics associated with autism spectrum disorder and intellectual disability. *PLoS One, 8*(1), e50963.

Leonard, H., de Klerk, N., Bourke, J., & Bower, C. (2006). Maternal health in pregnancy and intellectual disability in the offspring: A population-based study. *Annals of Epidemiology, 16*(6), 448–454.

Lichtenberg, E. O., Mather, N., Kaufman, N. L., & Kafuman, A. S. (2004). *Essentials of assessment report writing*. Hoboken, NJ: Wiley.

Loman, S., Vatland, C., Strickland-Cohen, K., Horner, R., & Walker, H. (2010). *Promoting self-determination: A practice guide*. Kansas City, KS: National Gateway to Self-Determination.

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

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.

Matthews, T., Weston, N., Baxter, H., Felce, D., & Kerr, M. (2008). A general practice-based prevalence study of epilepsy among adults with intellectual disabilities and of its association with psychiatric disorder, behaviour disturbance and carer stress. *Journal of Intellectual Disability Research, 52*(Pt 2), 163–173.

McSherry, B. (2015). Decision-making, legal capacity and neuroscience: Implications for mental health laws. *Laws, 4*(2), 125–138.

Morgan, V. A., Leonard, H., Bourke, J., & Jablensky, A. (2008). Intellectual disability co-occurring with schizophrenia and other psychiatric illness: Population-based study. *The British Journal of Psychiatry: the Journal of Mental Science, 193*(5), 364–372.

Moss, S. (2002). *The mini-PASADD interview pack*. Shoreham-by-Sea, UK: Pavilion.

Nettelbladt, P., Göth, M., Bogren, M., & Mattisson, C. (2009). Risk of mental disorders in subjects with intellectual disability in the Lundby cohort 1947–97. *Nordic Journal of Psychiatry, 63*(4), 316–321.

Oliver, M. N., Leimkuhl, T. T., & Skillman, G. D. (2003). Training needs, work related stressors, and job satisfaction of community staff supporting adults with mental retardation: Implications for ensuring optimal support quality. *The NADD Bulletin, 6*(4), 68–73.

Oliver, M. N. I., Miller, T. T., & Skillman, G. D. (2005). Factors influencing direct-care paraprofessionals' decisions to initiate mental health referrals for adults with mental retardation. *Mental Retardation, 43*(2), 83–91.

Perry, J. (2004). Interviewing people with intellectual disabilities. In *International handbook of applied research in intellectual disabilities* (pp. 116–131). Chichester, UK: Wiley.

Schneider, W. J., Lichtenberger, E. O., Mather, N., & Kaufman, N. L. (2018). *Essentials of assessment report writing*. Hoboken, NJ: Wiley.

Schrank, F. A., McGrew, K. S., Mather, N., Wendling, B. J., & LaForte, E. M. (2014). *Woodcock-Johnson IV tests of achievement*. Rolling Meadows, IL: Riverside Publishing Company.

Skarstad, K. (2018). Ensuring human rights for persons with intellectual disabilities? *The International Journal of Human Rights, 22*(6), 774–800.

Tsakanikos, E., Sturmey, P., Costello, H., Holt, G., & Bouras, N. (2007). Referral trends in mental health services for adults with intellectual disability and autism spectrum disorders. *Autism, 11*(1), 9–17.

Vertue, F. M., & Haig, B. D. (2008). An abductive perspective on clinical reasoning and case formulation. *Journal of Clinical Psychology, 64*(9), 1046–1068.

Wilcox, G., & Schroeder, M. (2015). What comes before report writing? Attending to clinical reasoning and thinking errors in school psychology. *Journal of Psychoeducational Assessment, 33*(7), 652–661.

Wright, J. (2010). *The RIOT/ICEL matrix: Organizing data to answer questions about student academic performance & behavior*. Retrieved from Intervention Central website: www.interventioncentral.org

Functional Assessment in Dual Diagnosis

10

Renee O. Hawkins, Tai A. Collins, and Cara Dillon

Individuals with intellectual disabilities (ID) display challenging behavior at higher rates than their typically developing peers (Dekker, Koot, van der Ende, & Verhulst, 2002; Emerson & Einfeld, 2010). Behaviors such as self-injury, noncompliance, and aggression can negatively impact social, emotional, and academic functioning (Emerson et al., 2001). For some individuals with ID, these challenging behaviors and/or other impairments may be linked to a mental health disorder, leading to a dual diagnosis (DD). Research suggests that individuals with ID experience mental health disorders at higher rates than individuals without ID, with published estimates of the prevalence of mental health disorders among individuals with ID three to five times higher than for individuals without ID (Araten-Bergman & Werner, 2017). Individuals with ID experience significant deficits in cognitive and adaptive functioning, and these deficits can be exacerbated by the presence of a co-occurring mental health disorder (Baker et al., 2003; Cervantes & Matson, 2015; Rojahn, Rowe, Kasdan, Moore, & van Ingen, 2011).

To effectively support the complex needs of individuals with ID who also experience a mental health disorder, reliable and valid assessment approaches are needed to help understand the maladaptive behaviors that may be interfering with overall functioning and to inform effective treatment planning. One approach that is well supported in the literature for addressing challenging behavior is functional behavioral assessment (FBA). The goal of FBA is to identify variables in the environment that are supporting maladaptive behavior so these variables can be manipulated to promote positive behavior change (Cooper, Heron, & Heward, 2007; Steege & Watson, 2009). In addition to the challenging behaviors often displayed by individuals with ID, both externalizing mental health disorders (e.g., attention deficit hyperactivity disorder) and internalizing mental health disorders (e.g., anxiety, depression) have a number of observable, measurable, negative behaviors associated with them (Axelrod, 2017) that can be examined and ultimately improved with the use of FBA methods. For example, behaviors such as social withdrawal, crying, and aggression may be appropriate FBA targets.

Through FBA, data are collected to determine antecedents, stimuli that are present just prior to the occurrence of the maladaptive behavior, and consequences, stimuli that occur immediately following the occurrence of the maladaptive behavior. Both antecedents and consequences affect the chances that a behavior will occur. A discriminative stimulus is one type of antecedent that signals that reinforcement is available for a particular behavior. For example, a water fountain

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signals that a drink is available by pushing the lever. Motivating operations are antecedent events that affect the value of reinforcement. For example, if it is uncomfortably cold outside, extra time outside at recess may not be as reinforcing as it would be on a warm, sunny day. Consequences fall into three categories: (1) positive reinforcement, which includes social attention, activities, tangibles, and edibles; (2) negative reinforcement, which includes escape or avoidance of social attention, settings, task demands, or activities; and (3) automatic reinforcement, which refers to a change in sensory stimulation. By closely examining the relationships between antecedents, target behaviors, and consequences, hypotheses are generated as to the function of the behavior (i.e., why it is occurring). These hypotheses are then used to develop intervention plans that manipulate environmental variables as to increase appropriate behavior and decrease problem behavior (Cooper et al., 2007; Steege & Watson, 2009).

The use of FBA is well supported in the research as a technically sound approach for assessing problem behavior (Hanley, Iwata, & McCord, 2003). FBA approaches have been widely studied for examining challenging behaviors such as aggression, noncompliance, property destruction, stereotypy, off-task, self-injury, tantrums, elopement, inappropriate vocalizations (Anderson, Rodriguez, & Campbell, 2015; Hanley et al., 2003). FBA methods have been used across a variety of settings (i.e., school, home, clinic) and have been used to assess the behaviors of typically developing individuals as well as individuals with disabilities (Hanley et al., 2003). More specifically, FBA methods have been used to effectively support children with attention deficit hyperactivity disorder (Miller & Lee, 2013), autism spectrum disorder (Larkin, Hawkins, & Collins, 2016), emotional and behavioral disorders (Murdock, O'Neill, & Cunningham, 2005), and ID (Wadsworth, Hansen, & Wills, 2015). Repeatedly, researchers have documented the benefits of identifying a function of problem behavior through FBA to develop an effective intervention plan (Didden, Korzilius, Oorsouw, & Sturmey, 2006; Harvey, Boer, Meyer, & Evans, 2009). Further, there is empirical evidence that function-based interventions can be more effective than interventions that are developed without consideration of function (Ingram, Lewis-Palmer, & Sugai, 2005; Newcomer & Lewis, 2004; Payne, Scott, & Conroy, 2007).

Types of Functional Assessments

Three types of FBA have been identified in the literature including indirect FBA, direct descriptive FBA, and functional analysis (Cooper et al., 2007). Indirect and direct descriptive FBAs lead to the generation of functional hypotheses regarding the environmental variables supporting problem behavior. Functional analysis systematically manipulates the environment to confirm functional hypotheses. Each of the three types of FBA differs with regard to their goals, administration times, and procedures; however, they each have their place in isolation and in combination in the assessment of the maintaining variables of challenging behaviors. The following section details the methods and procedures as well as the advantages and disadvantages of each type of FBA.

Indirect FBA

Indirect FBA refers to the use of assessments such as interviews and rating scales to determine a hypothesized function of problem behavior (Cooper et al., 2007). The goals of indirect FBA are to (1) operationally define the problem behavior(s); (2) identify the antecedents that occur before the behavior; (3) identify the consequences maintaining the behavior; (4) generate acceptable alternatives to the problem behavior; and (5) review previous intervention attempts to address the challenging behavior. In the case of severe problem behaviors, practitioners should not conduct an indirect FBA in isolation. Rather, indirect FBA should be used as the first step in a systematic FBA process to inform more stringent assessments. In the case of relatively minor behaviors, indirect FBA may be used in isolation

to quickly determine a hypothesized function and design a function-based intervention to address the problem behavior.

Indirect FBA is associated with a number of advantages and disadvantages. With regard to advantages, indirect FBA is often convenient and can be administered relatively quickly (Cooper et al., 2007). It also does not require direct observation or the systematic manipulation of maintaining variables, so indirect FBA is often feasible in a variety of settings and fits within general consultation routines. With regard to disadvantages, indirect FBA relies on the recollections and reporting of individuals closely related to the situation (e.g., teachers, caregivers, staff), which may be biased, inaccurate, and incomplete (Cooper et al., 2007). Also, technically adequate data are difficult to obtain from interviews in particular because they are often semi-structured, resulting in limited overall technical adequacy (Dufrene, Kazmerski, & Labrot, 2017).

FBA Interviews Interviews are a key method of gathering information about target individuals and behaviors. Interviews are used to gather information from persons that interact with the target individual (e.g., teachers and parents) as well as to gather information that the individual may have about their own behaviors (Steege & Watson, 2009). The goal of FBA interviews is to collect information about the behaviors in question, the antecedents of the behaviors, the possible function of the behavior through consequences, and desired alternative replacement behaviors (Gresham, Watson, & Skinner, 2001). A variety of interview protocols have been developed to guide teams in gathering data for indirect FBAs. Most of the FBA interviews are semi-structured, which allows for the interviewer to have flexibility in gathering data on the strengths and weaknesses of the client, antecedents, behaviors, and consequences, and other aspects of the environment.

Functional Assessment Interview (FAI) The FAI (O'Neil et al., 1997) is a semi-structured interview protocol that takes 45–90 min to complete (Kelley, LaRue, Roane, & Gadaire, 2011). Questions are asked within 11 sections, including a description of the behavior, setting events, antecedents, consequences, efficiency of the behavior (i.e., how much the appropriate and inappropriate behaviors lead to reinforcement), functional alternative behavior, primary mode of communication, things to do/avoid (i.e., discussing the idiosyncrasies of the client), reinforcers, history of undesirable behavior, and summary statements (O'Neil et al., 1997). As such, the FAI can be used to get a comprehensive picture of the environment and determine a function-based intervention plan.

Functional Assessment Checklist: Teachers and Staff (FACTS) The FACTS (March et al., 2000) is a semi-structured interview developed to be more efficient than the FAI. During the 10–25 min administration time, a functional hypothesis is generated based on two parts of the interview (McIntosh et al., 2008). In Part A, a routine analysis is conducted whereby the challenging behavior and the parts of the client's schedule that are most and least often associated with the challenging behavior are identified. During Part B, an operational definition is determined, and setting events, antecedents, and consequences associated with each problem behavior routine are identified. The utility of this form is shown in the hypothesis agreement between the FACTS informed hypotheses and functional analysis hypotheses noted by McIntosh et al. (2008).

Functional Assessment Informant Record for Teachers (FAIR-T) The FAIR-T (Edwards, 2002) is a semi-structured interview with Likert-type responses and yes/no prompts. Four sections are included: referral information, problem behaviors, antecedents, and consequences. Researchers have created a preschool version (FAIR-T P; Dufrene, Doggett, Henington, & Watson, 2007) for use in early intervention settings.

Student-Guided Functional Assessment Interview Another semi-structured interview used in schools is the Student-Guided Functional Assessment Interview (Reed, Thomas, Sprague, & Horner, 1997). It can establish the target individual's perspective on their own behaviors by asking questions about what the individual likes, dislikes, struggles with and when these struggles occur.

Rating Scales In addition to interviews, rating scales can provide valuable information about the context of challenging behaviors. Rating scales are typically more structured than interviews, as they consist of standardized questions and prompts that key stakeholders complete. They can be used in isolation or with interviews to inform function-based intervention or more intensive FBA procedures (Cooper et al., 2007). Behavior rating scales are a resource in assessment of behavior that can cover a wide breadth of knowledge with a variety of informants (Cooper et al., 2007). Behavior rating scales can provide information about behaviors that may not be able to be readily observed during a short observation period or in an adjacent setting.

A benefit of some behavior rating scales is that they are norm-referenced, which can give information about where an individual compares to a large sample, and this offers the ability to screen many individuals in a short time for a variety of topics (Cooper et al., 2007). For example, there are omnibus tools like the Behavior Assessment System for Children – Third Edition (BASC-3; Kamphaus & Reynolds, 2015), which addresses externalizing and internalizing behaviors, adaptive skills, anxiety, and depression over general and clinical samples (Cooper et al., 2007). There are more focused tools like the Conners Third Edition ADHD Index (Conners, 2008) and the subsequent full form that covers comorbid disorders, family issues, and peer issues and the Behavioral and Emotional Screening (BESS; Kamphaus & Reynolds, 2015), which is a more focused tool developed from the BASC-3. However, these behavior rating scales are limited in nature and should be used in conjunction with other methods like interviews, as the antecedents and consequences of behaviors are often not captured (Gresham et al., 2001). Moreover, these scales can fall prey to reputational biases, effects of central tendency, and rater and setting variance (Cooper et al., 2007). In addition to omnibus and more targeted rating scale tools, several FBA rating scales have been developed.

Questions About Behavior Function (QABF) The QABF (Matson & Vollmer, 1995) is the most frequently researched FBA rating scale, with more studies than all other rating scales combined (Matson & Williams, 2014). The QABF consists of 25 items in 5 categories, including attention, escape, nonsocial, physical, and tangible functions (Matson, Tureck, & Rieske, 2012). Taking an average of 20 min to administer, the QABF does not require extensive training and is associated with strong psychometric properties (Matson et al., 2012). This assessment is notable because it assesses a large range of functions, including social avoidance, tangible reinforcement, and physical discomfort, which are not covered in other rating scales (Kelley et al., 2011). Evidence exists that the QABF, which was modeled after a traditional functional analysis (FA), yields similar results to an FA with much less time, effort, and required training (Matson & Williams, 2014).

Functional Analysis Screening Tool (FAST) The FAST (Iwata & DeLeon, 1996) is a rating scale consisting of 16 items across 4 categories: social-positive reinforcement (i.e., access, tangible); social-negative reinforcement (i.e., escape from demands and/or social situations); automatic-positive reinforcement (i.e., self-stimulatory behavior); and automatic-negative behavior (i.e., pain or discomfort attenuation; Iwata, DeLeon, & Roscoe, 2013). The functional category with the most "yes" responses is typically hypothesized as the function of the problem behavior (Kelley et al., 2011). Mixed results have been published regarding the psychometric properties of the FAST (Iwata et al., 2013; Matson et al., 2012).

Motivation Assessment Scale (MAS) The MAS (Durand & Crimmins, 1992) is a 16-item scale with Likert-type ratings from 0 to 6 (Kelley et al., 2011). The four factors of the MAS are escape from demands, attention, tangible, and sensory reinforcement (Matson & Williams, 2014). Similar to the FAST, psychometric results on the MAS are mixed and additional research is needed (Matson & Williams, 2014).

Other Scales In addition to the previously mentioned rating scales, many other indirect FBA rating scales have been developed, including the Contextual Assessment Inventory (CAI; McAtee, Carr, & Schulte, 2004; Carr, Ladd, & Shulte, 2008) and the Problem Behavior Questionnaire (PBQ; Lewis, Scott, & Sugai, 1994). Steege and Watson (2009) included sample indirect FBA forms and hypothetical examples, including the Behavioral Stream Interview (BSI) and Functional Behavioral Assessment Screening Form (FBASF).

Direct Descriptive FBA

Direct descriptive FBA involves more intensive assessment methods and is distinguished by the use of direct observation in the setting of interest (Cooper et al., 2007). Similar to indirect FBA, direct descriptive FBA starts with the use of interviews and rating scales to collect information on the individual and the behaviors of concern. However, this information is then verified and further explored through direct observation. Practitioners often utilize the results of an indirect FBA to inform the target behaviors and procedures of a direct descriptive FBA. Direct descriptive FBA is advantageous because it capitalizes on direct observation as the hallmark of assessment within applied behavior analysis, does not require a change to the typical routine, and yields quantifiable data (Cooper et al., 2007). Major disadvantages include the problems with direct observation (e.g., observer effects, biases, reactivity, etc.; Cooper et al., 2007), infrequent behaviors may not be detected, and the results are correlational and not causal (i.e., observers identify events associated with the problem behavior, but do not systematically manipulate those events). Direct observation data can be collected through systematic direct observation methods, ABC recording, scatterplots, and direct behavior ratings.

Systematic Direct Observation (SDO) There are several SDO methods, including event recording, timing, time sampling, and planned activity checks (PLACHECK; Cooper et al., 2007). All of these methods rely on operationalized behaviors that not only include a clear and specific definition of behaviors being observed but examples and non-examples of the behaviors (Cooper et al., 2007). Concerns of SDO encompass the time that it takes to observe in the environment, whether or not the behavior will actually occur during the observation sessions, and lack of context surrounding the behaviors recorded (Lewis, Scott, Wehby, & Wills, 2014).

Event recording methods record frequency of behaviors and can be accomplished in a wide variety of ways in a variety of settings. Event recording can be completed with a paper and pencil, a mechanical clicker counter, or beads put into a bowl, and this method is easy to train caregivers or teachers to do (Cooper et al., 2007). Behaviors that are appropriate for event recording include those behaviors that have an explicit beginning and end, such as hand raising or positive comments to peers (Gresham et al., 2001).

Timing behaviors involve the measurement of the duration, latency, or inter-response time of behaviors (Cooper et al., 2007). Duration is a measure of how long a behavior occurs from start to finish. A percentage of time a behavior occurs during an observation session can be determined by adding the separate durations together, dividing by the total time observed, and multiplying by 100 to get a percentage (Gresham et al., 2001). Duration recording is an appropriate method for assessing behaviors for which there are concerns that the duration is too brief or extended and initial intervention goals may relate to increasing or decreasing the duration of the behavior. Tantrums

and crying are example behaviors for which duration recording may be considered. Latency is a measure of time between an antecedent and a behavior, for example, the time between a caregiver instructing the individual to put away their dishes and the target individual performing the task of putting away the dishes. Inter-response time is a measure from the end of one occurrence of a target behavior to the onset of the next incidence of the target behavior. This method could be used to measure time between appropriate peer interactions. Stop watches or digital timers would be the most precise method of collecting timing data (Cooper et al., 2007).

Time sampling methods divide a longer duration observation session into brief (i.e., 10–30 s) observation intervals to determine estimates of the duration or rate of the target behavior (Cooper et al., 2007). Whole-interval recordings measure whether or not a behavior occurred during the entire interval (Cooper et al., 2007). With partial-interval recording, a behavior is recorded if it occurs at any time in the interval (Gresham et al., 2001). When using momentary time sampling, behaviors are only recorded if they occur at the very end of the interval (Cooper et al., 2007). For these three types of time sampling procedures, data are reported in terms of the percentage of intervals in which the behavior occurred (Cooper et al., 2007). Lastly, as another time sampling method, PLACHECK can be used to assess the behavior of multiple target individuals at the same time. This method uses a head count in order to determine which individuals, through separate percentages, are participating in an activity more so than others (Cooper et al., 2007).

When deciding which SDO methods to use, it is important to consider that different methods may overestimate or underestimate the occurrence of behavior (Cooper et al., 2007). For example, behaviors that are short lived, like answering a yes/no question, would not be appropriate for 10 s whole-interval recording because there would be a tendency to underestimate the occurrences of that behavior if it does not last the full 10 s (Cooper et al., 2007). Partial-interval recording may also underestimate the occurrence of behaviors as repeated occurrences of the behavior may occur during the interval, but the data will show only one occurrence during the interval (Cooper et al., 2007). This would be true for rapid self-injurious behaviors that could be better recorded through event recording. Alternatively, partial-interval recording may overestimate behavior if it only occurs very briefly during the intervals. Momentary time sampling also suffers from underestimation because a behavior may not occur at the end of the interval but in the middle (Cooper et al., 2007).

When planning SDO sessions, it is important to recognize that observing people in their own environment can change how they behave and to be conscience of possible observer effects (Baer, Wolf, & Risley, 1968). Three to five observations are recommended to achieve a more accurate interpretation of the target individual's behavior (Briesch, Chafouleas, & Riley-Tillman, 2010). Also, interviews can identify certain times (transitions between activities, bedtime, lunch) in which the behavior is most likely to occur to increase the chances that the behavior is captured during SDO sessions.

Observation forms or protocols for SDO methods can be created or obtained from standardized forms. Different forms can be used to include the observation of multiple target individuals, multiple target behaviors, and using a variety of forms of time sampling. The method for the observation needs to consider the target behaviors and the appropriateness of different approaches as discussed above. For example, the Behavior Observation of Students in Schools (BOSS; Shapiro, 2004) combines momentary time sampling of a continuous behavior (e.g., engagement) and partial-interval time sampling for behaviors that are more inconsistent (e.g., off-task verbal). Other considerations would include the feasibility of the observation system and if it fits the environment. For example, 5 s momentary time sampling where peer interactions are recorded on the playground may be difficult for the observer as the children are often mobile.

When creating observational codes for target behaviors, operationalization of the behaviors should be done precisely so any observer can use

the observation code without ambiguity (Baer et al., 1968). This becomes especially salient when considering the need for inter-observer agreement data that verifies the reliability and validity of observation data (Cooper et al., 2007). Function and topography are two ways that behaviors can be defined. A function-based definition notes the common effect on the environment, and several behaviors can create the effect (Cooper et al., 2007). For example, a tantrum, stomach ache, and crying can all lead to attention from an adult, and a function-based definition would encompass all these behaviors. This type of target behaviors is considered best practice when possible because it can include several behaviors, the function of the behavior is paramount in observation and intervention, and these definitions can be more succinct (Cooper et al., 2007). The other way to define a behavior is through a topography-based definition. This type is based on what a specific behavior looks like to an observer, and this method can be used for behaviors that do not have clear functions or unreliable functions (Cooper et al., 2007). Both function- and topography-based definitions should also be behavioral and objective in nature without subjective terms that try to infer states of the target individual (e.g., sad, frustrated, mean-spirited; Baer et al., 1968). Also, as noted earlier, boundary conditions should be created with examples and nonexamples of the target behavior (Cooper et al., 2007).

ABC Recording With ABC recording, observers record the antecedents, behaviors, and consequences in the natural environment. Two types of ABA recording are continuous and narrative. With ABC continuous recording, pre-specified codes (often the result of an indirect FBA) are utilized to observe the context of the behavior(s) of interest. This method is often conducted with a time sampling protocol such as whole- or partial-interval recording (Cooper et al., 2007).

With ABC narrative recording, data are collected in a less structured manner (i.e., without pre-determined codes), only when the problem behavior or other target behaviors occur (Cooper et al., 2007). An example of ABC narrative recording is the Functional Behavioral Assessment Observation Form (FBAOF; Steege & Watson, 2009), which includes a table whereby observers record the date and time, setting events, antecedents, behaviors, consequences, and related staff member associated with each instance of problem behavior. ABC narrative recording requires little training and is flexible and relatively easy to conduct (Thompson & Borrero, 2011). One issue associated with ABC narrative recording is, since only the context surrounding problem behaviors is recorded, it is possible that the same patterns of contextual variables could occur just as often or more frequently with other behaviors (Cooper et al., 2007). As such, teams should carefully use ABC narrative recordings and avoid making misleading decisions based on correlational data.

Scatterplots Scatterplots are recordings of target behaviors and the times and dates of which they occur (Cooper et al., 2007). Rather than utilizing a trained staff member to directly observe behaviors, scatterplot data are typically collected as records of tallies that can be completed by teachers and caregivers as a part of their normal routine. Teams look to identify patterns within scatterplot data, and conditional probabilities (i.e., the likelihood that the problem behavior occurs in a specific environment and under specific conditions) can be calculated (Dufrene et al., 2007). Scatterplots can be useful in determining when to conduct ABC recording (i.e., the times most and least associated with problem behaviors); however, there is limited research on the utility of scatterplots and gathering accurate data may be difficult (Thompson & Borrero, 2011).

Direct Behavior Ratings Direct behavior ratings (DBRs) represent a combination of direct observation and rating scales methods. At the end of a predetermined period of time, the observer rates the individual on specific behaviors. DBRs can be used to assess several behaviors at once, with recommended scales of 5–10 gradients (Chafouleas, Christ, & Riley-Tillman, 2009). For

example, a parent could use a Likert scale to rate the amount of time the child was compliant during bedtime routine. While DBRs are not as reliable as SDO methods, they still offer a method that can be considered to have utility in practice (Briesch et al., 2010). DBRs benefit from their ease of use and ability to have multiple informants use them in multiple natural settings. Moreover, DBRs can be completed by someone naturally occurring in the environment which might mitigate possible changes in behavior due to an observer being present. DBRs can also be personalized to the target individual's behaviors which cannot be said of formal rating scales (Chafouleas et al., 2009). Just like rating scales, however, DBRs are subject to reputational biases and rater reliability problems (Briesch et al., 2010). Like most of the methods discussed, DBRs should be used in conjunction with other methods to create a holistic report of the target individual.

Functional Analysis

Functional analysis (FA) refers to a variety of methods in which the antecedents and consequences of target behaviors are systemically and experimentally manipulated (Cooper et al., 2007). FA is the most intensive of the FBA approaches. FA procedures require experimenters to "systematically vary stimuli, one at a time, in an attempt to determine which environmental stimuli covary most often with the CB [challenging behavior]" (Matson & Williams, 2014, p. 59). The term was coined by Skinner (1953), who discussed the demonstration of experimental functional relationships of cause and effect between variables. Two foundational studies have led to a vast literature of research studies on FA procedures and associated function-based interventions (Carr & Durand, 1985; Iwata, Dorsey, Slifer, Bauman, & Richman, 1982/1994)

Iwata and colleagues (1982/1994) published the seminal article on FA, as they assessed the problem behavior of nine children with developmental disabilities who engaged in self-injurious behavior (SIB). Four conditions were implemented, including social disapproval, academic demand, unstructured play, and alone. In the social disapproval (attention) condition, the participants were instructed to play with toys while the experimenter pretended to read. Contingent on problem behavior, the experimenter expressed disapproval or concern and utilized "brief physical contact" (Iwata et al., 1994, p. 202) in close proximity to the participant. In the academic demand (escape) condition, the experimenter presented an academic task at a table using a series of prompts to promote engagement with the task. Contingent on SIB, the academic demand was removed and the experimenter turned away from the participant for 30 s. During the unstructured play condition, a variety of toys were available in the room, the experimenter delivered praise every 30 s contingent on the absence of SIB, and relatively minor forms of SIB were ignored. The participant was placed in a bare room with "minimal amounts of stimulation" (Iwata et al., 1994, p. 203) in the alone condition, which was instituted to determine if automatic reinforcement was maintaining the problem behavior. Within this methodology, the attention, alone, and escape conditions are compared to the unstructured play (control condition) to determine a function of the problem behavior.

Carr and Durand (1985) conducted an influential study on Functional Communication Training (FCT) that involved the systematic manipulation of contextual variables. In their study with four children engaging in a variety of problem behaviors in a day school, Carr and Durand (1985) utilized four conditions to determine if behaviors were maintained by attention and escape. The conditions either included easy matching-to-sample tasks or a difficult vocabulary task that was decided upon based on the participants' performance on a standardized assessment, as well as attention delivered by the experimenter during 100% or 33% of intervals. As such, the researchers compared the participants' rates of problem behaviors during Easy 100 (i.e., easy task and 100% attention), Easy 33, Difficult 100, and Difficult 33 sessions. In Study 2, FCT was used to intervene on the problem behaviors based on the results of the FA.

Since the Iwata et al. (1982/1994) and Carr and Durand (1985) studies, a number of studies have been conducted with variations of FA procedures to address a variety of problem behaviors in many settings (Hanley et al., 2003). Advantages of FA include that it is most likely to yield a clear function of the problem behavior (as compared to indirect and direct descriptive FBA methods), it results in quantifiable data and has high treatment validity (i.e., the results can directly inform treatment), and the contextual variables are systematically manipulated to determine functional relationships (Cooper et al., 2007).

Although FA is associated with advantages and has been repeatedly demonstrated to be useful in the design of function-based interventions, a number of disadvantages of the traditional FA limit its feasibility in a variety of settings. One of the major disadvantages of FA is that it is time-consuming. For example, the FA in the Iwata et al. (1982/1994) study averaged 30 sessions (range 24–53) over 8 days (range 4–11). FA also requires at least two experimenters with considerable training in applied behavior analysis and single case design in order to properly conduct the procedures (Matson & Williams, 2014). There are also problems associated with allowing access to reinforcement contingent on problem behavior, particularly if the behavior is dangerous (Cooper et al., 2007). FA procedures may result in a manufactured function, and challenging behaviors could increase after FA sessions. There are also a limited range of problem behaviors that can be subject to an FA, as low-frequency, high-intensity behaviors would not be appropriate to analyze using an FA (Cooper et al., 2007). Undifferentiated results can occur if the behavior is similar across all conditions or is highly variable, which is particularly problematic given the time-consuming nature of FA (Cooper et al., 2007). FAs are also conducted in an analog setting, which is often not representative of the natural environment. Researchers have indicated that the traditional FA is not feasible to implement in a number of settings, and more resource-efficient versions of FA have been developed (Steege & Watson, 2009).

Structural Analysis Structural analysis refers to a variety of procedures involving systematically manipulating antecedent conditions of problem behaviors and recording whether the challenging behaviors are evoked (Steege & Watson, 2009). Structural analyses can be particularly helpful when specific, idiosyncratic antecedents need to be tested (Wacker, Berg, Harding, & Cooper-Brown, 2011). Although the antecedents are implemented to evoke the problem behaviors, structural analyses do not involve the reinforcement of problem behaviors. One disadvantage of structural analysis is that it also occurs in an analog setting that may not represent the setting of interest (Steege & Watson, 2009).

Brief Functional Analysis (BFA) BFA involves fewer and briefer sessions than the traditional FA. Northup et al. (1991) conducted a BFA with three participants demonstrating aggressive behavior. Using alone, escape, social attention, and tangible (i.e., access to a preferred item for 15–20 s) conditions, the researchers identified a clear function for each participant. They also implemented a contingency reversal condition, in which a functionally equivalent replacement behavior was reinforced. In this study, BFAs were conducted within one 90 min outpatient session, with condition sessions lasting 5–10 min each (Northup et al., 1991). A number of studies have been conducted demonstrating the use of BFA in different settings to design function-based interventions (e.g., Cihak, Alberto, & Fredrick, 2007). Also, Tincani, Castrogiavanni, and Axelrod (1999) demonstrated that BFA and traditional FA yielded similar results with regard to the function of challenging behaviors of three adults with developmental disabilities; however, the BFA took an average of 20% of the time of the FA.

The major advantages of the BFA are its feasibility and brevity, as it can be more efficiently conducted than the FA (Northup et al., 1991; Tincani et al., 1999). With regard to disadvantages, BFA may not be appropriate to detect automatic reinforcement as a maintaining variable, as

additional sessions within a more traditional FA may be required (Tincani et al., 1999). Also, since BFA is typically conducted in one session, any pre-session setting events or motivating operations may significantly alter the results and influence decision-making (Tincani et al., 1999). BFA requires rapid discrimination within each session to determine which condition is in effect, which may not be appropriate for all clients (Cihak et al., 2007). Finally, BFA may sometimes result in an unclear function after brief, rapidly alternated sessions. Vollmer, Marcus, Ringdahl, and Roane (1995) conducted a BFA as part of a systematic FA process leading to more intense FA procedures for the 14 out of 20 participants whose BFA results were unclear.

Trial-Based Functional Analysis (TBFA) TBFA is distinguished by discrete trials that are incorporated into the typical classroom routine (e.g., conducting a demand trial during the completion of academic tasks; tangible trials during lunch or snack times). Trials are distributed throughout the day and consist of two parts: a test part similar to a traditional FA when the relevant motivating operation is created and a control part when free access to the functional reinforcer is allowed (Larkin et al., 2016). Problem behaviors are recorded for test and control parts separately and compared to determine a function.

Sigafoos and Saggers (1995) demonstrated the utility of TBFA with two students who demonstrated aggressive behavior. During the test part of the attention trials, the staff member sat near the child, turned away, and ignored the student for up to 60 s. Contingent on problem behavior, the test part was immediately stopped, and the student was allowed 60 s of uninterrupted attention (the control part). The control part also occurred if the test part lasted for 60 s without problem behavior. Tangible trials included a test trial in which edible items or drinks were within view but out of the reach of the student, and the child was instructed that they could have access to the item in 1 min. The control condition (i.e., access to the item for 60 s) was implemented contingent on 60 s without problem behavior or upon the demonstration of problem behavior. Finally, the task condition involved an academic task being presented to the student and a verbal or physical prompt to complete the task every 10 s if the child did not interact with the task. After 60 s or continent on problem behavior, the task was removed during the control part.

A growing body of literature supports the use of TBFA with different staff members conducting the assessments (Rispoli, Ninci, Neely, & Zaini, 2014). A major advantage of TBFA is its feasibility and ecological validity, as it is conducted within the natural environment by teachers or other stuff who require little training (Larkin et al., 2016). Function-based interventions designed using the results of TBFA have repeatedly been demonstrated as effective, and teachers who conduct TBFAs often indicate that the procedures are socially valid (Austin, Groves, Reynish, & Francis, 2015). Also, the distributed trials of TBFA make it less susceptible to pre-session motivating operations, such as with BFA.

A number of disadvantages are also associated with TBFA. Researchers have debated whether teachers and other school staff should be tasked with being the primary experimenters in TBFAs (Rispoli et al., 2015). There is some evidence that teachers may have difficulty fitting the TBFA into their daily routine, and some teachers have indicated that reinforcing problem behaviors in the presence of other students may be problematic (Rispoli et al., 2015). Although studies have demonstrated agreement between identified functions based on TBFA and FA, false positives and false negatives are possible (Rispoli et al., 2014).

Interview-Informed Synthesized Contingency Analysis (IISCA) IISCA (Hanley, Jin, Vanselow, & Hanratty, 2014) has recently emerged as a potentially advantageous method of FA. This method involves a 30–45 min open-ended interview with parents and a 15–30 min observation of the client, which are then used to inform the control (i.e., hypothesized reinforcer is continuously available) and test conditions (i.e., hypothesized reinforcer is removed every 30 s and reinstated contingent on problem

behavior) of a time-efficient functional analysis. IISCA has been extended to the home environment (Rose & Beaulieu, in press) and used to integrate children into the general education school environment (Taylor, Phillips, & Gertzog, 2018).

IISCA is an efficient assessment, as it takes an average administration time of 25 min (Jessel, Hanley, Ghaemmaghami, & Metras, in press). It is also personalized to the client, as only relevant conditions based on the results of the interview and observation are tested (Hanley et al., 2014). This also allows for synthesized functions (e.g., with two or more hypothesized functions acting simultaneously) to be detected, as these may be similar to the natural environment (Hanley et al., 2014).

A major disadvantage of IISCA is that, in order to determine a function, the problem behavior has to be immediately responsive to the different motivating operations in the test and control conditions (Jessel et al., in press). As such, IISCA may not be appropriate for long-duration behaviors, such as tantrums, or for low-frequency behaviors (Jessel et al., in press). Researchers have also indicated that interviews may need to be semi-structured (i.e., future research may need to include sample questions and prompts) so that the required information regarding the context of problem behaviors is reliably obtained (Jessel et al., in press).

Conclusion

FBA is an evidence-based approach to assessing problem behavior and informing treatment planning (Hanley et al., 2003). FBA methods are applicable to the wide range of maladaptive behaviors that may be displayed by individuals with ID, including individuals also experiencing a mental health disorder. By clearly defining, observing, and measuring problem behaviors; collecting data on the relationship between antecedents, problem behaviors, and consequences; and determining functional relationships between the environment and problem behavior, effective interventions can be developed to manipulate the environment in such ways to promote adaptive behavior and improve social, emotional, and adaptive behavior outcomes for individuals with ID.

References

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.

Araten-Bergman, T., & Werner, S. (2017). Social workers' attributions towards individuals with dual diagnosis of intellectual disability and mental illness. *Journal of Intellectual Disability Research, 61*, 155–167.

Austin, J. L., Groves, E. A., Reynish, L. C., & Francis, L. L. (2015). Validating trial-based functional analyses in mainstream primary school classrooms. *Journal of Applied Behavior Analysis, 48*, 274–288.

Axelrod, M. I. (2017). *Behavior analysis for school psychologists*. New York, NY: Routledge.

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., 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*, 217–230.

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*(3), 408–421.

Carr, E. G., & Durand, V. M. (1985). Reducing behavior problems through functional communication training. *Journal of Applied Behavior Analysis, 18*, 111–126.

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.

Cervantes, P. E., & Matson, J. L. (2015). Comorbid symptomology in adults with autism spectrum disorder and intellectual disability. *Journal of Autism and Developmental Disorders, 45*, 3961–3970.

Chafouleas, S. M., Christ, T. J., & Riley-Tillman, T. C. (2009). Generalizability of scaling gradients on direct behavior ratings. *Educational and Psychological Measurement, 69*(1), 157–173.

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.

Conners, C. K. (2008). *Conners 3rd edition (Conners 3)*. North Tonawanda, NY: Multi-Health Systems.

Cooper, J., Heron, T., & Heward, W. (2007). *Applied behavior analysis* (2nd ed.). Columbus, OH: Prentice Hall/Merrill.

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. *Journal of Child Psychology and Psychiatry, 43*, 1087–1098.

Didden, R., Korzilius, H., Oorsouw, V., & Sturmey, P. (2006). Behavioral treatment for challenging behavior in individuals with mild mental retardation: A 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. (2017). 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.

Emerson, E., & Einfeld, S. (2010). Emotional and behavioural difficulties in young children with and without developmental delay: A bi-national perspective. *Journal of Child Psychology and Psychiatry, 51*, 583–593.

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.

Gresham, F. M., Watson, T. S., & Skinner, C. H. (2001). Functional behavioral assessment: Principles, procedures, and future directions. *School Psychology Review, 30*(2), 156–172.

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*, 16–36.

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.

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. A., &, DeLeon, I. G. (1996). *The Functional Analysis Screening Tool (FAST)*. Unpublished Manuscript. University of Florida: Gainesville, FL.

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.

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)

Jessel, J., Hanley, G. P., Ghaemmaghami, M., & Metras, R. (in press). An evaluation of the single-session interview informed synthesized contingency analysis. *Behavioral Interventions, 34*, 62–78.

Kamphaus, R. W., & Reynolds, C. R. (2015). *Behavior assessment system for children—Third edition (BASC-3): Behavioral and emotional screening system (BESS)*. Bloomington, MN: Pearson.

Kelley, M. E., LaRue, R. H., Roane, H. S., & Gadaire, D. M. (2011). Indirect behavioral assessments: Interviews and rating scales. In W. W. Fisher, C. C. Piazza, & H. S. Roane (Eds.), *Handbook of applied behavior analysis*. New York, NY: The Guilford Press.

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.

Lewis, T. J., Scott, T. M., & Sugai, G. (1994). The problem behavior questionnaire: A teacher based instrument to develop functional hypotheses of problem behavior in general education classrooms. *Diagnostique, 19*(2–3), 103–115.

Lewis, T. J., Scott, T. M., Wehby, J. H., & Wills, H. P. (2014). Direct observation of teacher and student behavior in school settings: Trends, issues and future directions. *Behavioral Disorders, 39*(4), 190–200.

March, R. E., Horner, R. H., Lewis-Palmer, T., Brown, D., Crone, D., Todd, A. W., et al. (2000). *Functional assessment checklist: Teachers and staff (FACTS)*. Eugene, OR: Educational and Community Supports.

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: Scientific Publishers.

Matson, J. L., & Williams, L. W. (2014). Functional assessment of challenging behavior. *Current Developmental Disorders Reports, 1*, 58–66.

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.

Murdock, S. G., O'Neill, R. E., & Cunningham, E. (2005). A comparison of results and acceptability of functional behavioral assessment procedures with a group of middle school students with emotional/behavioral disorders. *Journal of Behavioral Education, 14*, 5–18.

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.

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.

Reed, H., Thomas, E., Sprague, J. R., & Horner, R. H. (1997). The student guided functional assessment interview: An analysis of student and teacher agreement. *Journal of Behavioral Education, 7*, 33–49.

Rispoli, M., Ninci, J., Burke, M. D., Zaini, S., Hatton, H., & Sanchez, L. (2015). Evaluating the accuracy of results for teacher implemented trial-based functional analysis. *Behavior Modification, 39*(5), 627–653.

Rispoli, M., Ninci, J., Neely, L., & Zaini, S. (2014). A systematic review of trial-based functional analysis of challenging behavior. *Journal of Developmental and Physical Disabilities, 26*, 271–283.

Rojahn, J., Rowe, E. W., Kasdan, S., Moore, L., & van Ingen, D. J. (2011). Psychometric properties of the *Aberrant Behavior Checklist*, the *Anxiety Depression and Mood Scale*, the *Assessment of Dual Diagnosis*, and the *Social Performance Survey Schedule* in adults with intellectual disabilities. *Research in Developmental Disabilities, 32*, 2309–2320.

Rose, J. C., & Beaulieu, L. (in press). Assessing the generality and durability of interview-informed functional analyses and treatment. *Journal of Applied Behavior Analysis, 52*(1), 271–285.

Shapiro, E. S. (2004). *Academic skills problems workbook (rev.)*. New York, NY: Guilford Press.

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.

Skinner, B. F. (1953). *Science and human behavior*. New York, NY: Macmillan.

Steege, M. W., & Watson, T. S. (2009). *Conducting school-based functional behavioral assessments (second edition): A practitioner's guide*. New York, NY: The Guilford Press.

Taylor, S. A., Phillips, K. J., & Gertzog, M. G. (2018). Use of synthesized analysis and informed treatment to promote school reintegration. *Behavioral Interventions, 33*, 364–379.

Thompson, R. H., & Borrero, J. C. (2011). Direct observation. In W. W. Fisher, C. C. Piazza, & H. S. Roane (Eds.), *Handbook of applied behavior analysis*. New York, NY: 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.

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*, 561–576.

Wacker, D. P., Berg, W. K., Harding, J. W., & Cooper-Brown, L. J. (2011). Functional and structural approaches to behavioral assessment of problem behavior. In W. W. Fisher, C. C. Piazza, & H. S. Roane (Eds.), *Handbook of applied behavior analysis*. New York, NY: The Guilford Press.

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.

Checklists and Structured Interviews

11

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A psychiatric diagnosis is based on comprehensive information about signs, symptoms, and problems and the duration and frequency of the problems. Typically, diagnosis is based on descriptions of the person's problems and experiences obtained in an interview and/or by use of self-rating checklists (Othmer, Othmner, & Othmner, 2005). Thus, diagnostic systems (e.g., DSM-5, American Psychiatric Association, 2013; ICD-10, World Health Organization, 1992, 1993) rely heavily on descriptions of the subjective experience of the individuals who are being diagnosed. The reliability of ordinary psychiatric diagnostic for individuals with intellectual disability (ID) has therefore been questioned (Einfeld & Aman, 1995).

Most individuals with ID, including individuals with good verbal abilities, have difficulties in self-report of experience and inner states (Helverschou, Bakken, & Martinsen, 2011; Underwood, McCarthy, Chaplin, & Bertelli, 2015) and hence in reporting information needed to identify a psychiatric disorder. In individuals with ID, psychiatric disorders may have to be identified by observable behaviors, and there is a need to recognize the possible impact of ID in modifying the symptoms of psychiatric illness. This has been extensively described in DC-LD (Royal College of Psychiatrists, 2001) and in DM-ID – 2 (Fletcher, Barnhill, & Cooper, 2016). In persons with both autism spectrum disorders (ASD) and ID, the diagnostic process is further complicated by the combination of the comprehension and communication difficulties related to autism and the problems in self-report related to ID.

In order to assist the challenges of identifying psychiatric disorders among individuals who have difficulties reporting about their problems, researches started in the 1980s to develop normed measures to assess mental health issues and behavior disorders among persons with ID. This chapter provides an overview of these measures and the rationale for their development. Strengths and weakness of various assessment instruments and checklists are covered. The focus is a broad

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overview of developments and trends since specific details on specific conditions will be covered in subsequent chapters. The more general aspects of assessment of mental health issues, such as adaptations and considerations due to verbal and cognitive deficits among individuals with ID, are covered in Chap. 8.

Several checklists and instruments have been developed to assist the process of assessment of challenging behavior and psychiatric disorders in individuals with ID. The different checklists and structured interviews developed may be categorized into the following subcategories: (1) measures of challenging behaviors, (2) instruments to identify specific psychiatric disorders among individuals with ID generally, and (3) measures of psychiatric disorders among individuals with ASD and ID, due to the specific challenges that this group represents. Few instruments specifically for those with ASD and ID have been developed, and these will therefore be presented in more detail. In addition, the chapter will discuss the use of checklists and instruments not particularly adapted to individuals with ID or ASD. Instruments developed for the general population have been used to measure behavioral problems and aspects of psychiatric comorbidity in this group, and the usefulness and pitfalls of such use will be discussed. The chapter focuses on checklists used for adolescents and adults as mental disorders including the main categories, psychoses, mood disorders, and anxiety and stress disorders, which are most likely to occur in adolescents and adults. The checklists are presented in each section in chronological order according to the year first published. Overview of the checklists and interviews are presented in Tables 11.1, 11.2, and 11.3 encompassing type of tool, outcome measures, corresponding criteria, population, and psychometric properties.

Checklists for Challenging Behavior

There are numerous checklists for the assessment of challenging behavior in individuals with ID. The association between challenging behavior and mental health issues has been identified in numerous studies (Bowring, Totsika, Hastings, Toogood, & Griffith, 2017; Kearney & Healy, 2011; Myrbakk & von Tetzchner, 2008a, 2008b; Rojahn, Matson, Naglieri, & Mayville, 2004) – as have associations with autism spectrum disorder (Bowring et al., 2017; McCarthy et al., 2010; Rojahn, Matson, Lott, Esbensen, & Smalls, 2001), limited communication ability, somatic health problems, and other sources of emotional discomfort (Bowring et al., 2017). While checklists for challenging behavior may be useful, challenging behavior may also be viewed as a symptom rather than an explanation, and further investigation of possible causes is recommended.

Level of ID as well as other conditions such as autism spectrum disorder or genetic conditions may influence the expression of challenging behavior and thus the appropriateness of various checklists (Bowring et al., 2017; Flynn et al., 2017; Rojahn et al., 2001). In individuals with severe to profound ID, behavior is typically less differentiated than in individuals with mild or moderate ID (Myrbakk & von Tetzchner, 2008a, 2008b). Checklists found to be valid in individuals with mild ID may therefore not necessarily be appropriate for individuals with severe to profound ID and vice versa.

The *Disability Assessment Schedule* (DAS) (Holmes, Shah, & Wing, 1983) was constructed to assess the level of functioning in individuals with intellectual disabilities. It includes a behavioral problems scale (DAS-B) which has been used to measure challenging behavior (McCarthy et al., 2010). While showing good internal consistency (Tsakanikos, Underwood, Sturmey, Bouras, & McCarthy, 2011), questions remain regarding its validity as no association has been identified between this measure challenging behavior and comorbid psychopathology (McCarthy et al., 2010), a finding contrary to other studies in the field. Furthermore, concurrent validity with other measures is yet to be explored.

The *Aberrant Behavior Checklist (ABC)* was one of the earliest checklists developed for individuals with ID (Aman, Singh, Stewart, & Field, 1985a, 1985b). Though it was originally

Table 11.1 Checklists and rating scales assessing challenging behavior in individuals with ID

Year published	Type of tool	Outcome	Population	Psychometric properties
1982	DAS-B: Structured interview with parents or care staff. 14 items rated on a three-point scale	Two underlying factors: Disruptive/distractive, antisocial/delinquent	Individuals with ID	Holmes et al. (1983): Good inter-rater and test-retest reliability McCarthy et al. (2010): Good internal consistency (0.87) Tsakanikos et al. (2011): Good internal consistency (0.87)
1985	ABC: Informant completed checklist. 58 items rated on a four-point scale	Five subscales: Irritability/agitation/crying, lethargy/social withdrawal, stereotypic behavior, hyperactivity/noncompliance, inappropriate speech.	Individuals with ID	Aman et al. (1985a, 1985b): Confirmed factor structure, good internal consistency (alpha coeff. 0.86–0.95). High test-retest reliability. Demonstrated criterion group validity, convergent validity with other scales, divergent validity with IQ
				Newton and Sturmey (1988): Confirmed factor structure, good internal consistency (alpha coeff. 0.94–0.92)
				Sturmey and Bertman (1994): Good concurrent validity with the Reiss Screen
				Ono (1996): Confirmed similar factor structure, good internal consistency (alpha coeff. 0.85–0.95), good test-retest reliability (4 weeks, 0.84–0.90), good inter-rater reliability (0.58–0.78)
				Walsh and Shenouda (1999): Good concurrent validity with the Reiss Screen and the Adaptive Behavior Scales – part II
				Rojahn et al. (2003): Good concurrent validity with the BPI-01
				Flynn et al. (2017): In a systematic review, excellent internal consistency and test-retest reliability and good inter-rater reliability was reported for severe ID, as well as good criterion and construct validity
1988	RSMB: Informant completed checklist. 38 items rated on a three-point scale	Eight subscales: Aggressive behavior, autism, psychosis, paranoia, depression (behavioral signs), depression (physical signs), dependent personality disorder, avoidant personality disorder. Six maladaptive behavior items	Individuals with mild to moderate ID	Sturmey, Burcham, and Perkins (1995): Good internal consistency (0.50–0.85), varying inter-rater reliability (0.07–0.83), modest to good test-retest reliability (2 weeks, 0.01–0.69) Sturmey et al. (1996): Factor structure not confirmed.

(continued)

Table 11.1 (continued)

Year published	Type of tool	Outcome	Population	Psychometric properties
				Havercamp and Reiss (1997): Factor structure confirmed
				Gustafsson and Sonnander (2002): Excellent internal consistency (0.90), good inter-rater reliability (0.60). Good criterion validity
				Flynn et al. (2017): Good internal consistency
1991	DASH/DASH-II: Informant completed checklist. 84 items rated for frequency, duration, and severity, all on a three-point scale	Thirteen subscales: Anxiety, depression, mania, PDD/autism, schizophrenia, stereotypies, self-injury, elimination, eating, sleep, sexual, organic, impulsivity/challenging behavior	Individuals with severe/profound ID	Matson, Gardner, et al. (1991)/ Matson, Coe, et al. (1991): Good inter-rater reliability, varying internal consistency (0.20–0.84). Confirmed factor structure
				Paclawskyj et al. (1997): Good concurrent validity with the ABC
				Flynn et al. (2017): In a systematic review, DASH-II was found to have excellent test-retest and inter-rater reliability and good internal consistency. Excellent criterion and construct validity, good content validity
1997	BPI-01/BPI-S: Informant completed checklist. 49/30 items rated on a five-point scale for frequency and a three-point scale for severity	Three subscales: Self-injurious behavior, stereotyped behavior, aggressive/destructive behavior	Individuals with ID	Rojahn et al. (2001): Good inter-rater reliability (0.59–0.96) for subscales, good internal consistency for subscales (0.61–0.82), and the full scale (0.83). Confirmed factor structure
				Rojahn et al. (2003): Good concurrent validity with the ABC
				González et al. (2009): Good internal consistency for subscales (0.40–0.87), good inter-rater reliability for subscales (0.41–0.80), and varying test-retest reliability for subscales (0.28–0.70)
				Rojahn et al. (2012b): Good concurrent validity with the ABC, DASH-II, and the Nisonger Child Behavior Rating Form. Good internal consistency of subscales for both BPI-01 (0.73–0.92) and BPI-S (0.68–0.89)

(continued)

Table 11.1 (continued)

Year published	Type of tool	Outcome	Population	Psychometric properties
				Bowring et al. (2017) (BPI-S): Good internal consistency for frequency (0.89) and severity (0.77), with subscales varying from 0.63 to 0.87

constructed to assess behavioral and psychopharmacological treatment effects in individuals with ID, it has been found to be useful for general assessment of challenging behavior (Aman, 2012). It is by far the most thoroughly researched checklist for challenging behavior, with several hundred studies investigating its psychometric properties, and it is currently available in more than 25 languages (Aman, 2012).

The ABC seems to be applicable for individuals with mild ID and moderate ID (Aman, 2012), as well as individuals with severe to profound ID (Flynn et al., 2017). It has been successfully applied in children, adolescents, and adults (Aman, 2012). The ABC is easy to administer, and scores are easily calculated. Its availability, applicability, and usefulness across age ranges and levels of ID are likely to be contributing to its wide usage. Psychometric properties have been found to be varying from satisfactory to excellent (Aman et al., 1985a, 1985b; Flynn et al., 2017; Ono, 1996), and its factor structure has been confirmed in numerous studies (Aman et al., 1985a; Newton & Sturmey, 1988; Ono, 1996). True to its original intention, the ABC seems to be sensitive to change and treatment effects and may therefore be a good instrument to aid in the evaluation of intervention (Aman, 2012).

One common misuse of the ABC is the publication or interpretation of a "total ABC score." Calculation and use of such a score is strongly discouraged by the ABC's authors (Aman, 2012), as it disregards the empirical origins and factor structure of the instrument and therefore lacks construct validity. Only summation scores on the subscales of the ABC may be interpreted meaningfully.

Though it has strong psychometric properties, a limitation of the ABC is the lack of norms and cutoff scores, necessitating an idiographic approach in individual clinical assessments and making the instrument challenging to use for diagnostic purposes. Higher scores are generally found to be associated with measures of psychiatric disorder (Katz, Berry, & Singh, 1997; Rojahn & Helsel, 1991; Sturmey & Ley, 1990). However, the lack of cutoff values on the subscales may make it challenging for clinicians to discern what constitutes a high score and thereby what behavior should be considered sufficiently challenging to warrant further investigation. Furthermore, diagnostic implications of the subscales may also be challenging to interpret, as their associations with specific diagnostic categories are not necessarily obvious. However, subscales measuring irritability/agitation/crying and lethargy/social withdrawal both seem to be associated with mood disorders (Flynn et al., 2017).

The *Reiss Screen for Maladaptive Behavior* (RSMB) (Reiss, 1988) is another of the older, well-researched scales for evaluation of challenging behavior (Matson, Belva, Hattier, & Matson, 2012). It was designed to identify individuals to be referred for further, psychiatric evaluation. (The scale is also presented in the next section focusing on mental health assessment.) The Reiss Screen has 38 items which are scored on a three-point scale, and all items are related to a possible symptom. The checklist provides cutoff values (Reiss, 1988), and studies have found scores on the Reiss Screen to correspond well with scores on the ABC (Walsh & Shenouda, 1999). As an assessment tool, the Reiss Screen has been criticized for being in need of a more comprehensive set of items (Prout, 1993), but it seems to be useful as a screening tool. A separate version for children, Reiss Scale for Children's Dual Diagnosis (Reiss & Valenti-Hein, 1994), also

Table 11.2 Checklists and rating scales assessing mental health problems in individuals with ID

Year published	Type of tool	Outcome	Corresponding diagnostic criteria	Population	Psychometric properties
1988	PIMRA: Structured interview, two versions; self-rated and informant completed. 56 items rated present/not present	Eight subscales (7 items each): schizophrenia, adjustment disorder (D), anxiety, psychosexual D, affective D., somatoform D, personality D, and inappropriate adjustment	DSM-III	Adults with mild-moderate ID	Gustafsson and Sonnander (2005): 101 adults in community residences and adults in specialized mental health services. PIMRA informant version Internal consistency (Cr. Alpha): moderate – acceptable for 5 of 7 mental health subscales, 2 not acceptable. Criterion validity: Sensitivity and specificity compared to DSM were 0.53 and 0.94, respectively, for psychosis and 0.75 and 0.76 for anxiety.
					Matson et al. (1984): 110 adults, clinical mental health sample. Internal consistency self-rated 0.85 and informant completed 0.83
1988	RSMB: Screening checklist. Informal informant completed. 38 items rated on a three-point scale. Each item defined and is followed by example	Seven subscales: aggression, psychosis, paranoia, depression, dependent personality disorder, and avoidant personality disorder	DSM-III-R	Adults with mild-moderate ID	Gustafsson and Sonnander (2002): 140 adults with ID in communities + 17 in a clinical mental health services sample. Reliability inter-rater agreement 0.60. Internal consistency (Cr. alpha) 0.90. Criterion validity: Cohen's kappa $k = 0.58$
					Kishore et al. (2010): 56 adolescents and adults. RSMB differentiated well between those with and without psychiatric diagnoses, using ICD-10 as external validator
1991	DASH/DASH-II: Diagnostic scale 84 items rated for duration and frequency, and symptoms on a three-point scale	DASH-II has 13 subscales, anxiety, depression, mania, autism, schizophrenia, stereotypies, self-injury, elimination, eating, sleep, sexual, organic, impulse	DSM, and research on ID and mental disorders	Adults with severe-profound ID	Matson et al. (1991): 506 adults, severe or profound ID. Inter-rater reliability: severity = 0.96, duration = 0.95, anxiety and sleep = 0.98, less than 0.60 = depression + impulse control. Internal consistency Cr. alpha ranging from 0.84 (elimination) to 0.20 (schizophrenia)

1993	PAS-ADD clinical interview: Diagnostic tool Patient and third-party interviews	13 domains: physical illness, eating and weight change, sleep, energy, tension, worries and fears, depression, substance abuse, auditory hallucinations, other hallucinations, thought interference/ delusions, and observational items covering: catatonia, behavior, speech and affect	ICD-10 and DSM-IV	Adults with ID	Moss et al. (1993): 25 adults in a selected sample for this study. A mixture of mentally healthy and unhealthy adults with ID. To independent interviewers. Inter-rater reliability. Kappa for all items = 0.77. Most questions had alpha > 0.60 Moss et al. (1997): 95 adults with ID in a clinical mental health sample. Mean IQ = 37.6. Two independent interviewers. Factor analysis extracted 5 factors, accounting for 61.5% of the items (expansive mood, psychosis, negative symptoms, depression, anxiety)
1997	Mini PAS-ADD: Checklist Trained informant completed checklist 86 items	7 subscales: depression, anxiety and phobias, mania, OCD, psychosis, unspecified disorder, and autism	ICD-10	Adults with ID	Prosser et al. (1998): 68 persons with ID (age + level of ID not specified) in a clinical mental health services sample. Internal consistency Cr. Alpha; for all subscales 0.60. Rater agreement: 91% agreement between the trained informant raters and psychiatrists. Trained informant raters scored averagely higher symptom burden than psychiatrists.
					Myrbakk and von Tetzchner (2008a). 181 adolescents and adults with all levels of ID (mental health services and in community services): 29.6% of the sample scored above cutoff compared to 44.7% with the DASH-II

(continued)

Table 11.2 (continued)

Year published	Type of tool	Outcome	Corresponding diagnostic criteria	Population	Psychometric properties
1998	PAS-ADD Checklist: Untrained rater completed checklist 29 items on a four point scale	7 subscales: appetite and sleep, tension and worry, phobias and panics, depression and hypomania, obsessions and compulsions, psychoses, autism	ICD-10 & DSM-IV	Adults with ID	Moss et al. (1998): Study 1: 201 adults in community settings. Study 2: 66 adults in a mixed community & hospital sample, all levels of IQ in both samples. Both untrained raters. Factor analysis: 8 factors explained 65.3%. 7 of 8 factors were consistent with the subscales defined. Internal consistency Cr. Alpha overall was god, but somewhat lower for some of the subscales. Reliability: overall mean Kappa 0.42 (low)
					Hatton and Taylor (2008): 1115 adults randomly divided in two groups. Group A scores gave an optimal 7-factor structure, accounted for 61.25%. Group B revealed mediocre to poor fit. Further analyses confirmed this finding
1998	The ADD Assessment scale 79 items Screening tool. Third-party information. trained interviewers Duration and frequency and symptoms on a three-point scale	13 subscales: mania, depression, anxiety, PTSD, substance abuse, somatoform disorder, dementia, conduct disorder, pervasive developmental disorder, schizophrenia, personality disorder, eating disorder, sexual disorder	DSM-IV	Adults with mild or moderate ID	Matson and Bamburg (1998): 101 adults with mild or moderate ID in community settings. 18% mild ID, the rest moderate. Internal consistency: Cr. Alpha 0.93 for all items. Sub-scales variated 0.77–0.95
					Inter-rater reliability: correlations 0.98 for all items. Test-retest reliability scored for 67 participants: correlations 0.98 for all items
					Daniel, Passmore, and Sewell (2003): 58 adults with mild or moderate ID. Correlations of scores of the MMPI-168(L) and ADD found that (except for Mania subscale) the correlations between similar subscales in the two tools were low. MMPI-168(L) is self-rated

2005	DBC-A Carer-completed checklist 107 items scored on a three-point scale	6 subscales: disruptive, communication and anxiety disturbance, self-absorbed, antisocial, depressive, social relating	Definitions of disturbed behavior and emotion in Graham and Rutter (1970)	Adults with ID and/or developmental disabilities	Mohr, Tonge, and Einfeld (2005): 100 files scored retrospectively by two independent raters. Item-by-item correlations = 0.76. 34 adults with moderate-profound ID in community setting. Paid carers. Test-retest reliability correlated 0.75 Validity: high correlation between DBC-A scores, and ABC scores, and PAS-ADD Checklist scores.
					Mohr et al. (2011). 38 adults, all levels of ID. Representative community settings. Pair of independent scorers. Inter-rater reliability: kappa = 0.85 for all items
2008	The P-AID. Checklist 280 items Diagnostic tool Professional carer completed	10 subscales for mental health: dementia, schizophrenia, depression, mania, agora phobia, social phobia, specific phobia, general anxiety, panic anxiety, OCD. Additional 8 scales for behavior problems	DC-LD	Adults with ID	Hove and Havik (2008): Two scorers for 66 adults in a 593 patient sample completed the P-AID. All levels of ID. Internal consistency 0.83–0.93 for mental health subscales; 0.86–0.95 for behavior subscales. Inter-rater reliability: correlations 0.63–0.88. Factor analysis indicated 4 factors explaining 54.9% of the variance. Sensitivity acceptable, specificity poor

Table 11.3 Checklists and rating scales assessing psychiatric symptoms in individuals with ASD and ID

Year published	Type of tool	Outcome	Corresponding diagnostic criteria	Population	Psychometric properties
1994	SAPPA Diagnostic tool, semi-structured interview between trained professional and informant	Psychiatric disorders identified as being absent, possible, or definite	Research diagnostic criteria	Children and adults with ID and/or ASD	No published reports on psychometric properties
2006	ASD-CA Screening checklist interview 84 items 37 items	Five subscales: Anxiety/repetitive behaviors, conduct problems, irritability/behavior excesses, attention/hyperactivity/impulsivity, depressive symptoms	DSM-IV-R and ICD-10	Adults with ASD and ID	Matson and Boisjoli (2008): Inter-rater reliability (*k*) 0.30–0.77; mean = 0.43. Test-retest reliability (*k)* 0.35–0.92, mean = 0.59. Subscales internal consistencies KR-20 confidents 0.44–0.85. Internal consistency KR-20 coefficient of 0.91 LoVullo and Matson (2009): The reliability and validity of each subscale varied widely. No significant differences on mean scores between two ASD and ID groups, with and without psychopathology
2009	PAC Informant completed checklist 42 items	Five subscales: General adjustment problems, psychosis, depression, anxiety, obsessive-compulsive disorder (OCD)	DSM-IV and ICD-10	Adolescents and adults with ASD and ID	Helverschou et al. (2009): All subscales had acceptable and good internal consistency (Cronbach's α) and inter-rater agreement (Cohen's kappa) psychosis $\alpha = 0.89$ and $k = 0.51$, depression $\alpha = 0.85$ and $k = 0.67$, anxiety disorder $\alpha = 0.78$ and $k = 0.58$, OCD $\alpha = 0.88$ and $k = 0.53$, general adjustment problems $\alpha = 0.88$ and $k = 0.66$). Significant difference between groups of ASD + ID with or without psychiatric comorbidity

The Schedule for the Assessment of Psychiatric Problems Associated with Autism (and Other Developmental Disorders), SAPPA; Bolton and Rutter (1994). The Autism Spectrum Disorder-Comorbidity for Adults, The ASD-CA; Matson, Terlonge & Gonzalez (2006), Matson and Boisjoli (2008), Lovullo and Matson (2009). The Psychopathology in Autism Checklist, PAC; Helverschou et al. (2009)

exists. Most studies to date have included individuals with mild to moderate ID, and its psychometric properties have been sparsely explored in individuals with severe to profound ID (Flynn et al., 2017). Questions also remain as to the underlying factor structure of its subscales (Sturmey, Jamieson, Burcham, Shaw, & Bertman, 1996).

The *Diagnostic Assessment of the Severely Handicapped-II (DASH-II)* has been included here as well as in the next section, as it has a subscale for impulsive/challenging behavior (Matson, Gardner, Coe, & Sovner, 1991). It is the only checklist for challenging behavior beside the ABC found to be appropriate for individuals with severe to profound ID (Flynn et al., 2017). It is something of a paradox that so few checklists are available for evaluation of challenging behavior in severe and profound ID, as these individuals will have greater difficulties answering questions and assessments thus have to rely more heavily on checklists. The subscales of the DASH-II have been confirmed by factor analyses (Matson, Coe, Gardner, & Sovner, 1991). High scores on these subscales are associated with high scores on similar scales of the ABC (Paclawskyj, Matson, Bamburg, & Baglio, 1997), with the impulse control subscale on the DASH-II correlating highly with the irritability and hyperactivity subscale of the ABC.

The *Behavior Problems Inventory* (BPI) was first used in a prevalence study of self-injurious behavior (Rojahn, 1986). It has later been expanded and translated from German to several languages. It has also been used in epidemiological research (Rojahn et al., 2001). The BPI was developed with a more specific focus on problematic behaviors, as these typically receive relatively little attention in broader instruments such as the DASH, the ABC, or RSMB. It was designed for use in clinical assessment, treatment outcome measures, survey/epidemiological use, as well as for micro-analyses of common problem behaviors. The BPI was developed in a similar manner to the ABC, identifying subscales by use of factor analysis (Rojahn et al., 2001). Three subscales have been identified, focusing on self-injurious behavior, stereotyped behavior, and aggressive/destructive behavior (Rojahn et al., 2001). One study (Rojahn, Aman, Matson, & Mayville, 2003) found that the BPI and the ABC cross-validated each other where expected, while subscales assumed to have little relationship were not associated. Various editions have had varying number of items from 29 (Rojahn, Polster, Mulick, & Wisniewski, 1989) to 49 (Rojahn et al., 2003) and 52 (Rojahn et al., 2001) items. It currently exists in two versions, the full version often referred to as the BPI-01 (Rojahn et al., 2003), and a shorter version, the BPI-S, which was empirically shortened using archival data (Rojahn et al., 2012a, 2012b). Comparable psychometric properties have been found for the short version as to the longer one (Bowring et al., 2017).

Concluding Remarks

There are several checklists available for assessment of challenging behavior in individuals with ID. These checklists have varying strengths and weaknesses, and the choice of checklist should be made based on the characteristics of the individual to be assessed. As described, some of these checklists seem to be more appropriate for individuals with severe or profound ID, while others seem to be more appropriate with mild or moderate ID. The ABC currently seems to be the only checklist for challenging behavior with repeatedly studied and confirmed validity across the range of ID levels. However, as it is also the checklist backed by the largest number of studies, the other checklists may have similarly strong psychometric characteristics, but further research is needed to confirm this. As all these instruments have their individual strengths and weaknesses, combining two or more of them in individual assessments would be recommended – a process sometimes referred to as triangulation (Yin, 2014) – to avoid relying on a single instrument that may not always be appropriate for the individual being assessed.

Instruments on Psychiatric Disorders

This section covers the scales and related methods designed to help to arrive at a diagnosis of psychopathology in individuals with ID. Checklists and structured interviews that aim to identify mental disorders in persons with ID who do not have additional ASD will be presented. Only tools that cover main domains of mental disorders will be presented in full, while useful checklists covering one or two specific disorder(s) will be mentioned with one key reference. In mental health services, a wide range of elements are assessed in order to plan treatment and follow-up; diagnostic categories being one.

The *psychopathology inventory for mentally retarded* (PIMRA) was beside the Reiss Screen the two checklists developed for the purpose of identifying mental health problems in ID already in the 1980s. The PIMRA is developed in the USA and is widely used. It has been translated into other languages like Swedish (Gustafsson & Sonnander, 2005), Norwegian (Linaker & Helle, 1994), Dutch (Minnen, Savelsberg, & Hoogduin, 1994), and Indian. The PIMRA is derived from DSM-III and adjusted for observational use (Matson, Kazdin, & Senatore, 1984). The checklist is available in two versions, self-report and informant, and the checklist include eight subscales: seven measuring mental health problems, schizophrenia, adjustment disorder, anxiety disorder, psychosexual disorder, affective disorder, somatoform disorder, and personality disorder, and one subscale measuring inappropriate adjustment. Each scale has 7 items, all together 56 items. The scale has two versions, one self-rated, which is recommended for persons with mild ID only, and one informant completed version. The PIMRA is recommended for persons on all levels of ID, who are suspected of having mental health problems: for assessment and treatment evaluation. Strengths include having a self-rated version, including all levels of ID, and good reliability. The limitations include questionable validity (Gustafsson & Sonnander, 2005), and the items may be challenging to score without thorough knowledge about phenomenology of mental disorders in ID, which may explain the rather low sensitivity found in some studies (i.e., Gustafsson & Sonnander, 2005).

The *Reiss Screen for Maladaptive Behavior* (RSMB) was published in 1988 (Reiss, 1988). The RSMB is developed in the USA and has been widely used there, both clinically and in research (Havercamp & Reiss, 1997), and also in European countries. This RSMB was constructed for detecting psychiatric symptoms, self-harm, and other challenging behavior (Gustafsson & Sonnander, 2002; Reiss, 1988). It has seven subscales with five items each, measuring aggression, psychosis, paranoia, depression, dependent personality disorder, and avoidant personality disorder. The RSMB has a three-point evaluation scale of severity of problems. The scale is developed with 38 items in alphabetical order. Each item is presented in three parts: the symptom, a definition of the symptom, and an example. This is to facilitate the scoring for non-professionals. The RSMB is recommended for persons with mild or moderate ID. The strengths include the facilitation for non-professional – that often know the person well and therefore may score behavior change more reliable than professionals (Helverschou, Bakken, & Martinsen, 2009) and also that the RSMB differentiate well between persons with and without mental disorders (Kishore, Nizamie, & Nizamie, 2010). The limitations relate to the subscales included. The instrument does not cover anxiety disorders, which are known to be highly prevalent in persons with ID. The autism subscale is found to have weak diagnostic utility and should not be used for diagnostic or screening purposes (Bertelli, Rossi, Scuticcio, & Bianco, 2015).

The *Diagnostic Assessment for the Severely Handicapped Scale* (DASH) was published in 1988 (Matson). The DASH is complementary to the ADD (Matson & Bamburg, 1998; Matson et al., 1991). See below. The scale was developed to measure severity, duration and frequency of mental health problems in persons with severe or profound ID (ibid.). The DASH encompass 13 subscales including anxiety, depression, mania, autism, schizophrenia, stereotypies, self-injury, elimination, eating, sleep, sexual, organic, and

impulse. It has 84 items. The DASH-II version has been widely used both in the USA and in Europe (Flynn et al., 2017; Myrbakk & von Tetzchner, 2008c; Paclawskyj et al., 1997; Perez-Achiaga, Nelson, & Hassiotis, 2009). The items are scored on a three-point scale. The DASH-II is recommended for persons with severe and profound ID with suspected mental illness but may be used for all levels of ID (Myrbakk & von Tetzchner, 2008c; Paclawskyj et al., 1997). The strengths include good reliability and validity in a number of research papers (Flynn et al., 2017), and the broad spectrum of subscales, and that the DASH-II has norming data that has elicited cutoffs. DASH-II address autism as one of the disorders to be identified and may work as a screening instrument for ASD. However, there is reason to question whether psychiatric comorbidity can be identified among individuals with ASD since reliability and validity in identifying psychiatric comorbidity in individuals with autism not yet have been examined (Leyfer, Folstein, Bacalman, et al., 2006).

The *Psychiatric Assessment Schedule for Adults with a Developmental Disability* (PAS-ADD) was published during the 1990s. It is a system of assessment tools developed by Moss and colleges in the UK in order to detect mental disorder symptoms in adults with intellectual disability. The PAS-ADD encompass three tools: a semi-structured clinical interview, a checklists for scoring by professional caregivers (the MINI PAS-ADD), and a checklist for scoring by informal caregivers like family members (the PAS-ADD Checklist). This system is widely used in the European countries (Bertelli et al., 2015). More information about the PAS-ADD system is found here: https://www.pavpub.com/?s=pass+-+add&post_type=product&type_aws=true

The *PAS-ADD Clinical Interview* was published in 1993 (Moss et al.). It is a semi-structured comprehensive interview. It is developed to correspond to criteria both in ICD-10 and DSM-IV. The development of the PAS-ADD is based on the assumption that manifestations of mental illness are the same in people with ID as in the general population (Moss et al., 1997). The interview is recommended for patients with mild and moderate ID and suspected mental disorder. Both the patients and caregivers may be interviewed. It is recommended to score checklists for aberrant behavior additionally, such as the ABC (described above), for differential diagnostic purposes. The strengths of this interview is that it is comprehensive with a large number of items and that it is constructed for merging different items in a coding system that generates diagnoses within the main categories (psychosis, affective, and anxiety disorders). The subscales have cutoffs. Further, the interview is supplied by 26 observational items for patients with less verbal capacity. The limitations are mainly that the interview will not be feasible for patients with severe and profound ID and that it is quite time-consuming. See also PAS-ADD Clinical Interview handbook https://www.pavpub.com/pas-add-clinical-interview-handbook

The *Mini PAS-ADD* assessment scale is as the PAS-ADD interview developed by Steve Moss and colleges and published in 1998 (Prosser et al.). It is supposed to enable non-clinicians (basically community carers) to accurately recognize mental disorders in people with ID (ibid.). It includes seven subscales; depression, anxiety and phobias, mania, OCD, psychosis, unspecified disorder, and autism. The items are scored by care staff in order to make informed referrals to mental health services (Myrbakk & von Tetzchner, 2008c). The 86 items are scored on a four-point scale. This assessment tool is recommended for patients with mild to profound ID and suspected mental health problems. It is recommended to be scored by professional caregivers such as nurses and social pedagogues. The strengths encompass that it is comprehensive and covers the main groups of mental illness. It has cutoffs. The limitations are mainly the relatively low inter-rater reliability found in research studies and that there is a lack of data from larger-scale studies, which would have been an advantage for such a comprehensive tool.

The *PAS-ADD Checklist* is a screening tool for care staff. It has 29 items covering life events and psychiatric symptoms and is scored on a four-point scale (Moss et al., 1998). The items are worded in

everyday language. This checklist is widely used in European countries (Hatton & Taylor, 2008). The items cover seven subscales: appetite and sleep, tension and worry, phobias and panics, depression and hypomania, obsessions and compulsions, psychoses, and autism. Recommended use: The PAS-ADD checklist is recommended for patients with mild-profound ID and suspected mental health problems in community settings. It is designed to be a quick and easy-scored tool for untrained professional caregivers (Moss et al., 1998). Strengths and limitations: The strengths of this checklist are basically the feasibility in community settings (Moss et al., 1998). This may explain the widespread use. It also has cutoffs. The limitations are mainly linked to the relatively poor psychometric properties found in research studies (Hatton & Taylor, 2008; Moss et al., 1998), which include low inter-rater reliability and somewhat low internal consistency for some subscales.

The Assessment of Dual Diagnoses, ADD. The ADD is complementary to the DASH-II (Matson et al., 1991); see above. It is a screening tool, collecting third-party information (Matson & Bamburg, 1998). The ADD has 79 items in 13 subscales (like the DASH): depression, anxiety, PTSD, substance abuse, somatoform disorder, dementia, conduct disorder, pervasive developmental disorder, schizophrenia, personality disorder, eating disorder, and sexual disorder. Each item is scored for duration and frequency additionally to symptom severity on a three-point scale. Scoring the ADD should take about 20 minutes (ibid.). The ADD checklist is recommended for patients with mild to moderate ID and suspected mental health problems. Primary caregivers should be interviewed by trained interviewers (Matson & Bamburg, 1998). The strengths of this checklist are the broad variety of subscales and that it is not a time-consuming tool (Moss et al., 1998). The reliability and validity seems to be good. The limitations are mainly linked to the ADD not having cutoffs (Myrbakk & von Tetzchner, 2008c). It is not tested for self-report, as many persons with mild and moderate ID may answer questions about mental health (Douma, Dekker, Verhulst, & Koot, 2006; Rose, Willner, Shead, Johoda, et al., 2013).

The *Developmental Behaviour Checklist for Adults*, DBC-A. The DBC-A is developed in Australia by Mohr, Tonge, and Einfeld and published in 2005. It was built on the Developmental Behaviour Checklist (DBC), a widely used checklist for detecting mental health problems in children and adolescents (Hassiotis & Turk, 2011). Some items of the DBC were deleted, and new items added. The DBC-A has 107 items in 6 subscales: disruptive, communication and anxiety disturbance, self-absorbed, antisocial, depressive, and social relating. The item is scored on a three-point scale and a manual is available. The DBC-A is recommended for use by family members or professional service providers to score adults above 18 for possible mental health and behavior problems (Mohr et al., 2011). As the DBC-A have a number of items in common with the child/adolescent version, mental health issues may be monitored from childhood into adulthood by using this system. It has good reliability and validity. It may be scored by both family members and service providers in community settings. It has cutoffs. Limitations are related to the few subscales, especially not including psychosis and mania.

The *Psychopathology Checklist for Adults with Intellectual Disability*, P-AID. The P-AID is a newer tool based on the Diagnostic Criterion for psychiatric disorder for use with adults with Learning Disabilities, DC-LD (Royal College of Psychiatrists, 2001), which again is based on ICD-10. The P-AID has 280 items in 10 subscales for mental health (dementia, schizophrenia, depression, mania, agora phobia, social phobia, specific phobia, general anxiety, panic anxiety, and OCD) and 8 subscales for behavior problems (verbal, physical, destructive, SIB, sexual, oppositional, demanding, wandering). It is a professional caregiver completed checklist. It is recommended for adults with all levels of ID. It should be scored by professional caregivers, with a minimum of 3-year college university education. This tool is comprehensive and combines mental health problems and behavior problems. However, the tool is not translated to English, which limits the use to the Nordic countries, and there is no available replication study of the ini-

tial findings of Hove and Havik (2008). There are no cutoffs. Information about scoring values is not available in English (Hove & Havik, 2008). The comprehensiveness of the checklist is both strength and a limitation since it is quite time-consuming to complete.

Recommended Checklists for Individual Disorders

Because of overlapping symptoms, checklists for specific disorders may be helpful in the further assessment process. The following checklists mentioned are recommended for this purpose:

Anxiety and depression: *Anxiety, Depression and Mood Scale* (ADAMS, Esbensen, Rojahn, Aman, & Ruedrich, 2003). *Glasgow Anxiety Scale for people with an Intellectual Disability* (GAS-ID, Mindham & Espie, 2003). *Glasgow Depression Scale for people with a Learning Disability* (GAS-LD, Cuthill, Espie, & Cooper, 2003). *Mood, Interest & Pleasure Questionnaire* (MIPQ, (Ross & Oliver, 2003). *Mood and Anxiety Semi-Structured Interview* (MASS) for patients with intellectual disability (Charlot, Deutsch, Hunt, Fletcher, & McIlvane, 2007).

Eating disorders: The Screening Tool of Eating Problems (STEP, Matson, Fodstad, & Boisjoli, 2008).

Emotions: Scale of Emotional Development-Short (SED-S, Sappok et al., 2016).

Trauma: The Lancaster and Northgate Trauma Scales (LANTS, Wigham, Hatton, & Taylor, 2011). Impact of Event Scale Revised for people with Intellectual Disabilities (IES-ID, Hall, Jobson, & Langdon, 2014). Bangor Life Events Schedule for Intellectual Disabilities (BLESID, Hulbert-Williams, Hastings, Crowe, & Pemberton, 2011).

Concluding Remarks

Most of the tools presented in full are relatively comprehensive and cover main categories of mental disorders. However, some important disorders like anorexia nervosa and PTSD are not covered by the commonly used tools; additional tools and clinical observations should be conducted when needed. Another important topic is the suggested discrepancy between patient reported symptoms and family/professional caregiver reported symptoms (Douma et al., 2006; Finlay & Lyons, 2001; Moss, Prosser, Ibbotson, & Goldberg, 1996; Rose et al., 2013). Generally, patient-reported outcome measures (PROM) are during the last decade found to be more clinically relevant related to interventions than measures reported by health professionals (Mercieca-Bebber et al., 2017). A number of papers which have included informants with ID report findings in line with this new approach in clinical research. Already in 1996, Moss and colleges report that adults with mild and moderate ID reported more anxiety and depressive symptoms than professional caregivers, who reported more stress and psychotic symptoms. One study on anger and ID reported that the users with mild and moderate ID report more internal emotions and mental health problems, whereas professional caregivers reported more behavior problems (Rose et al., 2013). Most of the tools presented here use caregiver report. In sum, studies indicate that individuals with ID themselves report more mental health problems, whereas caregivers report more behavior problems. This discrepancy should influence clinicians to use third-party information with caution, as well as seek information from multiple sources.

Assessment in Individuals with ASD and ID

In most studies higher prevalence rates of psychiatric disorders are reported in individuals who have both ASD and ID compared to ID "only" (Bakken et al., 2010; Bakken, Helverschou, Høidal, & Martinsen, 2016; Helverschou et al., 2011,) and ASD are recognized as a more severe developmental risk than most other disabilities (Matson & Cervantes, 2014; Wing & Gould, 1979). The identification of psychiatric disorders

in individuals with ASD and ID is considered particularly difficult due to the considerable symptom overlap between ASD and psychiatric disorders and the problems of distinguishing between the conditions (Bakken et al., 2016; Clarke, Baxter, Perry, & Prasher, 1999; Clarke, Littlejohns, Corbett, & Joseph, 1989; Ghaziuddin, Alessi, & Greden, 1995; Ghaziuddin & Greden, 1998; Ghaziuddin, Tsai, & Ghaziuddin, 1992; Helverschou et al., 2011; Helverschou, Bakken, & Martinsen, 2008; Kobayashi & Murata, 1998; Lainhart, 1999; Long, Wood, & Holmes, 2000; McDougle, Kresch, & Posey, 2000; Reaven & Hepburn, 2003; Volkmar & Cohen, 1991; Wing, 1996). The complexity is illustrated by the large variation in reported prevalence rates of mental illness in persons with ASD, between 16% and 73% (Lai, Lombardo, & Baron-Cohen, 2014). The main explanation to the *large variation* in prevalence is probably associated with problems in the delineation between ASD and mental illness (Helverschou, 2010). The communication difficulties that characterize ASD further complicate the diagnostics. However, few instruments have been developed to aid the process of identification of psychiatric disorders in individuals with ASD (Underwood, McCarthy, & Tsakanikos, 2011). Three different diagnostic interview and screening instruments especially for use with individuals with ASD and ID will be presented below and are summarized in Table 11.3.

The Schedule for the Assessment of Psychiatric Problems Associated with Autism – SAPPA. Only one diagnostic tool is available in order to diagnose additional mental health problems in individuals with ASD, the Schedule for the Assessment of Psychiatric Problems Associated with Autism – SAPPA (Bolton & Rutter, 1994). The SAPPA is developed in the UK, and the items are derived from Research Diagnostic Criteria (Spitzer, Endicott, & Robins, 1978). The SAPPA is a semi-structured interview for use by a trained professionals with an informant and has been developed for clinical research to assess prevalence of psychiatric disorders. There are no reports on psychometric properties. The SAPPA is described to provide an assessment framework for use by clinicians experienced in the psychiatric assessment of persons with autism and with intellectual disabilities and has been used in a number of follow-up studies (Bolton et al., 2011; Bradley & Bolton, 2006; Hutton, Goode, Murphy, Couteur, & Rutter, 2008).

The authors describe the criteria to differentiate between autism and psychiatric disorders as strict. To consider a disorder as a comorbid psychiatric disorder to ASD, termed "new psychiatric disorder" by the SAPPA, there is an explicitly demand of emergence of a condition that represents more than a worsening of already existing autism features and that constitute a clear break from the preexisting autism. Among studies addressing prevalence of psychiatric comorbidity in ASD, the studies which have used the SAPPA report the lowest rates of co-occurring psychiatric disorders. For example, Hutton and colleagues (2008) report an onset of new psychiatric disorders at follow-up in only 16 percent of adults diagnosed with ASD as children. Another 5% of the individuals were identified with a possible new psychiatric disorder. However, by using such strict criteria, the researchers may fail to recognize all symptoms and overlook the very specific appearances of psychiatric disorders in individuals with ASD. The use of such strict criteria contrasts with the view of researchers who consider the appearance of new maladaptive behaviors and an increase in typical autism symptoms (i.e., more intense ruminations and repetitive and ritualistic behavior) as indicators of psychiatric disorders in individuals with autism (Ghaziuddin, 2005; Tantam, 2000; Wing, 1996). Due to confounding between anxiety and autism (Morgan, 2006; Weisbrot, Gadow, DeVincent, & Pomeroy, 2005) and the finding by Hutton and colleagues (2008) of only one person with an anxiety disorder, there is reason to suspect that anxiety is not adequately identified by these criteria.

The Autism Spectrum Disorder-Comorbidity for Adults (ASD-CA) (ASD-CA; Matson & Boisjoli, 2008) is one of two screening instruments designed for identifying psychiatric disorders in this population. It contains items based on a review of the literature, diagnostic criteria (DSM-IV-TR), and in reference to other scales that measure psychopathology in ID popu-

lations (Matson & Boisjoli, 2008). The items included are judged by the authors as characteristic of the most probable psychiatric disorders in the ASD population and are constructed to screen for comorbid psychopathology in adults with ASD and ID. The ASD-CA is an informant-based measure with items scored on a two-point scale: (0) not different, no impairment, or (1) different, some impairment. Items load into five subscales that were empirically derived through factor analysis: (1) anxiety/repetitive B, (2) conduct problems, (3) irritability/behavioral excesses, (4) attention/hyperactivity/impulsivity, and (5) depressive symptoms (Matson & Boisjoli, 2008). Subscales scores are calculated by summing item scores within each factor. The primary subscales are intended to provide a measure of psychopathology consistent with the respective labels. A total subscale score are afterward calculated by summing subscale scores. Total subscale score is intended to provide an overall measure of psychopathology. Two reports have been published on the ASD-CA. The first study evaluated reliability and factor structure of the scale (Matson & Boisjoli, 2008). Inter-rater reliability ranged from 0.30 to 0.77 with an average kappa of 0.77 for all items. Kappa values were in the range of 0.35–0.92 for test-retest reliability with an overall average kappa of 0.59. The internal consistencies of the subscales consisted of KR-20 confidents ranging from 0.44 to 0.85. The internal consistency of the entire scale was good with a KR-20 coefficient of 0.91. In a second study, comparison was performed between a group with ID and ASD and psychopathology and a group with ASD and ID only (LoVullo & Matson, 2009). The reliability and validity of each subscale of the ASD-CA varied widely, and it was unable to distinguish between the group with ASD and ID and co-occurring psychopathology the group with ASD and ID alone.

The Psychopathology of Autism Checklist – PAC (Helverschou et al., 2008, 2009). The PAC is developed based on an understanding that differentiation between ASD and mental illness is a prerequisite for developing more valid psychiatric diagnoses in this group. An initial study, a conceptual analysis based on definitions of psychiatric disorders in diagnostic manuals, i.e., ICD-10 and DSM-IV, demonstrated that a panel of clinicians were able to identify symptoms of four groups of mental illness that do not overlap with the core characteristics of ASD (Helverschou et al., 2008). These items were considered as indicators of psychiatric disorders in persons with ASD and ID and were included in the PAC. In addition, the conceptual analysis identified a set of nonspecific items regarded as representing at least three disorders and not specific to the disorder they originally were selected to measure. These general indicators of impaired functioning or mental health problems were included in the general adjustment problem (GAP) subscale of the PAC to increase the probability of identifying all individuals in need of further psychiatric assessment. Symptoms such as sleeping problems, general passivity, challenging behavior, and general distress, which are among the items in the GAP subscale, are typically associated with most psychiatric disorders in individuals with ASD and ID as well as reactions to a difficult life situation (Ghaziuddin, 2005; Lainhart, 1999; Reiss, 1988; Stavrakiki, 1999).

The PAC is a caregiver-completed screening checklist for the identification of individuals with ASD and ID in need of psychiatric services. The PAC comprises 42 items distributed across 5 subscales; *psychosis* (10 items), *depression*, (7 items), *anxiety disorders* (6 items), *obsessive-compulsive disorder* (OCD) (7 items), and *general adjustment problems* (12 items). Thirty items represent symptoms assessed as specific to one of four major psychiatric disorders and not to autism. Twelve items are assessed as indicators of general problems often observed in individuals with adjustment problems and/or psychiatric disorders, e.g., sleep disturbances, self-harm, irritability, passivity, and restlessness.

Each item is assessed on two domains: "Extent of problems," addresses the current extent of problems (1 = no problem; 2 = minor problem; 3 = moderate problem; 4 = severe problem), and "Change from usual behavior," addresses whether the behavior has changed in relation to premorbid or typical patterns of behavior (worsened, unchanged, and improved). The rating "Change

from usual behavior" is used for clinical interpretation, and "Extend of problems" is used for diagnostic evaluation. A two-step procedure secures identification of individuals with a possible psychiatric disorder. First, individuals with *severe general adjustment problems* are identified, i.e., average GAP score above cutoff. Thereafter, those individuals who obtain an average score above cutoff for any of the psychiatric subscales are classified as suspected as having a psychiatric disorder. They should be referred to a comprehensive psychiatric examination.

In the first validation study (Helverschou et al., 2009), the PAC was found to discriminate between adults with autism and ID with and without psychiatric disorders and to a certain extent between individuals with different psychiatric disorders, especially psychosis and OCD. The psychometric properties, i.e., internal consistency computed by Cronbach's α and inter-rater agreement computed by Cohen's kappa, were also acceptable (psychosis $\alpha = 0.89$ and $k = 0.51$; depression $\alpha = 0.85$ and $k = 0.67$; anxiety disorder $\alpha = 0.78$ and $k = 0.58$; OCD $\alpha = 0.88$ and $k = 0.53$; general adjustment problems $\alpha = 0.88$ and $k = 0.66$). Cutoff values for the subscales have been established based on the results of the validation study (Helverschou et al., 2009). Comparison between anxiety assessment by the PAC and clinical assessment demonstrates, however, that the anxiety items in the PAC are not sufficient to identify all individuals with anxiety problems but have to be combined with severe general adjustment problems, i.e., a GAP score above cutoff (Helverschou & Martinsen, 2011).

In a screening study of a representative sample, psychiatric disorders and severe adjustments problems were found in more than 50% of the ASD and ID group compared to approximately 20% in a representative sample of ID only (Bakken et al., 2010). Thus the PAC identifies higher rates of psychiatric disorders than studies using the strictest criteria, criteria that do not include deterioration of already existing autism features or increase in typical autism symptoms as symptoms of psychiatric disorders. This may indicate that the very specific ways psychiatric disorders are manifested in individuals with ASD and ID are identified by the PAC. These symptoms may have been overlooked clinically and in studies where autism and psychiatric disorders have been identified at the same time (i.e., Melville et al., 2008; Tsakanikos et al., 2006) as well as in the studies that demand "a clear break" from the preexisting autism (i.e., Hutton et al., 2008).

Comparison Between ASD-CA and PAC

Both the Autism Spectrum Disorder-Comorbidity for Adults (ASD-CA, Matson & Boisjoli, 2008, Lovullo & Matson, 2009) and the Psychopathology in Autism Checklist (PAC) (Helverschou et al., 2008, 2009) have been published parallel in time and have been developed with the same purpose, namely, to screen for comorbid psychopathology in adults with ASD and ID. However, the construction of the two checklists differs. Based on definitions of psychiatric disorders in diagnostic manuals, i.e., ICD-10 and DSM-IV and the conceptual analysis, the approach chosen in the development of the PAC can be described as a top-down approach, while the strategy chosen in the development of the ASD-CA can be described as a bottom-up approach (e.g., Achenbach, Dumenci, & Rescorla, 2003). The ASD-CA contains items judged by the authors as characteristic of the most probable additional disorders in the ASD population. This instrument addresses neurodevelopmental disorders such as ADHD, as well as "core" psychiatric disorders like anxiety and depression. Psychosis is, however, not explicitly included. The development of the ASD-CA draws on the authors' wide experience developing instruments of psychiatric assessment in individuals with ID only and thus is likely to include key items for identification of psychiatric disorders in adults with ASD and ID. The DASH-II (Matson et al., 1991), which is among the few instruments especially designed to assess psychiatric disorders in individuals with severe and profound ID, and which has been used in a study on psychiatric disorders in individuals with autism and ID, reported prevalence similar to

those found by the PAC (e.g., Bradley, Summers, Hayley, & Bryson, 2004). At the present time, cutoff values and psychometric properties have been reported on the ASD-CA, which the authors consider as important steps in the development of the scale, but they argue for the need for more studies to further examine the scale's usefulness (Lovullo & Matson, 2009). More studies on the properties of the PAC are also warranted, particularly on external validity and its sensitivity and specificity. Since the ASD-CA and the PAC represent different approaches but with a similar aim, they might prove useful complementary tools. Important steps in the development of these scales have been taken, but there is a need for more studies to further examine the scales usefulness. Probably, each of these much needed assessments instruments may be useful tools and is likely to contribute to the identification of psychiatric disorders in this population becoming less challenging.

Concluding Remarks

At present, there is no consensus on diagnostic practice and criteria for the use in diagnosing mental illness in persons with ASD and ID. Confounding between autism and mental illness may cause diagnostic shadowing both ways; mental illness may be overlooked when symptoms are attributed to impairments caused by autism, and autism may be overlooked by mental illness symptoms overshadowing characteristics of autism (Cholemkery, Mojica, Rohrmann, et al., 2014; Geurts & Jansen, 2011; Helverschou et al., 2011). The large variation in prevalence estimates thus indicates that mental illness is both under- and overdiagnosed in people with autism. There are two main pitfalls; the first of which would be to interpret symptoms of autism as mental illness, which may result in too many being identified with at psychiatric disorder. By using measurements not adjusted for persons with autism, such as instruments for the general population or for persons with intellectual disability (ID), ordinary autism symptoms are likely to be misinterpreted as psychiatric symptoms. This may be the case for the studies reporting the highest prevalence rates.

The second pitfall is to overlook mental illness in persons with ASD. Several of the instruments designed for screening for psychopathology in individuals with ID address autism as one of the disorders to be identified and have not yet been examined for reliability and validity in identifying psychiatric comorbidity in individuals with autism (Leyfer et al., 2006). Concurrent identification of autism and mental illness may result in psychiatric symptoms being overshadowed by autism symptoms. This interpretation is suggested by the lower rates of depression and anxiety reported (e.g., Melville et al., 2008; Tsakanikos et al., 2006). Likewise, when some studies report that persons with ID only more often suffer from psychiatric disorders compared to persons with both ASD and ID, these reports may be explained by psychiatric symptoms in the persons with ASD being overshadowed by autism symptoms. When the assessment has been done simultaneously, more psychiatric disorders in the ID group only have been reported despite higher rates of problem behavior (e.g., Melville et al., 2008) and significant higher proportion of patients receiving medication (e.g., Tsakanikos et al., 2006) in the autism group.

Thus, diagnosing mental illness in persons with autism and ID poses formidable challenges and indicates that diagnostic criteria (ICD, World Health Organization, WHO, 1993 or DSM, American Psychiatric association, APA, 2013) may not be used unmodified. The diagnostic process is time-consuming and requires skilled professionals who work in teams and contribute in different ways. Information including psychometrics, interviews with formal and informal caregivers, observations, and physiological measures is crucial (Bakken & Høidal, 2014; Helverschou et al., 2011; Kildahl, Bakken, Holm, & Helverschou, 2017; MacNeil, Lopes, & Minnes, 2009).

Use of Conventional Assessment Tools

There is currently little consensus on the use and adaptation of conventional assessment tools in the assessment of mental health in ID. Though their properties in this population have been the object of little systematic study, there are indications that such instruments may be useful in clinical settings (Hassiotis et al., 2011; Kildahl et al., 2017, 2018; Manohar et al., 2016). Most available ID-specific tools are designed as screening measures or checklists, not as diagnostic instruments. Relying on these ID-specific measures alone involves a risk that diagnosable mental health problems not belonging in the main symptom categories may be overlooked. Supplementing ID-specific measures with conventional assessment tools seems to be one promising approach to reduce this risk (Manohar et al., 2016; Kildahl et al., 2017, 2018), as do seeking information from multiple sources in these assessments (Kildahl et al., 2017).

Conventional assessment tools have generally been more thoroughly studied than most ID-specific tools, including their psychometric properties. They are also more well-known to practitioners who do not work with ID, easing communication with practitioners in general mental health care. However, application of conventional assessment tools in the ID population is not without potential problems, and there is a need for further research on how they may be adapted as well as whether and how this adaptation affects their psychometric properties.

In individuals with intellectual disability and good verbal abilities, conventional tools may be applied in a standardized manner, as long as questions are simplified and/or shortened. In individuals with more severe intellectual disability, examples of adaptions include using such tools to systemize information from several sources, including interviews with family and professional caregivers (Kildahl et al., 2018). However, findings from such non-standardized use of the instruments must be interpreted with care. Certain symptoms of mental health problems, for instance, hearing voices or experiencing flashbacks, are inherently intra-psychic phenomena and may be difficult for others to observe if they do not know what behavioral equivalents to look for (Kildahl et al., 2017; Kildahl, Bakken, Iversen, & Helverschou, 2019). Lack of report of such symptoms does in other words not exclude their presence. In cases involving ASD, there is also a risk that use of conventional assessment tools may result in symptoms of ASD being interpreted as symptoms of psychiatric disorder or vice versa (Bakken & Høidal, 2014).

Examples of general conventional assessment tools described as useful in the ID population include the MINI neuropsychiatric interview in its versions both for children (Manohar et al., 2016) and adults (Kildahl et al., 2017, 2018, 2019), the Brief Symptom Inventory (Kellett, Beail, Newman, & Hawes, 2004), and the Schedule for Affective Disorders and Schizophrenia for School-Age Children-Present and Lifetime Version (Kiddie-SADS) (Kildahl, Engebretsen, & Helverschou, 2019).

For affective disorder, the Inventory of Depression Symptoms (Kildahl et al., 2018), Beck's Depression Inventory (BDI) (Perez-Achiaga et al., 2009), Montgomery-Asberg Depression Rating Scale (MADRS) (Kildahl et al., 2017), and the Hamilton Depression Rating Scale (Charlot et al., 2007) have all been used to assess depressive symptomatology in individuals with ID. The Young Mania Rating Scale (Kildahl et al., 2018) has been reported to be useful for assessment of mania symptoms. For psychotic symptoms, use of the Positive and Negative Syndrome Scale (PANSS) (Engebretsen, Kildahl, Hoy, & Bakken, 2017; Hatton et al., 2005; Kildahl et al., 2017) and the PSYRATS (Hatton et al., 2005) has been described.

Further examples include assessment of obsessive-compulsive disorder using the Yale-Brown Obsessive-Compulsive Scale in its adult or child versions (Manohar et al., 2016; Kildahl, Engebretsen, Horndalsveen, et al., 2019). This instrument also exists in an adapted version for children with ASD (Scahill et al., 2006). Personality disorders have been assessed with the Structured Clinical Interview for the DSM-IV-TR – Axis 2 (Hassiotis et al., 2011).

Hatton et al. (2005) compared conventional assessment tools for psychosis to ID-specific measures and concluded that these held up well in the ID group for measures of positive symptoms (PANSS) and hallucinations (PSYRATS) but showed poorer psychometric properties for negative symptoms (PANSS) and delusions (PSYRATS). This underlines the importance of specific knowledge concerning each instrument's strengths and weaknesses in the ID group, and that such knowledge is needed also for individual subscales and measures within instruments.

In conclusion, the application of a combination of screening tools developed for individuals with ID and adaptations of conventional assessment tools seems to be a promising approach for the clinical assessment of mental health problems in individuals with ID. However, further investigation beyond case reports or group studies focusing on single instruments is necessary to conclude regarding the viability of this approach. Future research comparing ID-specific measures and conventional assessment tools would provide knowledge necessary for further systematic application of conventional assessment tools in this population.

Conclusion

Several checklists and structured interviews have been presented in this chapter. They have been developed to assist the process of measuring symptoms displayed by individuals with ID. The main goal is to reach to an understanding of each individual's problems and provide adequate treatment and support. The presented tools have varying strengths and weaknesses, and the choice of checklist should be based on the characteristics of the individual to be assessed. As overlapping symptoms is highly prevalent in mental disorders generally, and diagnostic overshadowing is a main challenge in assessing mental health problems in people with cognitive impairments (Reiss, Levitan, & Szyszko, 1982), there is a need for clinical judgment beside using checklists and interviews. There is also an overrepresentation of physical health problem in people with ID, making the use of checklists for mental health purposes one part of a multi modal assessment (Charlot et al., 2011). It is further recommended that assessment should be multidisciplinary to include different angels of understanding the phenomenology that each patient represents (ibid.). The more severe ID, the more behavior equivalents are used when identifying symptoms (Matson et al., 1991). As behavior equivalents are less valid than self-reported symptoms (Charlot, 2003), a triangulated approach is recommended.

The psychometric properties of the different checklists vary tremendously. More research is warranted both on existing instruments presented here, the relationship between them, and on how they function in populations who have not been examined in previous studies. Particularly, for the use in individuals with ASD and ID, there is a lack of instruments as well a lack of research on how instruments that have been developed for individuals with ID only or for the general population function. This indicates care in interpreting checklist scores in clinical judgments.

References

Achenbach, T. M., Dumenci, L., & Rescorla, L. (2003). DSM-oriented and empirically based approaches to constructing scales from the same item pools. *Journal of Clinical Child and Adolescent Psychology, 32*, 328–340.

Aman, M. G. (2012, June Update). *Annotated biography on the Aberrant Behavior Checklist (ABC)*. Unpublished manuscript, The Ohio State University, Columbus, OH.

Aman, M. G., Singh, N. N., Stewart, A. W., & Field, C. J. (1985a). The Aberrant Behavior Checklist: A behavior rating scale for the assessment of treatment effects. *American Journal of Mental Deficiency, 89*, 485–491.

Aman, M. G., Singh, N. N., Stewart, A. W., & Field, C. J. (1985b). Psychometric characteristics of the Aberrant Behavior Checklist. *American Journal of Mental Deficiency, 89*, 492–502.

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

Bakken, T. L., Helverschou, S. B., Eilertsen, D. E., Hegglund, T., Myrbakk, E., & Martinsen, H. (2010). Psychiatric disorders in adolescents and adults with autism and intellectual disability: A representa-

tive study in one county in Norway. *Research in Developmental Disabilities, 31*, 1669–1677.

Bakken, T. L., Helverschou, S. B., Høidal, S. H., & Martinsen, H. (2016). Mental illness in people with intellectual disabilities and autism spectrum disorders (Chapter 11). In C. Hemmings & N. Bouras (Eds.), *Psychiatric and behavioural disorders in intellectual and developmental disabilities* (3rd ed., pp. 119–128). Cambridge: Cambridge University Press. isbn: 9781107645943.

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), 215–222.

Bertelli, M. O., Rossi, M., Scuticcio, D., & Bianco, A. (2015). Diagnosing psychiatric disorders in people with ID: Issues and achievements. *Advances in Mental Health and Intellectual Disabilities, 9*(5), 230–242.

Bolton, P. F., Carcani-Rathwell, I., Hutton, J., Goode, S., Howlin, P., & Rutter, M. (2011). Epilepsy in autism: Features and correlates. *The British Journal of Psychiatry, 198*(4), 289–294. https://doi.org/10.1192/bjp.bp.109.076877

Bolton, P. F., & Rutter, M. (1994). *Schedule for assessment of psychiatric problems associated with autism (and other developmental disorders) (SAPPA): Informant version*. Cambridge, UK: University of Cambridge; and London: Institute of Psychiatry.

Bowring, D. L., Totsika, V., Hastings, R. P., Toogood, S., & Griffith, G. 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.

Bradley, E., & Bolton, P. (2006). Episodic psychiatric disorders in teenagers with learning disabilities with and without autism. *The British Journal of Psychiatry, 189*(4), 361–366. https://doi.org/10.1192/bpj.bp.105.018127

Bradley, E. A., Summers, J. A., Hayley, L., & Bryson, S. E. (2004). Comparing rates of psychiatric and behavior disorders in adolescents and young adults with severe intellectual disability with and without autism. *Journal of Autism and Developmental Disorders, 34*, 151–161.

Charlot, L. (2003). Mission impossible? Developing an accurate classification of psychiatric disorders for individuals with developmental disorders. *Mental Health Aspects of Developmental Disabilities, 6*(1), 26–35.

Charlot, L., 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., Deutsch, C., Hunt, A., Fletcher, K., & McIlvane, 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.

Cholemkery, H., Mojica, L., Rohrmann, S., et al. (2014). Can autism spectrum disorder and social anxiety disorders be differentiated by the social responsiveness scale in children and adolescents? *Journal of Autism and Developmental Disorders, 44*, 1168–1182.

Clarke, D., Baxter, M., Perry, D., & Prasher, V. (1999). The diagnosis of effective and psychotic disorders in adults with autism: Seven case reports. *Autism, 3*, 149–164.

Clarke, D. J., Littlejohns, C. S., Corbett, J. A., & Joseph, S. (1989). Pervasive developmental disorders and psychoses in adult life. *British Journal of Psychiatry, 155*, 692–699.

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. *British Journal of Psychiatry, 182*, 347–353.

Daniel, W. F., Passmore, C. E., & Sewell, H. M. (2003). The MMPI-168(L) and ADD is assessing psychopathology in individuals with mental retardation: Between and within instruments associations. *Research in Developmental Disabilities, 264*, 19–32.

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 American Child & Adolescent Psychiatry, 45*(10), 1224–1231.

Einfeld, S. L., & Aman, M. (1995). Issues in the taxonomy of psychopathology in mental retardation. *Journal of Autism and Developmental Disorders, 25*, 143–167.

Engebretsen, M. H., Kildahl, A. N., Hoy, I. H., & Bakken, T. L. (2017). Metyrosine treatment in a woman with chromosome 22q11.2 deletion syndrome and psychosis: A case study. *International Journal of Developmental Disabilities, 65*(2), 1–6. https://doi.org/10.1080/20473869.2017.1401257

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 Disabilities, 33*(6), 617–629.

Finlay, W. M. L., & Lyons, E. (2001). Methodological issues in interviewing and using self-report questionnaires with people with mental retardation. *Psychological Assessment, 13*(3), 319–335.

Fletcher, R., Barnhill, J., & Cooper, S.-A. (2016). *Diagnostic manual-intellectual disability 2: A textbook of diagnosis of mental disorders in persons with intellectual disability*. New York, NY: NADD Press. ISBN 9781572561342.

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.

Geurts, H. M., & Jansen, M. D. (2011). A retrospective chart study: The pathway to a diagnosis for adults referred for ASD assessment. *Autism, 16*(3), 299–305.

Ghaziuddin, M. (2005). *Mental health aspects of autism and Asperger syndrome*. London, UK: Jessica Kingsley.

Ghaziuddin, M., Alessi, N., & Greden, J. F. (1995). Life events and depression in children with pervasive developmental disorders. *Journal of Autism and Developmental Disorders, 25*, 495–502.

Ghaziuddin, M., & Greden, J. F. (1998). Depression in children with autism/pervasive developmental disorders: A case-control family history study. *Journal of Autism and Developmental Disorders, 28*, 111–115.

Ghaziuddin, M., Tsai, L., & Ghaziuddin, N. (1992). Comorbidity of autistic disorder in children and adolescents. *European Child and Adolescent Psychiatry, 1*, 209–213.

González, M. L., Dixon, D. R., Rojahn, J., Esbensen, A. J., Matson, J. L., Terlonge, C., & Smith, K. R. (2009). The behavior problems inventory: Reliability and factor validity in institutionalized adults with intellectual disabilities. *Journal of Applied Research in Intellectual Disabilities, 22*, 223–235. https://doi.org/10.1111/j.1468-3148.2008.00429.x

Graham, P., & Rutter, M. (1970). Selection of children with psychiatric disorder. In M. Rutter, J. Tizard, & K. Whitmore (Eds.), *Education, health and behavior* (pp. 148–150). London, UK: Longman.

Gustafsson, C., & Sonnander, K. (2002). Psychometric evaluation of a Swedish version of the Reiss Screen for Maladaptive Behavior. *Journal of Intellectual Disability Research, 46*, 218–229. https://doi.org/10.1046/j.1365-2788.2002.00398.x

Gustafsson, C., & Sonnander, K. (2005). A psychometric evaluation of a Swedish version of the psychopathology inventory for mentally retarded adults. *Research in Developmental Disabilities, 26*, 183–201.

Hall, J. C., Jobson, L., & Langdon, P. E. (2014). Measuring symptoms of post-traumatic stress disorder in people with intellectual disabilities: The development and psychometric properties of the Impact of Event Scale-Intellectual Disabilities (IES-ID s). *British Journal of Clinical Psychology, 53*(3), 315–332.

Hassiotis, A., Gazizova, D., Akinlonu, L., Bebbington, P., Meltzer, H., & Strydom, A. (2011). Psychiatric morbidity in prisoners with intellectual disabilities: Analysis of prison survey data for England and Wales. *British Journal of Psychiatry, 199*(2), 156–157. https://doi.org/10.1192/bjp.bp.110.088039

Hassiotis, A., & Turk, J. (2011). Mental health needs in adolescents with intellectual disabilities: Cross-sectional survey of s service sample. *Journal of Applied Research in Intellectual Disabilities, 25*, 252–261.

Hatton, C., Haddock, G., Taylor, J. L., Coldwell, J., Crossley, R., & Peckham, N. (2005). The reliability and validity of general psychotic rating scales with people with mild and moderate intellectual disabilities: An empirical investigation. *Journal of Intellectual Disability Research, 49*(7), 490–500.

Hatton, T., & Taylor, J. L. (2008). Factor structure of the PAS-ADD checklist with adults with intellectual disabilities. *Journal of Intellectual and Developmental Disabilities, 33*(4), 330–336.

Havercamp, S. M., & Reiss, S. (1997). The Reiss screen for maladaptive behavior: confirmatory factor analysis. *Behavior Research and Therapy, 35*(10), 976–971.

Helverschou, S. B. (2010). *Identification of anxiety and other psychiatric disorders in individuals with autism and intellectual disability* (Dissertation for the degree of Ph.D.). Department of Psychology, University of Oslo. https://www.duo.uio.no/handle/10852/18015

Helverschou, S. B., Bakken, T., & Martinsen, H. (2011). Psychiatric disorders in people with autism spectrum disorders: Phenomenology and recognition. (Chapter 5). In J. L. Matson & P. Sturmey (Eds.), *International handbook of autism and pervasive developmental disorders* (pp. 53–74). New York, NY: Springer.

Helverschou, S. B., Bakken, T. L., & Martinsen, H. (2008). Identifying symptoms of psychiatric disorders in people with autism and intellectual disability: An empirical conceptual analysis. *Mental Health Aspects of Developmental Disabilities, 11*, 105–115.

Helverschou, S. B., Bakken, T. L., & Martinsen, H. (2009). The Psychopathology in Autism Checklist (PAC): A pilot study. *Research in Autism Spectrum Disorders, 3*, 179–195.

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.

Holmes, N., Shah, A., & Wing, L. (1983). The disability assessment schedule: A brief screening device for use with the mentally retarded. *Psychological Medicine, 12*(4), 879–890.

Hove, O., & Havik, O. E. (2008). Psychometric properties of psychopathology checklist for Adults with Intellectual Disability (P-AID) on a community sample of adults with intellectual disability. *Research in Developmental Disabilities, 29*, 467–482.

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.

Hutton, J., Goode, S., Murphy, M., Couteur, A. L., & Rutter, M. (2008). New-onset psychiatric disorders in individuals with autism. *Autism, 12*, 373–390.

Katz, R. C., Berry, E., & Singh, N. N. (1997). Using the Aberrant Behavior Checklist to assess problem behavior in older individuals. *Clinical Gerontologist, 18*, 5–12.

Kearney, D. S., & Healy, O. (2011). Investigating the relationship between challenging behavior, co-morbid psychopathology and social skills in adults with moderate to severe intellectual disabilities in Ireland. *Research in Developmental Disabilities, 32*(5), 1556–1563.

Kellett, S., Beail, N., Newman, D. W., & Hawes, A. (2004). The factor structure of the brief symptom inventory: Intellectual disability evidence. *Clinical Psychology and Psychotherapy, 11*, 275–281.

Kildahl, A. N., Bakken, T. L., Holm, O. H., & Helverschou, S. B. (2017). Assessment of psychosis in ASD/ID: A case study. *Advances in Mental Health and Intellectual Disabilities, 11*(1), 17–23.

Kildahl, A. N., Bakken, T. L., Iversen, T. E., & Helverschou, S. B. (2019). Identification of post-traumatic stress disorder in individuals with autism spectrum disorder and intellectual disability – A systematic review. *Journal of Mental Health Research in Intellectual Disabilities, 12*(1–2), 1–25.

Kildahl, A. N., Berg, L. K., Nilssen, A. L. E., Bjørgo, K., Rødningen, O., & Helverschou, S. B. (2018). Psychiatric assessment in Phelan-McDermid syndrome (22q13 deletion syndrome). *Journal of Intellectual & Developmental Disability*. https://doi.org/10.3109/13668250.2018.1440135

Kildahl, A.N., Engebretsen, M.H,. Helverschou, S.B. (2019). Attachment disorder in autism spectrum disorder and intellectual disability. *Advances in Mental Health and Intellectual Disabilities*.

Kildahl, A.N., Engebretsen, M.H.,Horndalsveen, K., Hellerud, J., Wiik, J.Y., Aasen, G.E., Helverschou, S. B. (2019). Psychiatric assessment in congenital blindness, ASD and ID: experience from two clinical cases. *Advances in Mental Health and Intellectual Disabilities, 13*(5) 194–203.

Kishore, M. T., Nizamie, S. H., & Nizamie, A. (2010). Utility of the Reiss screen in identifying psychiatric problems in persons with mental retardation. *Indian Journal of Psychological Medicine, 32*(1), 38–41.

Kobayashi, R., & Murata, T. (1998). Behavioral characteristics of 187 young adults with autism. *Psychiatry and Clinical Neuroscience, 52*, 383–390.

Lai, M.-C., Lombardo, M. V., & Baron-Cohen. (2014). Autism. *Lancet, 383*, 896–910.

Lainhart, J. E. (1999). Psychiatric problems in individuals with autism, their parents and siblings. *International Review of Psychiatry, 11*, 278–298.

Leyfer, O. T., Folstein, S. E., Bacalman, S., … Lainhart, J. E. (2006). Comorbid psychiatric disorders in children with autism: Interview development and rates of disorders. *Journal of Autism and Developmental Disorders, 36*, 849–861.

Linaker, O., & Helle, J. (1994). Validity of the Schizophrenia diagnosis of the psychopathology inventory for mentally retarded (PIMRA): A comparison of schizophrenic patients with and without mental retardation. *Research in Developmental Disabilities, 15*, 473–486.

Long, K., Wood, H., & Holmes, N. (2000). Presentation, assessment and treatment of depression in a young woman with learning disability and autism. *British Journal of Learning Disabilities, 28*, 102–108.

LoVullo, S. V., & Matson, J. L. (2009). Comorbid psychopathology in adults with autism spectrum disorders and intellectual disability. *Research in Developmental Disabilities, 30*, 1288–1296.

Manohar, H., Subramanian, K., Kandasamy, P., Penchilaiya, V. & Arun, A. (2016). Diagnostic masking and overshadowing in intellectual disability—how structured evaluation helps. *Journal of Child and Adolescent Psychiatric Nursing, 29*, 171–176. doi.org/10.1111/jcap.12160.

MacNeil, B. M., Lopes, V. A., & Minnes, P. M. (2009). Anxiety in children and adolescents with autism spectrum disorders. *Research in Autism Spectrum Disorders, 3*, 1–21.

Matson, J. L. (1988). *The PIMRA manual*. Orlando Park, IL: International Diagnostic Systems, Inc..

Matson, J. L., & Bamburg, J. W. (1998). Reliability of the Assessment of Dual Diagnosis (ADD). *Research in Developmental Disabilities, 19*(1), 89–95.

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.

Matson, J. L., & Boisjoli, J. A. (2008). Autism spectrum disorders in adults with intellectual disability and comorbid psychopathology: Scale development and reliability of the ASD-CA. *Research in Autism Spectrum Disorders, 2*, 276–287.

Matson, J. L., & Cervantes, P. E. (2014). Commonly studied comorbid psychopathologies among persons with autism spectrum disorder. *Research in Developmental Disabilities, 35*(5), 952–962. https://doi.org/10.1016/j.ridd.2014.02.012

Matson, J. L., Coe, D. A., Gardner, W. I., & Sovner, R. (1991). A factor analytic study of the diagnostic assessment for the severely handicapped scale. *Journal of Nervous and Mental Disease, 179*(9), 553–557.

Matson, J. L., Fodstad, J. C., & Boisjoli, J. A. (2008). Cutoff scores, norms and patterns of feeding problems for the The Screening Tool of Eating Problems (STEP) for adults with intellectual disabilities. *Research in Developmental Disabilities, 29*, 363–372.

Matson, J. L., Gardner, W. I., 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. *The British Journal of Psychiatry, 159*(3), 404–409.

Matson, J. L., Kazdin, A. E., & Senatore, V. (1984). Psychometric properties of the psychopathology inventory for mentally retarded. *Applied Research in Mental Retardation, 5*, 81–89.

McCarthy, 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.

McDougle, C. J., Kresch, L. E., & Posey, D. J. (2000). Repetitive thoughts and behavior in pervasive developmental disorders: Treatment with serotonin reuptake inhibitors. *Journal of Autism and Developmental Disorders, 30*, 427–435.

Melville, C. A., Cooper, S. A., Morrison, J., Smiley, J., Allan, L., Jackson, A., et al. (2008). The prevalence and incidence of mental ill-health in adults with

autism and intellectual disability. *Journal of Autism and Developmental Disorders, 38*, 1676–1688.

Mercieca-Bebber, R., Firedlander, M., Calvert, M., Stockler, M., Kyte, D., Kok, P. S., & King, M. T. (2017). A systematic evaluation of compliance and reporting of patient-reported outcome endpoints in ovarian cancer randomised controlled trials: Implications for generalisability and clinical practice. *Journal of Patient-reported Outcomes, 1*, 5. https://jpro.springeropen.com/articles/10.1186/s41687-017-0008-3

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.

Minnen, A., Savelsberg, P. M., & Hoogduin, K. A. (1994). A Dutch version of the Psychopathology Inventory for Mentally Retarded Adults (PIMRA). *Research in Developmental Disabilities, 15*, 269–278.

Mohr, C., Tonge, B. J., & Einfeld, S. L. (2005). The development of a new measure for assessment of psychopathology in adults with intellectual disability. *Journal of Intellectual Disability Research, 49*(7), 469–480.

Mohr, C., Tonge, B. J., Taffe, J., Rymill, A., Collins, D., Keating, C., & Einfeld, S. L. (2011). Inter-rater reliability of the Developmental Behaviour Checklist for adults in community accommodation settings. *Journal of Intellectual Disability Research, 55*(7), 710–713.

Morgan, K. (2006). Is autism a stress disorder? What studies of nonautistic populations can tell us. In M. G. Baron, J. Groden, G. Groden, & L. P. Lipsitt (Eds.), *Stress and coping in autism* (pp. 129–182). Oxford, UK: Oxford University Press.

Moss, S., Ibbotson, B., Prosser, H., Goldberg, D., Patel, P., & Simpson, N. (1997). Validity of the PAS-ADD for detecting psychiatric symptoms in adults with learning disability (mental retardation). *Social Psychiatry Epidemiology, 32*(6), 344–354.

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 disabilities (mental retardation). Part I: Development and reliability of the patient interview (PAS-ADD). *British Journal of Psychiatry, 163*, 471–480.

Moss, S., Prosser, H., Costello, H., Simpson, N., Patel, P., Rowe, S., … Hatton, C. (1998). 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.

Moss, S., Prosser, H., Ibbotson, B., & Goldberg, D. (1996). Respondent and informant accounts of psychiatric symptoms in a sample of patients with learning disability. *Journal of Intellectual Disability Research, 40*(5), 457–465.

Myrbakk, E., & von Tetzchner, S. (2008a). Psychiatric disorders and behavior problems in people with intellectual disability. *Research in Developmental Disabilities, 29*, 316–332.

Myrbakk, E., & von Tetzchner, S. (2008b). The prevalence of behavior problems among people with intellectual disability living in community settings. *Journal of Mental Health Research in Intellectual Disabilities, 1*(3), 205–222.

Myrbakk, E., & von Tetzchner, S. (2008c). Screening individuals with intellectual disability for psychiatric disorders: Comparison of four measures. *American Journal of Mental Retardation, 113*(1), 54–70.

Newton, J. T., & Sturmey, P. (1988). The Aberrant Behaviour Checklist: A British replication and extension of its psychometric properties. *Journal of Mental Deficiency Research, 32*, 87–92.

Ono, Y. (1996). Factor validity and reliability for the Aberrant Behavior Checklist- Community in a Japanese population with mental retardation. *Research in Developmental Disabilities, 17*, 303–309.

Othmer, E., Othmner, S. C., & Othmner, J. P. (2005). Psychiatric interview, history, and mental examination. In B. J. Kaplan & V. A. Sadock (Eds.), *Comprehensive textbook of psychiatry* (8th ed., pp. 794–827). Philadelphia, PA: Lippicott, Williams & Wilkins.

Paclawskyj, T. R., Matson, J. L., Bamburg, J. W., & Baglio, C. S. (1997). A comparison of the Diagnostic Assessment for the severely Handicapped-II (DASH-II) and the Aberrant behavior Checklist (ABC). *Research in Developmental Disabilities, 18*(4), 289–298.

Perez-Achiaga, N., Nelson, S., & Hassiotis, A. (2009). Instruments for the detection of depressive symptoms in people with intellectual disabilities. *Journal of Intellectual Disabilities, 13*(1), 55–76.

Prosser, H., Moss, S., Costello, H., Simpson, N., Patel, P., & Rowe, S. (1998). 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.

Prout, H. T. (1993). Assessing psychopathology in persons with mental retardation: A review of the Reiss scales. *Journal of School Psychology, 31*(4), 535–540.

Reaven, J., & Hepburn, S. (2003). Cognitive-behavioral treatment of obsessive-compulsive disorder in a child with Asperger syndrome. *Autism, 7*, 145–164.

Reiss, S. (1988). *Reiss screen for maladaptive behavior: Test manual*. Worthington, OH: IDS Publishing Corporation.

Reiss, S., Levitan, G., & Szyszko, J. (1982). Emotional disturbance and mental retardation: Diagnostic overshadowing. *American Journal of Mental Deficiency, 86*, 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.

Rojahn, J. (1986). Self-injurious and stereotypic behavior of noninstitutionalized mentally retarded people: Prevalence and classification. *American Journal of Mental Deficiency, 91*(3), 268–276.

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., & Helsel, W. J. (1991). The Aberrant Behavior Checklist with children and adolescents with dual diagnosis. *Journal of Autism and Developmental Disorders, 21*, 17–28.

Rojahn, J., Matson, J. L., Lott, D., Esbensen, A., & 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., Matson, J. L., Naglieri, J. A., & Mayville, E. (2004). Relationships between psychiatric conditions and behavior problems among adults with mental retardation. *American Journal on Mental Retardation, 109*(1), 21–33.

Rojahn, J., Polster, L. M., Mulick, J. A., & Wisniewski, J. J. (1989). Reliability of the Behavior Problems Inventory. *Journal of the Multihandicapped Person, 2*(4), 283–293.

Rojahn, J., Rowe, E. W., Sharber, A. C., Hastings, R., Matson, J. L., Didden, R., … Dumont, E. L. (2012a). The Behavior Problems Inventory-Short Form for individuals with intellectual disabilities: Part I: Development and provisional clinical reference data. *Journal of Intellectual Disability Research, 56*, 527–545. https://doi.org/10.1111/j.1365-2788.2011.01507.x

Rojahn, J., Rowe, E. W., Sharber, A. C., Hastings, R., Matson, J. L., Didden, R., … Dumont, E. L. (2012b). The Behavior Problems Inventory-Short Form for individuals with intellectual disabilities: Part II: Reliability and validity. *Journal of Intellectual Disability Research, 56*, 546–565. https://doi.org/10.1111/j.1365-2788.2011.01506.x

Rose, J., Willner, P., Shead, J., Johoda, A., et al. (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*, 410–419.

Ross, E., & Oliver, C. (2003). Preliminary analysis of the psychometric properties of Mood, Interest & Pleasure Questionnaire (MIPQ) for adults with severe and profound learning disabilities. *British Journal of Clinical Psychology, 42*(1), 81–93.

Royal College of Psychiatrists. (2001). *DC-LD: Diagnostic criteria for psychiatric disorders for use with adults with learning disabilities/mental retardation*. London, UK: Royal College of Psychiatrists.

Sappok,T., Barrett, B.F., Vandevelde, S., Heinrich, M., Poppe, L., Sterkenburg, L., Vonk, J., Kolb, J., Claes, C. Bergman, T., Dosen, A. & Morisse, F. (2016). Scale of emotional development—Short. *Research in Developmental Disabilities, 59*, 166–175. https://doi.org/10.1016/j.ridd.2016.08.019 Get rights and content.

Scahill, L., McDougle, C. J., Williams, S. K., Dimitropoulos, A., Aman, M. G., McCracken, J. T., et al. (2006). Children's Yale-Brown Obsessive Compulsive Scale modified for pervasive developmental disorders. *Journal of American Academy of Child and Adolescent Psychiatry, 45*, 1114–1123.

Spitzer, R. L., Endicott, J., & Robins, E. (1978). Research diagnostic criteria: Rationale and reliability. *Archives of General Psychiatry, 35*(6), 773–782.

Stavrakiki, C. (1999). Depression, anxiety and adjustment disorders in people with developmental disabilities. In N. Bouras (Ed.), *Psychiatric disorders in developmental disabilities and mental retardation* (pp. 175–187). Cambridge, UK: Cambridge University Press.

Sturmey, P., & Bertman, L. J. (1994). Validity of the Reiss screen for Maladaptive Behavior. *American Journal on Mental Retardation, 99*, 201–206.

Sturmey, P., Burcham, K. J., & Perkins, T. S. (1995). The Reiss screen for maladaptive behaviour: Its reliability and internal consistencies. *Journal of Intellectual Disability Research, 39*(3), 191–195.

Sturmey, P., Jamieson, J., Burcham, J., Shaw, B., & Bertman, L. (1996). The factor structure of the Reiss screen for maladaptive behaviors in institutional and community populations. *Research in Developmental Disabilities, 17*(4), 285–291.

Sturmey, P., & Ley, T. (1990). The psychopathology instrument for mentally retarded adults. Internal consistencies and relationship to behaviour problems. *British Journal of Psychiatry, 156*, 428–430.

Tantam, D. (2000). Psychological disorder in adolescents and adults with Asperger syndrome. *Autism, 4*, 47–62.

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*, 1123–1129.

Tsakanikos, E., Underwood, K., Sturmey, P., Bouras, N., & McCarthy, J. (2011). Psychometric properties of the Disability Assessment Schedule (DAS) for behavior problems: An independent investigation. *Research in Developmental Disabilities, 32*(2), 653–658.

Underwood, L., McCarthy, J., Chaplin, E., & Bertelli, M. O. (2015). Assessment and diagnosis of psychiatric disorder in adults with autism spectrum disorder. *Advances in Mental Health and Intellectual Disabilities, 9*(5), 222–229. https://doi.org/10.1108/AMHID-05-2015-0025

Underwood, L., McCarthy, J., & Tsakanikos, E. (2011). Assessment of comorbid psychopathology. (Chapter 17). In J. L. Matson & P. Sturmey (Eds.), *International handbook of autism and pervasive developmental disorders* (pp. 287–293). New York, NY: Springer.

Volkmar, F. R., & Cohen, D. J. (1991). Comorbid association of autism and schizophrenia. *American Journal of Psychiatry, 148*, 1705–1707.

Walsh, K. K., & Shenouda, N. (1999). Correlations among the Reiss screen, the Adaptive Behavior Scale Part II, and the Aberrant Behavior Checklist. *American Journal of Mental Retardation, 104*, 236–248.

Weisbrot, D. M., Gadow, K. D., DeVincent, C. J., & Pomeroy, J. (2005). The presentation of anxiety in

children with pervasive developmental disorders. *Journal of Child and Adolescent Psychopharmacology, 15*, 477–496.

Wigham, S., Hatton, C., & Taylor, J. L. (2011). The Lancaster and Northgate Trauma Scales (LANTS): The development and psychometric properties of a measure of trauma for people with mild to moderate intellectual disabilities. *Research in Developmental Disabilities, 32*, 2651–2659.

Wing, L. (1996). *The autistic spectrum: A guide for parents and professionals*. London, UK: Constable.

Wing, L., & Gould, J. (1979). Severe impairment of social interaction and associated abnormalities in children: Epidemiology and classification. *Journal of Autism and Developmental Disorders, 9*, 11–29.

World Health Organization. (1992). *The ICD-10 classification of mental and behavioral disorders. Clinical descriptions and diagnostic guidelines*. Geneva, Switzerland: Author.

World Health Organization. (1993). *The ICD-10 classification of mental and behavior disorders. Diagnostic criteria for research*. Geneva, Switzerland: Author.

Yin, R. K. (2014). *Case study research. design and methods* (5th ed.). Los Angeles, CA: Sage Publishing.

12 Assessment of Intellectual Disabilities: Considerations for Dual Diagnosis

Sabrina N. Grondhuis

Brief History of Intellectual Disability

Extremes of intelligence, both genius and intellectual disability, appear to be a natural part of human diversity (Amundson, 2000). Variations in cognitive functioning and examinations to measure such differences are documented throughout history and across countries. Englishman Anthony Fitzherbert is an early example, developing a test around 1535 comprised of simple questions and tasks (knowing one's age, counting 20 pence, etc.) to determine intellectual ability. That same century Juan Huarte de San Juan, a Spaniard, advocated for formal intelligence evaluations as a way to better understand physiology. German philosopher Christian Thomasius desired to use quantitative data to understand the mind at the end of the seventeenth century. Jean Marc Gaspard Itard, a French physician, gained renown when he documented the intellectual disparities possessed by a feral child he was attempting to educate in 1802 (Sattler, 2001).

The fascination with cognition increased substantially after Alfred Binet and Theodore Simon developed the first intelligence test (Binet & Simon, 1916). This assessment focused on whether an individual's mental age was comparable to their chronological age, as they were charged with identifying French children in need of special education. Soon after, Henry Goddard translated their instrument into English and endorsed its use for schools, the military, and in courts of law in the United States. Although Goddard was a proponent of eugenics, which is undoubtedly a black mark on the history of human thought, he was also a supporter of special education services and the limitation of criminal prosecution for persons with intellectual disabilities (Goddard, 1920).

The persistent reoccurrence of reduced cognitive ability is, today, commonly referred to as intellectual disability, although this has not always been the case. This recent shift in terminology from precursor *mental retardation* came from a desire to better operationalize the construct, ensure consistency across healthcare providers and their support services, and replace offensive vocabulary with a less blatantly stigmatizing term (Schalock et al., 2007). More colorful bygone descriptors include *mental handicap*, *mental subnormality*, *mental deficiency*, *idiocy*, *moron, fool, imbecile*, *lunatic*, and *feebleminded-ness* (Bach, 2007; Schalock et al., 2007). While intellectual disability is the current preferred term for the United States, other countries may use different terminology; for instance, Canada has used *developmental disability*, the Dutch term used in the Netherlands translates to *intellectual restriction*, and the United Kingdom has

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been known to use *learning disability* or *intellectual development disorder* as synonyms for the same condition (Bradley et al., 2007).

The DSM-5 (*Diagnostic and Statistical Manual of Mental Disorders*, *5th edition*; American Psychiatric Association (APA), 2013) asserts that the following three criteria must be satisfied to qualify for an intellectual disability diagnosis: (1) deficits in intellectual functioning based on standardized intelligence assessments and clinical evaluation, (2) deficits in adaptive behavior that impair functioning across multiple domains, and (3) onset of these deficits during development. Further specificity should be provided through the use of severity descriptions, including mild, moderate, severe, and profound. This definition is mirrored by the American Association on Intellectual and Developmental Disabilities (AAIDD), although they emphasize development as originating prior to 18 years of age (Schalock et al., 2010). The World Health Organization (WHO) includes a comparable definition in their International Classification of Diseases, 11th Revision (ICD-11; WHO, 2018), although under the name *disorder of intellectual development*.

Prevalence Rates of Intellectual Disability

Intellectual disability prevalence rates vary based on many factors. One meta-analysis estimated the overall prevalence rate of intellectual disabilities to be 10.37/1000 population but found that the prevalence rate changed drastically depending on the studies' sample characteristics (Maulik, Mascarenhas, Mathers, Dua, & Saxena, 2011). For instance, rates of intellectual disability prevalence were greater in countries classified as low-income (16.41/1000) or middle-income (15.92/1000) than they were for high-income countries (9.21/1000). Prevalence rates were highest in studies conducted in urban slums or mixed rural-urban settings (21.23/1000), as opposed to studies based on national data (6.23/1000). Likewise, prevalence rates were highest in studies that focused on children and adolescents (18.30/1000), as compared to those that covered the entire lifespan (5.04/1000).

While prevalence rates may differ across domains, at least one longitudinal evaluation demonstrated that annual prevalence rates of intellectual disability were relatively consistent across a 15–20 year period of time in the same geographical area. Van Naarden Braun and colleagues (2015) used surveillance data to determine that intellectual disability prevalence rates in 8-year-olds in the greater Atlanta area ranged from 10.1 to 15.5/1000 between 1991 and 2010; the average prevalence rate was 13.0/1000, and yearly estimates were consistently 11 to 13/1000 approximately, with the exception of two anomalous years (15.5 per 1000 in 1996 and 14.9 per 1000 in 2008). Interestingly, there was a decrease in rates of intellectual disability among non-Hispanic black females during the two decades investigated, but a significant *increase* among non-Hispanic white males during the latter half of the study from 2000 to 2010. This shift in gender distribution could be due to the very substantial increase in autism spectrum disorder diagnoses at the same time, as that condition is four times more prevalent in males than in females (Fombonne, 2003).

Most individuals diagnosed with an intellectual disability have the mild variant of the condition (IQ ranging from 50 to 70, in addition to meeting the other previously mentioned criteria including deficits in adaptive behavior). This is a statistical certainty given the standardized approach for interpreting intelligence assessments, as 2.23% of the population should have an IQ between 50 and 70 given the theorized normal distribution on which intelligence tracks, with a reduction in prevalence expected with each categorical severity increase. King, Toth, Hodapp, and Dykens (2009) estimated that of those diagnosed with an intellectual disability, 85% are classified as mild, 10% are deemed moderate, 4% are labeled severe, and 2% have profound intellectual disability. This distribution, with the greatest proportion being classified as mild, has also been seen in practice; Van Naarden Braun and colleagues (2015) reported that 63.7% of their sample with intellectual disability were

classified as mild and 60.1% of persons with intellectual disabilities included in Obi and colleague's evaluation were diagnoses as mild, for example.

The individuals who fall into the range of IQ scores that we characterize as moderate intellectual disability or lower are even more restricted. This creates additional challenges for measurement, as cognitive assessments are usually unable to develop adequate standardized scores that can accurately assess the lowest-performing segment of the population. This floor effect, where 40 is typically the lowest standardized score available, may be attributed to lack of representation in the norming sample and thus a reduction in instrument accuracy (Farmer, Golden, & Thurm, 2016). These standardization floors likely mean that at least some low-performing individuals will have their cognitive abilities inadvertently overestimated due to the conversion of raw scores to scaled scores provided in assessment manuals (Whitaker, 2010). Inaccuracies in cognitive measurement at the lowest end of the intelligence range could substantially impact intervention recommendations and limit availability of services to those who potentially need it most.

Tests for cognitive functioning are a vital component of psychological assessments, as these measures tend to be predictive of long-term outcomes. The presence of an intellectual disability alone or in combination with other disabilities, as is the case with dual diagnosis, can alter recommendations for therapeutic and education interventions and can necessitate adjustments to future treatment plans. An intellectual disability diagnosis will need to be monitored and reassessed throughout an individual's lifespan, as fluctuations can occur based on environmental, developmental, and educational changes. Including a cognitive assessment at the initial diagnostic appointment provides a baseline measure to which other timepoints can be compared. This provides insight into whether adjustments are necessary for existing treatments or if other factors, such as physical or other mental health concerns, are adversely impacting intellectual abilities.

Measurement of Intellectual Disability

The DSM-5 (APA, 2013) specified that standardized intelligence tests must be used in the diagnostic process for intellectual disabilities. There are many such assessments available, most of which are empirically constructed and are regularly updated to ensure appropriate norms. The typical structure of IQ tests involves the administration of incrementally more difficult questions, usually arranged by topical subsection. Individual test responses vary, but answers are ordinarily given either verbally or through the completion of a task. While subsections may focus on a particular aspect of intelligence (e.g., memory, vocabulary), composite scores combine these elements and are thought to represent a person's general cognitive ability. It is important to note that there is not a single "best" intelligence assessment; instead, individual characteristics, such as age and motor ability, must be taken into account to find a measure that is a good fit for the particular person. For instance, an individual's limited manual dexterity might make a psychometrician or clinician choose a cognitive assessment in which stimuli do not need to be manipulated. Analogously, deficits in communication might indicate that a nonverbal assessment would be the most appropriate choice.

Each intelligence test produces raw scores that are transformed into standardized scores or composites using an established distribution. The most common distribution has a mean of 100 and a standard deviation of 15, although this is not universal (e.g., the Stanford-Binet Intelligence Scale, 4th edition used a standard deviation of 16; Thorndike, Hagen, & Sattler, 1986). Community and clinical samples are tested using these assessments, from which age-related norms are derived. Every person evaluated using these measures is then placed within the existing context of what has been deemed "normal" for their age, and statistical deviations inform potential intellectual aberrations. Highlighted below are selected conventional intelligence tests (that include verbal components) which are frequently administered to persons with intellectual disabilities. The

options, listed alphabetically, include assessments that are appropriate for different age groups across the lifespan.

Bayley Scales of Infant and Toddler Development, Third Edition

The Bayley Scales of Infant and Toddler Development, Third Edition (Bayley-III; Bayley, 2006) assess development from 1 month to 42 months of age. Evaluating during this critical period of early development allows clinicians to track developmental milestones and potentially identify early delays. The Bayley-III has five scales, including adaptive behavior, cognitive, language, motor, and social-emotional. The play-based administration takes 50–90 minutes, with younger children requiring less time to be assessed. Results for the individual scales can be standardized to composite scores (M = 100, SD = 15). Split-half reliability is very good, and ranged from 0.91 (cognitive scale) to 0.93 (language scale). When using all ages, average stability coefficients were 0.80 or higher.

Differential Ability Scales-II

The Differential Ability Scales-II (DAS-II; Elliott, 2007) is an intelligence assessment for children ages 2 years, 6 months through 17 years, 11 months. The test is divided into two batteries: Early Years for children 2 years, 6 months to 6 years, 11 months and School-Age for children 7 years, 0 months to 17 years, 11 months. The Early Years Battery is further divided into Lower Level for the youngest children (2 years, 6 months through 3 years, 5 months) and Upper Level for those 3 years, 6 months through 6 years, 11 months. Each Battery and Level has slight differences in the core subtests used to derive General Conceptual Ability (GCA), which is the DAS-II's measure of global intellectual ability, similar to full scale IQ scores. GCA for the Lower Level is comprised of Verbal Ability (subtests: Verbal Comprehension, Naming Vocabulary) and Nonverbal Ability (subtests: Picture Similarities, Pattern Construction) clusters. GCA for the Upper Level includes the same Verbal Ability components, but different pieces for Nonverbal Reasoning Ability (subtests: Picture Similarities, Matrices) and a new Spatial Ability cluster (subtests: Pattern Construction, Copying). The School Age Battery GCA includes Verbal Ability (subtests: Word Definitions, Verbal Similarities), Nonverbal Reasoning Ability (subtests: Matrices, Sequential and Quantitative Reasoning), and Spatial Ability (subtests: Recall of Designs, Pattern Construction) clusters.

There are also three diagnostic clusters available for supplemental information. These clusters are Working Memory (subtests: Recall of Sequential Order, Recall of Digits Backwards), Processing Speed (subtests: Speed of Information Processing, Rapid Naming), and School Readiness (subtests: Early Number Concepts, Matching Letter-Like Forms, Phonological Processing) for those children whose ages are within the Early Years Battery – Upper Level. Children who are older and qualify for the School-Age Battery complete only the Working Memory and Processing Speed clusters (same subtests). Diagnostic clusters are not able to evaluate children who are in the age range for the Early Years Battery – Lower Level. Standard scores for core ability and diagnostic clusters range from 30 to 170, and internal reliability scores for core subtests ranged from 0.79 to 0.94 for the Early Years Battery and 0.74–0.96 for the School-Age Battery.

Kaufman Brief Intelligence Test-Second Edition

The Kaufman Brief Intelligence Test-Second Edition (KBIT-2; Kaufman & Kaufman, 2004) is a short assessment for persons aged 4 years, 0 months through 90 years, 11 months that measures verbal and nonverbal intelligence. It is comprised of three subtests, Verbal Knowledge, Matrices, and Riddles, and can be completed in under 20 minutes. Standardized scores range from 40 to 160 (M = 100, SD = 15). This assessment has good psychometric properties, as split-half

reliability for the composite IQ scores was 0.93, and it was highly correlated at 0.77 with the full scale IQ from the Wechsler Intelligence Scales for Children (fourth edition; Wechsler, 2003). The Kaufman Assessment Battery for Children, Second Edition Normative Update (Kaufman & Kaufman, 2018) is also available, but with a restricted age range from 3 to 18 years.

Stanford-Binet Intelligence Scales, 5th Edition

The Stanford-Binet Intelligence Scales, 5th edition, is an individually administered IQ test that provides standardized norms for persons aged 2–85 years (Roid, 2003). This measure includes five subtests (Fluid Reasoning, Knowledge, Quantitative Reasoning, Visual Spatial Processing, and Working Memory) which are each administered twice, once requiring verbal answers and once not requiring verbal answers. The combination of these scales produces a full scale IQ, nonverbal IQ, and verbal IQ, which provide clinicians an opportunity to see if a disparity exists between verbal and nonverbal performance on the same subscale. Should time be limited or the person under assessment is noncompliant, an abbreviated IQ that requires fewer subscales could be chosen instead. Standard scores range from 40 to 160 points, and the scale had excellent split-half reliability: 0.98 on full scale IQ, 0.95 on nonverbal IQ, 0.96 on verbal IQ, and 0.91 on abbreviated IQ.

Wechsler Tests

David Wechsler's first cognitive assessment was the Wechsler-Bellevue Intelligence Scale (Wechsler, 1939), and today, his family of intelligence tests includes standardized options for people between the ages of 30 months (Wechsler, 2012) and 90 years (Wechsler, 2008a). The Wechsler instruments are the most widely used cognitive assessments for persons with intellectual disabilities (Whitaker, 2010), and the individual instruments are discussed more fully below.

Wechsler Preschool and Primary Scale of Intelligence-Fourth Edition

The newest version of the Wechsler Preschool and Primary Scale of Intelligence-Fourth Edition (WPPSI-IV; Wechsler, 2012) divided the protocol into two age bands (2 years, 6 months to 3 years, 11 months and 4 years, 0 months to 7 years, 7 months) each of which includes different subtests to make up their respective batteries. These two paths allow the subtests to more intentionally assess intelligence in accordance with cognitive development. Composite scores range from 40 to 160. The WPPSI-IV Full Scale IQ demonstrated high internal consistency (coefficients were 0.95–0.96), high short-term test-retest stability (0.88 for all ages), and high inter-rater agreement (0.98–0.99).

Full Scale IQ for the 2 years, 6 months through 3 years, 11 month age band consists of the Verbal Comprehension Index (subtests: Receptive Vocabulary, Information, plus supplemental Picture Naming), Visual Spatial Index (Block Design, Object Assembly), and Working Memory Index (Picture Memory, Zoo Locations). Ancillary Index scores for General Ability, Nonverbal, and Vocabulary Acquisition are available for this age band in addition to the primary indices already included in Full Scale IQ.

Full Scale IQ for the 4 year, 0 month through 7 year, 7 month age band is more expansive. It consists of the Verbal Comprehension Index (subtests: Information, Similarities, with supplemental Vocabulary, Comprehension), Visual Spatial Index (Block Design, Object Assembly), Fluid Reasoning Index (Matrix Reasoning, Picture Concepts), Working Memory Index (Picture Memory, Zoo Locations), and Processing Speed Index (Bug Search, Cancellation, with supplemental Animal Coding). Ancillary Index scores for this age band include Cognitive Proficiency, General Ability, Nonverbal, and Vocabulary Acquisition.

Wechsler Intelligence Scales for Children-Fifth Edition

The Wechsler Intelligence Scales for Children-Fifth Edition (WISC-V; Wechsler, 2014a) is suitable for individuals aged 6 years, 0 months to

16 years, 11 months of age (Wechsler, 2014b). This assessment includes the following five factors: Verbal Comprehension Index (VCI; primary subtests: Similarities, Vocabulary), Visual Spatial Index (VSI; Block Design, Visual Puzzles), Fluid Reasoning Index (FRI; Matrix Reasoning, Figure Weights), Working Memory Index (WMI; Digit Span, Picture Span), and Processing Speed Index (PSI; Coding, Symbol Search). These five factors can be combined to produce a Full Scale IQ, although the subtests Visual Puzzles, Picture Span, and Symbol Search are not included in that calculation. These composite scores can range from 40 to 160.

Five ancillary indices are available when deemed necessary by clinicians. These include the Cognitive Proficiency Index, General Ability Index, Nonverbal Index, Auditory Working Memory Index, and Quantitative Reasoning Index. The first three ancillary options can be derived using the ten primary subscales listed above, but the final two require the addition of one secondary subtest to the original five areas. These secondary subscales include Information or Comprehension (for VCI), Picture Concepts or Arithmetic (for FRI), Letter-Number Sequencing (for WMI), and Cancellation (for PSI). Internal consistency estimates for the composite scores ranged from 0.88 for PSI to 0.96 for Full Scale IQ. Short-term test-retest stability estimates were also high for Full Scale IQ (0.91).

Wechsler Adult Intelligence Scales-Fourth Edition (WAIS-IV)

The Wechsler Adult Intelligence Scales-Fourth Edition (WAIS-IV) is normed for ages 16 years, 0 months to 90 years, 11 months (Wechsler, 2008b). Full Scale IQ consists of the Verbal Comprehension Index (subtests: Similarities, Vocabulary, Information), Perceptual Reasoning Index (Block Design, Matrix Reasoning, Visual Puzzles), Working Memory Index (Digit Span, Arithmetic), and Processing Speed Index (Symbol Search, Coding). A General Ability Index can also be computed by using only the Verbal Comprehension Index and Perceptual Reasoning Index; this could provide an alternative measure of cognitive ability in cases where there is an existing impairment in working memory or processing. Internal consistency estimates across all age groups ranged from 0.97 to 0.98 for the Full Scale IQ, from 0.87 to 0.98 for the other index scores (VCI, PRI, WMI, PSI), and 0.71–0.96 for subtests.

The previous versions of this assessment, WAIS-III (Wechsler, 1997), received criticism when the factor structure employed failed to generalize to adults with intellectual disabilities (Jones, van Schaik, & Witts, 2006). Only two factors, Verbal and Performance, were found when a sample with intellectual disability was used, in contrast to the four factors suggested by the manual. Due to the practical implications of the measurement's inconsistency when evaluating those at the lower end of the intelligence distribution, people were similarly concerned when the WAIS-IV was introduced. Thankfully, the WAIS-IV four-factor structure was deemed invariant for the standard subtests when investigated using a sample with intellectual disability (Reynolds, Ingram, Seeley, & Newby, 2013), which indicates the assessment should be more generalizable across this specialty population.

Nonverbal Intelligence Tests

Individuals naturally present with a range of ability levels, and occasionally, this includes ranges in communication. The previously mentioned intelligence assessments generally rely on spoken language for instructions and the majority of responses, which could place persons with communication deficits at a disadvantage when compared to their peers without such difficulties. There are, however, well-established cognitive measures available that remove language from the evaluation of intelligence. Such evaluations can be particularly useful to persons with intellectual disabilities who have impairments with either expressive or receptive communication. A range of popular nonverbal intelligence assessment options are discussed below.

Comprehensive Test of Nonverbal Intelligence, Second Edition

The Comprehensive Test of Nonverbal Intelligence, Second Edition (CTONI-2; Hammill, Pearson, & Wiederholt, 2009) is a cognitive assessment for persons aged 6 years, 0 months to 89 years, 11 months. This measure does not require reading, writing, verbal responses, or object manipulation. There are six subtests, including Pictorial Analogies, Geometric Analogies, Pictorial Categories, Geometric Categories, Pictorial Sequences, and Geometric Sequences, and they require approximately 60 minutes to administer. The three Pictorial subtests can be combined to form the Pictorial Scale, while the three Geometric subtests can be combined to form the Geometric Scale. The Full Scale uses all six subtests. Internal consistency was high for CTONI-2 composites (Cronbach's alphas were at least 0.90), and the assessment was highly correlated with other measures of nonverbal intelligence (corrected r = 0.74 to 0.70 when compared to the Test of Nonverbal Intelligence, Fourth Edition; Brown, Sherbenou, & Johnsen, 2010).

Leiter International Performance Scale, Third Edition

The Leiter International Performance Scale, Third Edition (Leiter-3; Roid, Miller, Pomplun, & Koch, 2013) is a nonverbal measure of intelligence used with non-native English speakers (Athanasiou, 2000) or for people with hearing or communication deficits, among others (Sparrow & Davis, 2000). Unlike the nonverbal section of the Stanford-Binet, for example, instruction for the Leiter-3 is provided in pantomime and answers are given through pointing, arranging stimuli, or other task completions, which completely removes language as an influencing factor on the subsequent scores (Roid et al., 2013). This assessment can evaluate persons between the ages of 3 years, 0 months and 75 years of age. There are two batteries of tests, the Cognitive Battery and the Attention/Memory Battery, each comprised of five subtests. The Cognitive Battery measures intellectual abilities and includes the subtests Figure Ground, Form Completion, Classification/Analogies, Sequential Order, and Visual Patterns (which is optional). The Attention/Memory Battery evaluates attention and memory performance in a nonverbal manner through the following subtests: Attention Sustained, Forward Memory, Attention Divided, Reverse Memory, and Nonverbal Stroop.

Internal consistency for the Cognitive Battery subtests across all ages was good to excellent and ranged from 0.78 (Visual Patterns) to 0.95 (Sequential Order). In the norming studies using specialty samples, the Leiter-3 was able to correctly identify a cognitive delay in respondents with IQs below 75 (specificity) 99.4% of the time. This assessment is significantly correlated with the Stanford-Binet Intelligence Scales, 5th edition (r = 0.77 for the nonverbal IQ, r = 0.64 for verbal IQ), WISC-IV (r = 0.73 with WISC-IV Perceptual Reasoning Index), and WAIS-IV (r = 0.72 with WAIS-IV Perceptual Reasoning Index), which instills confidence that this nonverbal measure can effectively evaluate global cognitive ability without the use of language.

Raven's Progressive Matrices

Raven's Progressive Matrices (Raven, Raven, & Court, 1998a, 1998b, 1998c) is an assessment that can be administered individually or to a group. Each item has a pattern with a piece missing, and six answer choices are available to complete the progression. These questions are divided into sets, and items within each set are presented in order of increasing difficulty. This test is designed to measure fluid intelligence, that is, the ability to solve novel problems, rather than crystallized intelligence or accumulated knowledge which may be assessed using more traditional instruments such as the Wechsler scales. At least one study (Facon, Magis, Nuchadee, & De Boeck, 2011) determined that the assessment had adequate discriminative power in a sample of children and adolescents with intellectual disabilities when compared to a typical sample. There are multiple versions of the Progressive Matrices. Coloured Progressive Matrices (Raven et al., 1998b) includes 36 items and is for children ages 5–11 years. Standard Progressive Matrices (Raven et al., 1998c) has 60 items and

was designed for youth ages 6–17 years, although it can also be given to adults. Advanced Progressive Matrices (Raven et al., 1998a) has 48 items and was designed for those over the age of 17 years with above average intellect. The Progressive Matrices have acceptable split-half reliabilities that ranged from 0.65 to 0.94 and test-retest reliabilities that ranged from 0.71 to 0.93.

Test of Nonverbal Intelligence, Fourth Edition

The Test of Nonverbal Intelligence, Fourth Edition (TONI-4; Brown et al., 2010) is a problem-solving-based cognitive assessment for persons aged 6 years, 0 months through 89 years, 11 months that limits dependency on language and motor skills. While there are no subtests, there are two forms to reduce practice effects, Form A and Form B. These forms are not interchangeable, although they were designed to be equivalent for content and item difficulty. This brief assessment can be completed in under 20 minutes and provides the examiner the option to deliver the instructions orally or nonverbally depending on the language proficiency of the respondent, which is an interesting deviation from standard assessment protocols. Raw scores are transformed into index scores that are normally distributed (M = 100, SD = 15). Alpha coefficients for internal consistency were high, as scores ranged from 0.92 to 0.97 across age groups for both forms.

Universal Nonverbal Intelligence Test 2

The Universal Nonverbal Intelligence Test 2 (UNIT 2; Bracken & McCallum, 2016) is a cognitive measure for persons aged 5 years, 0 months to 21 years, 11 months. The Full Scale Battery is comprised of six subtests (Symbolic Memory, Nonsymbolic Quantity, Analogic Reasoning, Spatial Memory, Numerical Series, and Cube Design), although performance on selected subtests may be combined to form other composite scores. These include Memory, Reasoning, Quantitative, Abbreviated Battery, Standard Battery with Memory, and Standard Battery without Memory. These batteries or composites can be transformed into traditional index scores (M = 100, SD = 15). Alpha coefficients for internal consistency were excellent and ranged from 0.89 (Spatial Memory) to 0.98 (Standard Battery without Memory and Full Scale Battery) for all age groups, and inter-rater reliability was consistently excellent, with scores from 0.98 to 0.99.

Although there are many intelligence assessments from which to choose, the decision as to what measure is most appropriate is not as straightforward as it may appear. One reason is that researchers and clinicians operate under the premise that we are able to measure "true" cognitive ability. In reality, that would only be possible if conditions were perfect and these instruments were free of measurement error (Whitaker, 2010). Whitaker (2010) identified several considerations that counter this claim. One area was situational variance; patient behavior and level of distraction were key concerns for those assessing persons with intellectual disabilities, as was the assessor's ability to properly administer the test given potentially challenging conditions. Deviations from instrument protocol would introduce unforeseen variability in scoring, which would ultimately reduce score confidence. The presence of the Flynn effect, an observation that intelligence tests inflate a person's score by 0.3 points each year since the measure was standardized, was also mentioned (Flynn, 1984). This means that cognitive assessments must be regularly updated and have new norms established in order to combat the artificial inflation of tests scores.

Additionally, standardized scores provide a structured framework for ease of communication and comparison across cognitive measures, as they are intended to be interchangeable. While this complimentary profile appears to be true for those with typical development (e.g., Nader, Courchesne, Dawson, & Soulieres, 2016), it does not always hold true for those with intellectual disabilities who may experience greater variance in cognitive ability across different domains. For instance, Silverman and colleagues (2010) found that scores on the WAIS (e.g., Wechsler, 2008a) were significantly higher (mean difference of 16.7 points) for adults with intellectual disabilities than were scores on the Stanford-Binet

Intelligence Scales (e.g., Roid, 2003), although the most current versions of these assessments were not used in evaluating their sample. Further, the presence of additional comorbidities, such as autism spectrum disorder, can also impact test scores due to language demands. Grondhuis and colleagues (2018) reported that children with autism spectrum disorder (a condition often comorbid with intellectual disability) scored significantly higher (mean difference of 9.6 points) on the nonverbal Leiter International Performance Scale-Revised (Roid & Miller, 1997) than on the Stanford-Binet Intelligence Scales, 5th edition (Roid, 2003). Clearly, decisions about which cognitive assessment is most appropriate for a given individual should be approached with caution and consideration.

Assessment of Adaptive Behavior

Cognitive functioning is an important component to an intellectual disability diagnosis, but, as previously stated in multiple diagnostic criteria, it is by no means the only construct of consequence. Adaptive behavior is, broadly speaking, how successfully individuals are able to adapt to their environment's demands. AAIDD critiqued the structure of many adaptive behavior definitions and concluded that the concept was best operationalized by including the following three factors: conceptual skills (e.g., reading, writing, language, time, number concepts), practical skills (activities of daily living, safety, schedules, transportation, occupational skills, etc.), and social skills (interpersonal skills, self-esteem, gullibility, rule following; Schalock et al., 2010). Adaptive behavior assessments are not interchangeable with intelligence tests but, instead, should be viewed as an additional, necessary piece of the diagnostic puzzle. Where cognitive assessments gauge the extent of intellectual ability, adaptive behavior assessments describe what an individual is actually able to do in real-world situations. Similar to intelligence tests, there are many adaptive behavior assessments from which to choose. Alphabetically below are scales based on the adaptive behavior factor structure endorsed by AAIDD that were deemed to have acceptable psychometric properties in a review by Tasse and colleagues (2012). Updated versions of these assessments were included when available, as well as a new measure developed since the Tasse et al. (2012) publication.

Adaptive Behavior Assessment System-Third Edition

The Adaptive Behavior Assessment System-Third Edition (ABAS-3; Harrison & Oakland, 2015) can assess persons between birth and 89 years of age. Ten areas of adaptive skills are evaluated on a 0–3 scale that indicates how frequently, if at all, a person performs a given activity. These ten areas are broken down into three scaled domains. They are the Conceptual Domain (subtests: Communication, Functional Academics, Self-Direction), Social Domain (subtests: Leisure, Social), and Practical Domain (subtests: Community Use, School Living, Health and Safety, Self-Care). These three domains can be combined to form a General Adaptive Composite (GAC) as a single data point to summarize performance across the previously mentioned areas. There is an additional subtest Work that is not included in any of the domains or GAC. The GAC has very good psychometric properties: 0.97–0.99 for internal consistency, 0.90 for test-retest reliability, and 0.82–0.91 for inter-rater reliability.

AAMR Adaptive Behavior Scale-School, Second Edition

The AAMR (American Association on Mental Retardation, now AAIDD) Adaptive Behavior Scale-School, Second Edition (ABS-S:2; Lambert, Nihira, & Leland, 1993) was developed for persons aged 3–21 years (with an intellectual disability) or 3–18 years (without an intellectual disability) and includes two parts. Part 1 evaluates personal independence and includes the following nine domains: Independent Functioning, Physical

Development, Economic Activity, Language Development, Numbers and Time, Prevocational/Vocational Activity, Self-Direction, Responsibility, and Socialization. Part 2 evaluates social behavior and includes the following seven domains: Social Behavior, Conformity, Trustworthiness, Stereotyped and Hyperactive Behavior, Self-Abusive Behavior, Social Engagement, and Disturbing Interpersonal Behavior. This assessment, which can be given in an interview or questionnaire format, takes approximately 30 minutes to complete. The ABS-S:2 has excellent test-retest reliability (0.90).

Diagnostic Adaptive Behavior Scale

The Diagnostic Adaptive Behavior Scale (DABS; Tasse et al., 2008) is an individually administered standardized test for evaluating persons between 4 and 21 years of age, inclusive. The measure evaluates Conceptual, Social, and Practical skills, so that it aligns with both AAIDD adaptive behavior specifications (Schalock et al., 2010) and those of the DSM-5 (APA, 2013; Tasse, Schalock, Balboni, Spreat, & Navas, 2016). The interview employs a respondent such as a family member, friend, or teacher who has been able to observe the person for an extended period of time, preferably in multiple settings. Each behavioral item is rated on a 0–3 scale, where 0 indicates that the person "rarely or never does it" and 3 indicates that the person "yes – does it always or almost always independently – never or rarely needs reminders" (Tasse, Schalock, Thissen, et al., 2016). Test-retest reliability coefficients ranged from 0.78 (Social domain) to 0.95 (Practical and Conceptual domains) and convergent validity with similar adaptive behavior skills on the Vineland Adaptive Behavior Scales (second edition; Vineland-II; Sparrow, Cicchetti, & Balla, 2005) was very high, ranging from 0.70 (DABS Practical Skills and Vineland-II Daily Living Skills) to 0.84 (DABS Total and Vineland-II Composite; Tasse, Schalock, Balboni, et al., 2016).

Scales of Independent Behavior-Revised

The Scales of Independent Behavior-Revised (SIB-R; Bruinicks, Woodcock, Weatherman, & Hill, 1996) is an assessment of behavior, both adaptive and maladaptive, that contains two pieces, an Adaptive Behavior Section and a Problem Behavior Section. The Adaptive Behavior Section has four areas of measurement: Motor Skills (subscales: Gross-Motor Function, Fine-Motor Function), Social Interaction and Communication Skills (subscales: Social Interaction, Expressive and Receptive Language), Personal Living Skills (subscales: Eating and Meal Preparation, Toileting and Self-Care, Dressing, Domestic Skills), and Community Living Skills (subscales: Time and Punctuality, Money and Value, Work Skills, and Home/Community Orientation). The Problem Behavior Section has three areas of measurement: Internalized (subscales: Hurts Self, Repetitive Habits, Withdrawn, or Inattentive), Asocial (subscales: Socially Offensive, Uncooperative), and Externalized (subscales: Hurts Others, Destructive to Property, Disruptive). This measure can assess persons from 3 months of age through 80 years, and the comprehensive full form has good reliability and validity (Maccow, 2001).

Vineland Adaptive Behavior Scales-3rd Edition

The Vineland Adaptive Behavior Scales-3rd Edition (Vineland-3; Sparrow, Cicchetti, & Saulnier, 2016) is an individually administered measure of adaptive behavior that is able to assess persons from birth to 90 years of age. It can be completed using the Interview Form, Parent/Caregiver Form, or Teacher Form either on paper or online (using a tablet, for instance). The Vineland-3 contains five domains: Communication (subdomains: Receptive, Expressive, Written), Daily Living Skills (subdomains: Personal, Domestic, Community), Socialization (subdomains: Interpersonal

Relationships, Play and Leisure, Coping Skills), Motor Skills (optional; subdomains: Fine Motor, Gross Motor), and Maladaptive Behavior (optional; subdomains: Internalizing, Externalizing, Critical Items). It also provides an Adaptive Behavior Composite (ABC), which is an indicator of overall level of adaptive functioning. Each domain scale, as well as the ABC, is standardized (M = 100, SD = 15) to compare the levels of adaptive abilities of individuals to their same aged peers. Internal consistency is excellent; reliability coefficients using the comprehensive form ranged from 0.94 to 0.99 for all adaptive domains and ABC. Concurrent validity with the ABAS-3 was generally higher for Teacher responses than for those of Parent/Caregiver.

Mental Health in Persons with Intellectual Disability

Early conventional wisdom theorized that those with intellectual disabilities were at reduced risk for comorbid mental health problems thanks to reduced exposure to everyday stressors brought about by overthinking and worrying. In reality, we now know that people with intellectual disabilities are at greater risk to present with mental health problems than are persons without an intellectual disability (e.g., Emerson, 2003; Oeseburg, Dijkstra, Groothoff, Reijneveld, & Jansen, 2011; Platt, Keyes, McLaughlin, & Kaufman, 2019). This association appears to be consistent across the lifespan.

One systematic review (Einfeld, Ellis, & Emerson, 2011) investigated intellectual disability and comorbid mental health disorders in children and adolescents and found nine studies that indicated consistently high prevalence rates of mental disorders (30–50%) within the youth with intellectual disability. This was in contrast to the lower rates of mental illness reported in children without intellectual disability, which ranged from 8% (Emerson & Hatton, 2007; Rutter, Tizard, & Whitmore, 1970) to 18% (Dekker, Koot, van der Ende, & Verhulst, 2002). One study included in the review (Molteno, Molteno, Finchilescu, & Dawes, 2001) observed this trend in children under the age of 13 years; a different study by Emerson and Hatton (2007) observed this trend in children 5–10 years, specifically with regard to greater levels of hyperactivity, and in older children (11–16 years) with regard to emotional disorders; Dekker and Koot (2003) observed this trend in older children (13–20 years) with regard to obsessive-compulsive disorder. Molteno and colleagues (2001) also indicated that two studies included in their review (Emerson & Hatton, 2007; Stømme & Diseth, 2000) reported higher rates of comorbid psychopathology in males with intellectual disabilities, but that three others investigations (Dekker & Koot, 2003; Einfeld & Tonge, 1996; Molteno et al., 2001) found no such relationship between persons with dual disorders and gender.

A different systematic review of 31 studies evaluated prevalence rates of chronic health conditions, both mental and physical, among children with intellectual disability and determined the most frequently occurring conditions (Oeseburg et al., 2011). Those conditions were epilepsy (weighted mean: 22%, range: 5.5% (Dekker & Koot, 2003) to 35% (Koskentaista, Iivanainen, & Almqvist, 2002)), cerebral palsy (weighted mean: 19.8%, range: 8.4% (Christianson et al., 2002) to 33.8% (Molteno et al., 2001)), any anxiety disorder (weighted mean: 17.1%, range: 11.4% (Emerson, 2003; Emerson & Hatton, 2007) to 39% (Gothelf et al., 2008)), oppositional defiant disorder (weighted mean: 12.4%, range: 11.1% (Emerson, 2003; Emerson & Hatton, 2007) to 13.9 (Dekker & Koot, 2003)), Down syndrome (weighted mean: 11.0%, range: 2.1% (Christianson et al., 2002) to 20.3% (Fernell, 1998)), and autistic disorder (weighted mean: 10.1%, range: 4.5% (Stømme & Diseth, 2000) to 25.1% (Bradley & Bolton, 2006; Bradley & Isaacs, 2006; Bryson, Bradley, Thompson, & Wainwright, 2008)). The range of prevalence estimates within conditions highlights the difficulty of obtaining accurate measurements of these phenomena. Although the variance could be attributed to differences in operationalization or assessment, the evidence still seems compelling that these conditions regularly occur together.

Platt and colleagues (9) took the investigation of dual diagnosis a step further in their study of US adolescents from a representative population sample, rather than a clinical sample. Unlike other estimates of dual disorders that used samples from schools and confirmed only the presence of reduced intellectual capacity for study inclusion (e.g., Dekker et al., 2002), Platt and colleagues (9) required measurement of both intelligence and adaptive behavior. This inclusion of adaptive behavior is important, as other researchers (e.g., Obi et al., 2011) have previously noted that excluding levels of adaptive behavior could potentially contribute to overestimating the prevalence rates of intellectual disability. Platt and colleagues (9) reported that 65.1% of their sample met lifetime criteria for a comorbid mental disorder and that specific phobia, agoraphobia, and bipolar disorder were significantly more common in those with an intellectual disability than in those without after adjusting for parental socioeconomic status and family composition.

Evaluations focusing on adults with intellectual disabilities report somewhat similar findings of comorbid psychopathology. For instance, a systematic review by Buckles, Luckasson, and Keefe (2013) included 16 intellectual disabilities studies with samples of persons at least 16 years of age. The overall prevalence of additional mental health disorders ranged from 13.9% (Cooper, Smiley, Morrison, Williamson, & Allan, 2007) to 75% (Strydom, Hassiotis, & Livingston, 2005), although the large range may be due to study methodology. For instance, Cooper, Smiley, Morrison, and colleagues (2007) only reached their estimate of 13.9% of their 16–83-year-old sample having "mental ill-health" when using DSM-IV-TR criteria (APA, 2000) and excluding problem behaviors and autism spectrum disorder. When not excluding those disorders, the prevalence was 15.7% for DSM-IV-TR criteria and increased to 16.6% when using ICD-10-DCR criteria (WHO, 1993), 35.2% when using DC-LD criteria (Diagnostic Criteria for Psychiatric Disorder for Use with Adults with Learning Disorders/Mental Retardation; Royal College of Psychiatrists, 2001), and 40.9% when using any of those diagnostic options. The study by Strydom and colleagues (2005), on the other hand, evaluated adults at least 65 years of age without Down syndrome and reported that 75% of their sample had psychiatric *symptoms*, as opposed to meeting diagnostic criteria for specific mental health disorders. While expecting exact agreement in methodologies and diagnostic criteria used across studies is unrealistic, it is important for researchers to be explicit in what their process was for determining reported prevalence rates of comorbid conditions.

Cooper, Smiley, Finlayson, and colleagues (2007) specifically used a sample of adults with profound intellectual disabilities and assessed prevalence rates of mental health problems for the first time in this particular group. As noted before, this classification represents those who are the most cognitively impaired, which is a sample not frequently included in the literature. This is likely due to a host of potential scientific complications, including limited communication abilities, developmental level, and possibly biased informant reports. Regardless, these researchers reported prevalence of any mental ill-health in this subgroup as 11.4% for DSM-IV-TR criteria, 10.9% when using ICD-10-DCR criteria (WHO, 1993), 45.1% when using DC-LD criteria (Royal College of Psychiatrists, 2001), and 52.2% when using any of those diagnostic options. These prevalence rates are important, as they speak to psychopathology occurring at every level of intellectual disability.

Intellectual Disability and Challenges Assessing Mental Health

Understanding that persons with intellectual disabilities who experience comorbid mental health problems at a greater rate than those without intellectual disabilities is one important part of the story. The ample evidence of this connection, particularly the existence of this finding across the lifespan and the various intellectual disability severity classifications, reinforces the idea that mental health evaluations should be regularly uti-

lized in this group. Unfortunately, this process is fraught with challenges that make comorbid diagnosis more difficult. Given the far-reaching impact intellectual disability has on multiple life domains, a further look into how such a diagnosis influences the assessment of mental health is warranted.

Mental health assessments require a patient to be evaluated by a clinician, usually with a diagnostic interview or questionnaire, to determine if the problematic symptoms being reported meet criteria for a particular condition. The cognitive impairments and language difficulties so common in persons with intellectual disabilities likely reduce their ability to accurately articulate their current mental state and the cause of their distress (Bradley et al., 2007). The greater the intellectual disability severity, the greater the challenge for coherent communication of symptoms. Often times, caregivers are expected to account recent changes in the person's daily patterns as a proxy for informant report. This may be of limited utility if the caregiver is not familiar with the breadth of a person's typical behaviors or if the caregiver incorrectly infers the reason for a change.

These reporting difficulties may be further exacerbated by the presence of comorbid psychopathology. Ryan, Woodyatt, and Copeland (2010) asked a sample of adults with intellectual disability and a sample of adults with dual diagnosis (intellectual disability and psychosis) to play a game involving dice. Both groups understood the game, but the group with intellectual disability alone was able to verbalize game elements to a greater extent than was the group with dual diagnosis. Both groups were similar in scores of IQ, adaptive behavior, and vocabulary. While the authors posited that the greater difficulty in verbalization for the dual diagnosis group was likely due to the thought problems associated with psychosis, persons with intellectual disability being initially assessed for comorbid conditions could be additionally hindered in that process by their as-of-yet undiagnosed comorbidity.

It is important that clinicians assess persons with intellectual disabilities from a variety of perspectives and take all relevant information into account prior to assigning a diagnosis for a comorbid mental health condition. One reason is that there are many psychological conditions that share symptoms; generalized anxiety disorder and major depressive disorder both include problems concentrating, fatigue, and sleep disturbance as possible symptoms (APA, 2013). While shared symptomology is a challenge in the differential diagnosis process regardless of intellectual disability status, cognitive deficits may make clinical interpretation of which pieces are most impactful to one's functioning more difficult. Consulting multiple assessments and informants may provide the clinician a more comprehensive impression. Further, Platt and colleagues (2019) reported that externalizing disorders are more likely to be diagnosed in persons with intellectual disabilities when adaptive behavior level was not taken into account. The presence of overlapping symptomology may inadvertently be viewed as evidence for a behavioral disorder rather than a facet of an individual's level of functioning. Persons with intellectual disabilities can also display atypical presentation of symptoms, which may prove particularly problematic.

The presence of an intellectual disability is undoubtedly a serious and important consideration in a person's life. It is not, however, the sole consideration from a healthcare perspective, mental, or otherwise. Unfortunately, research has demonstrated that diagnostic overshadowing, that is, healthcare providers overlooking symptoms of mental or physical illness and incorrectly attributing them to an individual's intellectual disability, is a reality for many (Mason & Scior, 2004; Reiss & Szyszko, 1983). One such example is when Mason and Scior (2004) provided clinical vignettes of either a person with low IQ or normal IQ to psychiatrists and clinical psychologists. The vignettes were identical, with the exception of reported intelligence. The normal IQ vignettes were significantly more likely to be recognized as having symptoms of schizophrenia or drug problems, and were more likely to be referred for admission to an inpatient treatment facility, a mental health evaluation, or medication, than were the same vignettes belonging to a person with low IQ. Although diagnostic over-

shadowing is likely unintentional, discounting symptoms that interfere with daily functioning is troublesome and could seriously impact future quality of life.

Conclusions

Intellectual disabilities are variations in cognitive ability that contribute to the diversity of humanity. The current diagnostic criteria recommended by the DSM-5, AAIDD, and WHO state that an intellectual disability is characterized by deficits in cognition and adaptive behavior that are present prior to the end of development. This chapter presented popular intelligence tests and adaptive behavior assessments for a variety of ages and ability levels that could be used to assist the diagnostic process. It also discussed the prevalence rates of intellectual disability alone and in combination with other mental or physical disorders, and challenges to diagnosing mental health conditions comorbid with intellectual disabilities were considered. The breadth of measures and prevalence rates included was to purposefully highlight the substantial variation present within a single diagnostic condition and reinforce the notion that assessments must be individualized to best serve a person's needs.

References

American Psychiatric Association. (2000). *Diagnostic and statistical manual of mental disorders* (revised 4 ed.). Washington, DC. https://doi.org/10.1176/appi.books.9780890423349.

American Psychiatric Association. (2013). *Diagnostic and statistical manual of mental disorders* (5th ed.). Arlington, VA. https://doi.org/10.1176/appi.books.9780890425596.

Amundson, R. (2000). Against normal function. *Studies in History and Philosophy of Science Part C: Studies in History and Philosophy of Biological and Biomedical Sciences, 31*(1), 31–53.

Athanasiou, M. S. (2000). Current nonverbal assessment instruments: A comparison of psychometric integrity and test fairness. *Journal of Psychoeducational Assessment, 18*, 211–229.

Bach, M. (2007). Changing perspectives on developmental disabilities. In *A comprehensive guide to intellectual and developmental disabilities* (pp. 35–43). Baltimore, MD: Paul H. Brookes.

Bayley, N. (2006). *Bayley scales of infant and toddler development: Bayley-III* (Vol. 7). San Antonio, TX: Harcourt Assessment, Psych. Corporation.

Binet, A., & Simon, T. (1916). *The development of intelligence in children: The Binet-Simon Scale* (No. 11). Baltimore, MD: Williams & Wilkins Company.

Bracken, B. A., & McCallum, R. S. (2016). *UNIT 2: Universal nonverbal intelligence test*. Pro-Ed.

Bradley, E., & Bolton, P. (2006). Episodic psychiatric disorders in teenagers with learning disabilities with and without autism. *Journal of Intellectual Disability Research, 44*(Pt. 5), 529–543.

Bradley, E. A., & Isaacs, B. J. (2006). Inattention, hyperactivity, and impulsivity in teenagers with intellectual disability, with and without autism. *Canadian Journal of Psychiatry, 51*, 598–606.

Bradley, E. A., Summers, J., Brereton, A. V., Einfeld, S. L., Havercamp, S. M., Holt, G., … Tonge, B. (2007). Intellectual disabilities and behavioral, emotional, and psychiatric disturbances. I. In I. Brown & M. Percy (Eds.), *A comprehensive guide to intellectual & developmental disabilities* (pp. 645–666). Baltimore, MD: Paul H. Brookes Publishing Co.

Brown, L., Sherbenou, R. J., & Johnsen, S. K. (2010). *Test of nonverbal intelligence* (4th ed.). Austin, TX: PRO-ED.

Bruinicks, R. H., Woodcock, R., Weatherman, R., & Hill, B. (1996). *Scales of independent behavior – Revised*. Chicago, IL: Riverside Publishing.

Bryson, S. E., Bradley, E. A., Thompson, A., & Wainwright, A. (2008). Prevalence of autism among adolescents with intellectual disability. *Canadian Journal of Psychiatry, 53*, 449–459.

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*, 181–207.

Christianson, A. L., Zwane, M. E., Manga, P., Rosen, E., Venter, A., Downs, D., & Kromberg, J. G. R. (2002). Children with intellectual disability in rural South Africa: Prevalence and associated disability. *Journal of Intellectual Disability Research, 46*(2), 179–186.

Cooper, S. A., Smiley, E., Finlayson, J., Jackson, A., Allan, L., Williamson, A., … Morrison, J. (2007). The prevalence, incidence, and factors predictive of mental-ill health in adults with profound intellectual disabilities. *Journal of Applied Research in Intellectual Disabilities, 20*, 493–501.

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.

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 Children and Adolescent Psychiatry, 42*, 915–922.

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. *Journal of Child Psychology and Psychiatry, 43*, 1087–1098.

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 & Developmental Disability, 36*(2), 137–143.

Einfeld, S. L., & Tonge, B. J. (1996). Population prevalence of psychopathology in children and adolescents with intellectual disability: I Rationale and methods. *Journal of Intellectual Disability Research, 40*(2), 91–98.

Elliott, C. D. (2007). *Differential ability scaled* (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.

Emerson, E., & Hatton, C. (2007). Mental health of children and adolescents with intellectual disability in Britain. *British Journal of Psychiatry, 191*, 493–499.

Facon, B., Magis, D., Nuchadee, M., & De Boeck, P. (2011). Do Raven's Colored Progressive Matrices function in the same way in typical and clinical populations? Insights from the intellectual disability field. *Intelligence, 39*, 281–291.

Farmer, C., Golden, C., & Thurm, A. (2016). Concurrent validity of the differential ability scales, second edition with the Mullen scales of early learning in young children with and without neurodevelopmental disorders. *Child Neuropsychology, 22*(5), 556–569.

Fernell, E. (1998). Aetiological factors and prevalence of severe mental retardation in children in a Swedish municipality: The possible role of consanguinity. *Developmental Medicine and Child Neurology, 40*, 608–611.

Flynn, J. R. (1984). The mean IQ of Americans: Massive gains 1932 to 1978. *Psychological Bulletin, 95*, 29–51.

Fombonne, E. (2003). Epidemiological surveys of autism and other pervasive developmental disorders: An update. *Journal of Autism and Developmental Disorders, 33*(4), 365–381.

Goddard, H. H. (1920). *Human efficiency and levels of intelligence*. Princeton, NJ: Princeton University Press.

Gothelf, D., Goraly, O., Avni, S., Stawski, M., Hartmann, I., Basel-Vanagaite, L., & Apter, A. (2008). Psychiatric morbidity with focus on obsessive–compulsive disorder in an Israeli cohort of adolescents with mild to moderate mental retardation. *Journal of Neural Transmission, 115*(6), 929–936.

Grondhuis, S. N., Lecavalier, L., Arnold, L. E., Handen, B. L., Scahill, L., McDougle, C. J., & Aman, M. G. (2018). Differences in verbal and nonverbal IQ test scores in children with autism spectrum disorder. *Research in Autism Spectrum Disorders, 49*, 47–55.

Hammill, D. D., Pearson, N. A., & Wiederholt, J. L. (2009). *Comprehensive test of nonverbal intelligence* (2nd ed.). Austin, TX: PRO-ED.

Harrison, P. L., & Oakland, T. (2015). *Adaptive behavior assessment system-third edition (ABAS-3)*. Torrence, CA: Western Psychological Services.

Jones, J. J. S., van Schaik, P., & Witts, P. (2006). A factor analysis of the Wechsler Adult Intelligence Scale 3rd edition (WAIS-III) in a low IQ sample. *British Journal of Clinical Psychology, 45*, 145–152.

Kaufman, A. S., & Kaufman, N. L. (2004). *Kaufman Brief Intelligence Test* (2nd ed.). Bloomington, MN: Pearson, Inc..

Kaufman, A. S., & Kaufman, N. L. (2018). *Kaufman assessment battery for children, second edition normative update (KABC-II NU)*. Bloomington, MN: Pearson, Inc..

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.

Koskentaista, T., Iivanainen, M., & Almqvist, F. (2002). Psychiatric disorders in children with intellectual disability. *Nordic Journal of Psychiatry, 56*, 126–131.

Lambert, N., Nihira, K., & Leland, H. (1993). *ABS-S:2: AAMR adaptive behavior scale: School*. Pro-ed.

Maccow, G. (2001). Test review of the scales of independent behavior-revised. In B. S. Plake & J. C. Impara (Eds.), *The fourteenth mental measurements yearbook*. [Electronic version]. Available from http://marketplace.unl.edu/buros/

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*, 85–90.

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

Molteno, G., Molteno, C. D., Finchilescu, G., & Dawes, A. R. (2001). Behavioural and emotional problems in children with intellectual disability attending special schools in Cape Town, South Africa. *Journal of Intellectual Disability Resarch, 45*, 515–520.

Nader, A. M., Courchesne, C., Dawson, M., & Soulieres, I. (2016). Does WISC-IV underestimate the intelligence of autistic children? *Journal of Autism and Developmental Disorders, 46*(5), 1582–1589.

Obi, O., Van Naarden Braun, K., Baio, J., Drews-Botsch, C., Devine, O., & Yeargin-Allsopp, M. (2011). Effect of incorporating adaptive functioning scores on the prevalence of intellectual disability. *American Journal on Intellectual and Developmental Disabilties, 116*(5), 360–370.

Oeseburg, B., Dijkstra, G. J., Groothoff, J. W., Reijneveld, S. A., & Jansen, D. E. M. C. (2011). Prevalence of chronic health conditions in children with intellectual

disability: A systematic literature review. *Intellectual and Developmental Disabilities, 49*(2), 59–85.

Platt, J. M., Keyes, K. M., McLaughlin, K. A., & Kaufman, A. S. (2019). Intellectual disability and mental disorders in a US population representative sample of adolescents. *Psychological Medicine*, *49*(6), 952–961.

Raven, J., Raven, J. C., & Court, J. H. (1998a). *Advanced progressive matrices*. Oxford, UK: Oxford Psychologists Press.

Raven, J., Raven, J. C., & Court, J. H. (1998b). *Coloured progressive matrices*. Oxford, UK: Oxford Psychologists Press.

Raven, J., Raven, J. C., & Court, J. H. (1998c). *Standard progressive matrices*. Oxford, UK: Oxford Psychologists Press.

Reiss, S., & Szyszko, J. (1983). Diagnostic overshadowing and personal professional experience with mentally retarded persons. *American Journal of Mental Deficits, 87*, 396–402.

Reynolds, M. R., Ingram, P. B., Seeley, J. S., & Newby, K. D. (2013). Investigating the structure and invariance of the Wechsler Adult Intelligence Scales, fourth edition in a sample of adults with intellectual disability. *Research in Developmental Disabilities, 34*, 3235–3245.

Roid, G. H. (2003). *Stanford-Binet intelligence scales* (5th ed.). Itasca, IL: Riverside.

Roid, G. H., & Miller, L. J. (1997). *Leiter international performance scale—revised*. Wood Dale, IL: Stoelting.

Roid, G. H., Miller, L. J., Pomplun, M., & Koch, C. (2013). *Leiter international performance scale, (Leiter-3)*. Los Angeles, CA: Western Psychological Services.

Royal College of Psychiatrists (2001). *DC-LD: Diagnostic Criteria for Psychiatric Disorders for Use with Adults with Learning Disabilities/Mental Retardation,* (Occastional Paper OP48). Gaskell.

Rutter, M., Tizard, J., & Whitmore, K. (Eds.). (1970). *Education, health and behavior*. London, UK: Longman.

Ryan, J., Woodyatt, G., & Copeland, D. (2010). Procedural discourse in intellectual disability and dual diagnosis. *Journal of Intellectual Disability Research, 54*(1), 70–80.

Sattler, J. M. (2001). *Assessment of children; cognitive applications*. La Mesa, CA: Jerome 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: Diagnosis, classification, and systems of supports* (11th ed.). Washington, DC: American Association on Intellectual and Developmental Disabilities.

Schalock, R. L., Luckasson, R. A., Shogren, K. A., Borthwick-Duffy, S., Bradley, V., Buntinx, W. H. E., … Yeager, M. H. (2007). The renaming of mental retardation: Understanding the change to the term intellectual disability. *Intellectual and Developmental Disabilities, 45*(2), 116–124.

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., & Balla, D. A. (2005). *Vineland-II: Vineland adaptive behavior scales* (2nd ed.). Minneapolis, MN: Pearson Assessments.

Sparrow, S. S., Cicchetti, D. V., & Saulnier, C. A. (2016). *Vineland adaptive behavior scales, third edition (Vineland-3)*. San Antonio, TX: Pearson.

Sparrow, S. S., & Davis, S. M. (2000). Recent advances in the assessment of intelligence and cognition. *The Journal of Child Psychology and Psychiatry and Allied Disciplines, 41*(1), 117–131.

Strydom, A., Hassiotis, A., & Livingston, G. (2005). Mental health and social care needs of older people with intellectual disabilities. *Journal of Applied Research in Intellectual Disabilities, 18*(3), 229–235.

Stømme, P., & Diseth, T. H. (2000). Prevalence of psychiatric diagnoses in children with mental retardation: Data from a population-based study. *Developmental Medicine and Child Neurology, 42*, 266–270.

Tasse, M. J., Schalock, R. L., Balboni, G., Bersani, H., Borthwick-Duffy, S. A., Spreat, S., … Zhang, D. (2008). *Diagnostic adaptive behavior scale: User's manual*. Washington, DC: American Association on Intellectual and Developmental Disabilities, in press-b.

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), 291–303.

Tasse, M. J., Schalock, R. L., Balboni, G., Spreat, S., & Navas, P. (2016). Validity and reliability of the diagnostic adaptive behaviour scale. *Journal of Intellectual Disability Research, 60*(1), 80–88.

Tasse, M. J., Schalock, R. L., Thissen, D., Balboni, G., Bersani, H., Borthwick-Duffy, S. A., … Navas, P. (2016). Development and standardization of the Diagnostic Adaptive Behavior Scale: Application of item response theory to the assessment of adaptive behavior. *American Journal on Intellectual and Developmental Disabilities, 121*, 79–94.

Thorndike, R. L., Hagen, E. P., & Sattler, J. M. (1986). *Stanford-Binet intelligence scale* (4th ed.). Chicago,IL: Riverside Publishing Company.

Van Naarden Braun, K., Christensen, D., Doernberg, N., Scheive, L., Rice, C., Wiggins, L., … Yeargin-Allsopp, M. (2015). Trends in prevalence of autism spectrum disorder, cerebral palsy, hearing loss, intellectual disability, and vision impairment, metropolitan Atlanta, 1991-2010. *PLoS One, 10*(4), e0124120.

Wechsler, D. (1939). *Wechsler-Bellevue intelligence scale*. New York, NY: Psychological Corporation.

Wechsler, D. (2003). *Wechsler intelligence scales for children* (4th ed.). San Antonio, TX: TX: The Psychological Corporation.

Wechsler, D. (2008a). *Wechsler adult intelligence scale—fourth edition*. San Antonio, TX: Pearson Assessment.

Wechsler, D. (2008b). *Wechsler adult intelligence scale—fourth edition: Technical and interpretive manual*. San Antonio, TX: Pearson Assessment.

Wechsler, D. (2012). *Wechsler preschool and primary scale of intelligence—fourth edition*. San Antonio, TX: Psychological Corporation.

Wechsler, D. (2014a). *Wechsler intelligence scales for children* (5th ed.). San Antonio, TX: NCS Pearson.

Wechsler, D. (2014b). *Wechsler intelligence scales for children-fifth edition technical and interpretive manual*. San Antonio, TX: NCS Pearson.

Whitaker, S. (2010). Error in the estimation of intellectual ability in the low range using the WISC-IV and WAIS-III. *Personality and Individual Difference, 48*(5), 517–521.

World Health Organization. (1993). *The ICD-10 classification of mental and behavioural disorders: Diagnostic criteria for research*. WHO.

World Health Organization (2018). *International classification of diseases for Mortality and Morbidity Statistics* (11th Revision).

Wechsler, D. (1997). *Wechsler adult intelligence scale, 3rd edn* (WAIS-III). San Antonio, TX: Psychological Corporation.

Assessment of Anxiety in Persons with Dual Diagnosis

13

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Introduction

Anxiety disorders are a group of problems based on an individual having excessive worry and/or fear that has begun to meaningfully interfere with daily life. This is significantly different from normative fear or worry as it is marked by a disproportionate occurrence and intensity. Typically, individuals with anxiety have both emotional and behavioral responses to stressors, and this manifestation can occur across the lifespan and across cultures. The overall prevalence of anxiety has been found to be higher in adulthood compared to childhood, adolescence, and emerging adulthood (Essau, Lewinsohn, Lim, Moon-ho, & Rohde, 2018).

Individuals with anxiety tend to experience interference across multiple settings including at home, at school, and at work, and the development and persistence of anxiety can negatively affect personal relationships. Furthermore, anxiety disorders are also one of the most common co-occurring disorders, and individuals of all levels of cognitive functioning can experience anxiety (Matson & Bamburg, 1998). Even so, there is limited research on the assessment of anxiety in individuals with intellectual disabilities (ID) due to the complex presentations of anxiety in this population. Early (or at least expedient) identification and intervention is still key; however, and it is imperative that anxiety is accurately diagnosed and the child, adolescent, or adult with or without intellectual disabilities is referred for appropriate treatment as soon as possible. It is to this expedient and accurate identification that we now turn. This chapter summarizes research related to the assessment of anxiety generally in children, adolescents, and adults with ID, the challenges of assessing anxiety in individuals with ID, and then outlines the current assessment tools (i.e., rating scales, behavioral interviews, behavioral observations, physiological measures) useful with anxiety in children, adolescents, and adults who also have co-occurring ID.

Anxiety Disorders Defined

While there is considerable overlap, anxiety disorders differ based on the predominance of worry versus fear, and they are further differentiated based on the types of stimuli that induce symptoms, distress, and avoidance. There are 11 anxiety disorders recognized by the *Diagnostic and Statistical Manual of Mental Disorders, 5th Edition (DSM-5)* (APA, 2013): separation anxiety disorder, selective mutism, specific phobia, social anxiety disorder, panic disorder, agoraphobia, generalized anxiety disorder, substance/medication-induced anxiety disorder, anxiety

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disorder due to another medical condition, other specified anxiety disorder, and unspecified anxiety disorder.

Separation anxiety disorder is characterized by fear or anxiety over separating from an attachment figure or the home environment itself (APA, 2013). The fearful or anxious thoughts or behaviors are developmentally inappropriate. Sufferers are fearful of events or harm that could happen to the attachment figure(s) or themselves and often behave in a manner to reduce separation from the attachment figure (e.g., tantrums when an attachment figure is about to leave without them, excessive clinginess, often sleeping in the same bed or bedroom as the attachment figure). There is also the possibility of nightmares and physical symptoms of distress associated with this disorder. Individuals may struggle with sleeping away from home, leaving the home, or being home alone. The symptoms must be persistent, lasting at least 4 weeks in children or adolescents and 6 months in adults (APA, 2013).

According to the *DSM*-5, selective mutism is characterized by the failure to speak in social situations in which there is an expectation for communication and conversation. The individual chooses not to speak in a given situation but has no trouble speaking in other situations—an important consideration in those also having intellectual disability or potentially other non-anxiety-related speech difficulties. The failure to speak must negatively impact the individual in multiple settings (e.g., school, occupation, social communication) and must not be from a lack of knowledge or comfort of the spoken language. The symptoms also cannot be better explained by a communication disorder. The duration must be at least 1 month but is not limited to the first month of school (APA, 2013).

Specific phobia is characterized by a fearful, anxious, or avoidant response to objects or situations lasting 6 months or more (APA, 2013). The phobic response follows the interaction with the phobic object or situation and is inappropriate for the actual risk posed. The types of specific phobia include animal, natural environment, blood-injection injury, situations, and other (APA, 2013). The disturbance should also not be better explained by other mental disorders (e.g., objects or situations related to obsessions in obsessive-compulsive disorder; APA, 2013).

Social anxiety disorder is characterized by fearful, anxious, or avoidant reactions to social situations or when an individual may feel he or she is being scrutinized (e.g., meeting unfamiliar people, performing in front of someone else; APA, 2013). This includes fearing negative evaluation by others to the point that the individual feels embarrassed, humiliated, or rejected. The fear of the social situations must be persistent, out of proportion to the actual threat posed (APA, 2013).

Per *DSM*-5, panic disorder is the reoccurrence of unexpected panic attacks and persistent worry about when the next panic attack will occur; this fear, in turn, maladaptively leads to changes in one's behavior to avoid situations where an attack may occur. Panic attacks (note: different than panic disorder) are characterized by an abrupt surge of fear from exposure to a fearful stimulus or unexpectedly with no apparent reason. Similarly, agoraphobia may be associated with panic attacks. Agoraphobia is distinguished by fear or anxiety about two or more of the following: using public transportation, being in open spaces, being in enclosed places, standing in line or being in a crowd, or being outside of the home alone (APA, 2013). A key component of agoraphobia, however, is the fear that something will go wrong with the individual (e.g., have a panic attack) and they will not be able to escape or get help (APA, 2013).

Generalized anxiety disorder is characterized by persistent and excessive worry that cannot be controlled (APA, 2013). This worry extends to various domains such as school, work, performance, one's own health and the health of others, and the community. Generalized anxiety disorder is also marked by physical symptoms. Symptoms must have been present for more days than not for at least 6 months (APA, 2013).

There are four additional anxiety disorders in the *DSM-5*. Substance/medication-induced anxiety disorder is characterized by anxiety due to substance or medication treatment intoxication or withdrawal. Anxiety disorder due to another

medical condition is associated with anxiety symptoms that are the physiological consequence of another medical condition. Additionally, the *DSM-5* describes another specified anxiety disorder, which applies to symptoms of anxiety disorders not meeting full criteria for an anxiety disorder due to a specified reason. Unspecified anxiety disorder applies to cases in which symptoms of an anxiety disorder do not meet full criteria, but the reason is not specified (APA, 2013).

Anxiety and Intellectual Disabilities

The study of the prevalence and presentation of secondary psychopathologies among individuals with intellectual disabilities has been steadily increasing. Historically, there has been diagnostic overshadowing of secondary psychopathology in those with intellectual disability, an erroneous assumption that individuals with intellectual disabilities could either not experience mental health problems at all or at least not in addition to the intellectual disability (Matson, Belva, Hattier, & Matson, 2012). Moreover, due to diagnostic overshadowing and the false attribution of comorbid mental health symptoms to the intellectual disability itself, many individuals' dual diagnoses went unrecognized and, hence, untreated. To the contrary, a meta-review by Dykens (2000) and similar literature show an increased risk of additional psychiatric disorders for individuals with intellectual disability compared to their typically developing peers. Specifically, a common trend with dual diagnosis has been the comorbidity of anxiety disorders and ID (Costello, Mustillo, Erkanli, Keeler, & Angold, 2003; Emerson & Hatton, 2007).

Reardon, Gray, and Melvin (2015) examined the prevalence rates of anxiety disorders in those with ID recently in a meta-analysis. Among children with ID, the 12-month prevalence rates for anxiety disorders ranged between 3% and 21.9% (Dekker & Koot, 2003; Stromme & Diseth, 2000). Separation anxiety had the highest prevalence rates for younger children (13.7–17.6%) and decreased substantially in older children to 2.1–2.7% (Baker, Neece, Fenning, Crnic, & Blacher, 2010; Dekker & Koot, 2003; Emerson, 2003; Emerson & Hatton, 2007; Green, Berkovits, & Baker, 2015). The prevalence rates for social anxiety disorder ranged from 0.8% to 2.7% across a wide age range; however, the rate increased substantially to 10.8% in a study by Green et al. (2015) who were looking at 9-year-olds. For generalized anxiety disorder, prevalence rates ranged from 0% to 5.4% (Dekker & Koot, 2003; Green et al., 2015). Lastly, among the few studies that included specific phobia, panic disorder, and agoraphobia, rates were each approximately 2%. Even so, large variability was also observed in the rates: for example, there was one study that reported the prevalence rate of specific phobia was 17.5% (Dekker & Koot, 2003). Additionally, prevalence rates for individuals with mild or moderate ID may vary greatly from those with severe and profound ID; however, systematic research using measures designed for severe and profound levels has not been conducted (Matson, Smiroldo, Hamilton, & Baglio, 1997).

Anxiety among typically developing individuals usually includes a form of subjective feeling (i.e., an individual's subjective, personal appraisal of their symptoms in total) (e.g., cognitive ideations, unrealistic thinking; Davis & Ollendick, 2005), which may not be possible for some individuals with ID depending on their degree of cognitive and developmental impairment. Much of the research delving into the presentation of anxiety disorders among individuals with ID focuses on the mild to moderate levels, and little research has focused on the severe and profound levels (Matson et al., 1997). Anxiety may present in a variety of ways among individuals overall, and the same is true with individuals also having ID. Since the subjective reporting of feelings may be too cognitively demanding, the symptoms of anxiety in this population may need to be ascertained from observable behaviors (e.g., avoidance, trembling, shortness of breath, facial expressions). There also may be difficulty with specifying fears which may make identifying the subtypes of anxiety troublesome leading to a more generalized nature (Matson et al., 1997).

Ollendick, Oswald, and Ollendick (1993) assessed individuals with severe and profound ID and found that the primary source of information for anxiety symptoms came from other reports such as self-report and informant measures. In a study by Matson et al. (1997), symptoms of the *DSM-IV* diagnosis of anxiety that could be reliably identified for the majority of individuals with severe or profound ID were identified: becomes upset, hides or shields face when confronted by unfamiliar people or situations, sudden movement, sudden vocal responses, occasionally seems upset or uncomfortable, looks down a lot, sleep problems, increased arousal/exaggerated startle response, restlessness, easily fatigued, muscle tension, sweating, and trembling or shaking. The most problematic symptoms to identify with the severe or profound population included excessive worry, fear of a particular stimulus, anxiety about and avoidance of potentially feared stimuli, unreasonable thoughts, and physiological signs (e.g., feeling of choking, heart palpitations; Matson et al., 1997).

Dual Diagnosis

Other mental health conditions should be considered with a dual diagnosis of anxiety disorders for those with severe or profound levels of ID. Matson et al. (1997) described other possible explanations for behavioral symptoms that seem to align with anxiety disorders so that differential diagnoses can be conducted, including:

- Sudden motor or vocal responses may be indicative of another movement disorder (Schroeder, 1991).
- Shaking may be linked to involuntary movement disorders or the side effects of psychotropic drugs, which many individuals with severe or profound intellectual disabilities are prescribed (Aman & Singh, 1991).
- Sleep difficulties may be a result of other diagnoses such as depression (APA, 2013).
- Individuals diagnosed with autism spectrum disorder also present with startle responses (Schopler, Reichler, & Renner, 1988).

Multiple behavioral observations need to be conducted to observe the limited number of reliable symptoms described by Matson et al. (1997).

Individuals with ID have a higher risk of mental health problems than others who have more typically developing intelligence (Dekker, Nunn, & Koot, 2002; Emerson, 2003). Some factors associated with the increased risk of psychological disorders include increasing age, low socioeconomic status, reduced household income, living with one biological parent, living in an institution, severity level inadequate socialization, limitations on daily living skills, poor communication skills, epilepsy, and mobility. As well, a higher risk for anxiety has been associated with specific genetic syndromes such as fragile X syndrome. Additionally, higher adaptive functioning resulted in decreased risk of overall psychopathology, suggesting that a person's ability to navigate demands in the environment successfully decreases the risk of having an additional mental health problem (Koskentausta, Livanainen, & Almqvist, 2007).

Associations between risk factors for a secondary diagnosis and the level of ID may be explained (at least partially) by differences in assessment practices. The higher risk of a dual diagnosis with moderate ID may be due to increased abilities to effectively communicate or observe behaviors compared to severe and profound levels of ID during an assessment. (Koskentausta et al., 2007). Some data suggests that decreasing IQ is a risk factor for additional psychopathology (Chadwick et al., 2000; Eyman & Call, 1977; Gillberg et al., 1986; Jacobson, 1982), which conflicts with the findings by Koskentausta et al. (2007). However, if the behaviors are harder to detect, any additional mental health problems may be missed due to diagnostic overshadowing. Overall, individuals with ID are at a higher risk for developing a secondary psychopathology than their respective typically developing peers.

As previously discussed, the presenting symptoms of anxiety may look different in those also having ID than in typically developing individuals (Ollendick et al., 1993). In a literature review by Hagopian and Jennett (2008), only 48 studies

in the previous 35 years had looked at anxiety among individuals with ID, while the prevalence rates of these dual diagnoses have drastically increased. There are a multitude of factors to consider when thinking about the causes of the lack of literature in these populations. Evidence-based protocols for anxiety disorders have not previously included individuals with ID in their sample. Additionally, clinicians assessing for ID may not focus on additional mental health problems but instead may focus on adaptive functioning or other related difficulties. The diagnosis of ID may be more prioritized than a differential diagnosis, particularly when working with an individual with a severe or profound ID (Ehrenreich-May & Remmes, 2013).

Purposes of Assessment

Assessment is a problem-solving process that leads to an informed decision based on the gathering of relevant information about an individual (American Psychological Association, 2000). It should be viewed as a decision-making process using both inductive and deductive reasoning alternately. Inductive reasoning begins with specific observations (e.g., trembling or shaking) and moves toward a formulation of potential explanations for these observations or behaviors (e.g., the individual might be anxious when encountering the stimuli). Alternatively, deductive reasoning begins with a concept or theory (e.g., individuals with anxiety tremble and shake) and works down to specific conclusions or observations (e.g., this person is anxious because they are shaking). Diagnostic decision-making emphasizes the dynamic relationship between inductive and deductive reasoning, particularly when distinguishing between multiple diagnoses. Research has shown that assessors typically formulate approximately three to five initial hypotheses based on observations using inductive reasoning and then consider these hypotheses when finding additional evidence. Using deductive reasoning, the hypotheses can be narrowed into diagnoses. On the other hand, an assessor might initially focus on one particular diagnosis, but with the evidence gained from the assessment, the problems the individual is experiencing may be accounted for by multiple diagnosis rather than just one. When parsing out symptoms between ID and anxiety disorders, switching between inductive and deductive reasoning may be helpful (Suhr, 2015).

Arriving at the correct diagnostic label is important. Diagnostic labels can enhance the communication between professionals and reduce confusion with subsequent recommendations and treatment. Additionally, understanding dual diagnoses can aid in recommending tailored interventions, particularly when searching among the current variety of empirically supported treatments which may be specifically tied to research evidence based on careful diagnostic grouping (Davis, May, & Whiting, 2011; Silverman & Ollendick, 2005). While researchers have shown that treating a primary diagnosis can aid in the reduction of a secondary diagnosis (e.g., Davis, Ollendick, & Öst, 2019), there are multiple constructs that may hinder the overall treatment process (e.g., fearful of unknown situations or people), and generalization of treatment effects may only be negligible for any given individual. Additionally, understanding comorbid anxiety in those with an ID allows clinicians to monitor treatment outcomes appropriately. There is variability in treatment progress between individuals with ID and typically developing individuals (particularly if a treatment is being adapted for use from a typically developing population; Davis, 2012); however, the reduction of fear or avoidance among people with ID is conducive to an increase in the quality of life (Sattler & Hoge, 2006; Silverman & Ollendick, 2005).

Challenges of Assessment

Biases can be problematic during the assessment process. Representative bias occurs when a symptom typical to a disorder is ascertained be representative of that disorder. If an individual is avoidant and fearful of a stimulus, a diagnosis of an anxiety disorder without other information would be an example of a representative bias.

Availability bias occurs when a diagnosis is made on the most available or salient information such as the presenting concern or referral question. A hindsight bias is characterized by a clinician being influenced to provide a diagnosis because it was once made in the past. Regret bias is when an evaluator overestimates the base rate of a diagnosis because of fear that they will miss the diagnosis. Confirmatory bias is the tendency to seek evidence for validation for your original hypothesis while ignoring counter evidence. Lastly, diagnostic bias involving clinicians seeing an oft-given diagnosis regardless of an individual's circumstances should be avoided. For example, if an individual presents with difficulties, assigning a diagnosis even though the problems are not at diagnostic threshold would be inappropriate and possibly a bias (Suhr, 2015).

Assessing for anxiety among typically developing children often requires a number of methods including self-report, parent/caregiver-report, and teacher-report, which often may leads to increased discrepancies when the informants disagree (Ehrenreich-May & Remmes, 2013). When assessing individuals with ID, informant reports from caregivers and other professionals that work closely with the individual are necessary due to often limited communication from the individual. Researchers have shown that there are typically larger discrepancies among informants when reporting internalizing behaviors compared to externalizing behaviors (De Los Reyes & Kazdin, 2004), increasing the difficulty of assessing for anxiety disorders. When assessing for anxiety among individuals with an ID, focusing on the behavioral aspects can be helpful to negate informant discrepancies. For example, paying more attention to the results of a functional assessment of avoidant behavior rather than emotional concerns may align more with informant report. For more severe or profound levels of ID, the observable behavior should be the main focus for moderate to frequently occurring symptoms and behaviors, which in turn should lead to less informant discrepancies (De Los Reyes & Kazdin, 2004).

Individual differences in severity of ID present a particular challenge to assessing anxiety disorders (e.g., cognitive functioning). People who have mild to moderate levels of ID may be able to identify and verbalize some cognitions and physiological symptoms, whereas people who have severe to profound levels of ID would not have this capability. Moreover, an individual's communication skills will also vary. Self-reports can aid in the assessment process; however, this is not always feasible, valid, or realistic.

A major challenge to the assessment of anxiety among individuals with ID is the determination of whether observed behavioral problems are due to anxiety or other problems (e.g., defiance, hyperactivity/impulsivity; Hagopian & Jennett, 2008). It is common for a clinician to attribute behavior and presenting symptomology to an individual's cognitive impairments; thus, the dual diagnosis of an anxiety disorder or another related psychological disorder is often missed (Reiss, Levitan, & Szyszko, 1982). Functional behavioral assessment of avoidant behavior can be helpful when probing for anxious behavior in populations with ID. However, individuals with ID may display behavioral avoidance or negative reactions to situations that are non-preferred as opposed to situations that are fear-inducing. The presence of reinforcers may potentially induce avoidance behavior. For example, if there are multiple positive reinforcers at home, the child may be avoidant of school; therefore, the avoidance is not potentially caused by fear but would be caused by the positive reinforcers at home (e.g., a preferred activity). On the contrary, if an individual is avoidant of a stimulus while presenting with fearful facial expressions and physiological arousal across environments, the distinction may become clear, and the clinician should be aware of the possible presence of anxiety (Hagopian & Jennett, 2008). The difficulty in differentiating the symptoms of anxiety from those related to having an ID has contributed in part to the lack of literature. The lack of literature further contributes challenges to selecting the most appropriate tools and methods for assessing anxiety disorders based on an informed, evidence base among individuals with ID.

Assessment of Anxiety and Intellectual Disabilities

In order to accurately assess anxiety in individuals with ID, a comprehensive approach is strongly recommended (Hagopian & Jennett, 2008). Multi-informant and multi-method assessments include the direct observation of behavior, parent/caregiver interviews and informant-reported rating scales, and self-reported information to the extent possible. Multimodal assessments improve diagnostic accuracy (Dykens, 2000). March and Albano (1996) suggested obtaining self-report data from children over the age of 7 years, but the communication and cognitive deficits associated with ID make collection of self-report even more difficult. Further, discrepancies can occur between the symptoms of anxiety and depression reported by adults with ID and the symptoms reported by their caregivers; adults with ID have reported more autonomic symptoms, while their caregivers reported that the adult was experiencing more affective symptoms of anxiety or depression (Moss, Prosser, Ibbotson, & Goldberg, 1996). Literature supports the notion that individuals with ID are less likely to report changes in their affective states; thus, caregivers are typically expected to provide these details based on their observations (Sovner & Hurley, 1983). Therefore, having multiple sources of information (caregiver and self-report) will aid the clinician in conceptualizing the individual with ID's anxiety as accurately as possible.

When conducting any type of assessment with an individual with ID, there are both safety and ethical concerns. Individuals with ID who have comorbid anxiety, at times, present with more aggressive behavior as an expression of their anxiety (Dykens, 2000; Stavrakaki, 1999), and the desire to avoid hastily could also potentially harm the individual as well. In order to ensure safety, clinicians should be trained and have experience working with this population. Furthermore, ethical issues need to be considered when working with any vulnerable population. It is important there is an adult guardian present during the consent process to ensure that informed consent is provided in the event the individual with ID being assessed does not have the cognitive capacity to provide his or her own consent (Carlson, 2013). One should also attempt to obtain informed assent if at all possible: the individual's agreement to proceed based on developmentally appropriate descriptions even if it is not the legally binding consent. Additionally, modifying the consent documents to include simpler language and visual representation of the main points may be helpful for an individual with ID to understand, especially if the individual is nonverbal. In either case, as a major competent of efficacious treatment for anxiety and fear is exposure, additional care with regard to the safe and ethical planning and executions of exposure is recommended (see Davis, Ollendick, & Öst, 2012 for more on these issues).

Assessment Methods

The majority of instruments that are used to assess psychopathology in this population were not originally created or standardized for individuals with ID; often the items on these scales have been designed for individuals with typical cognitive functioning (Esbensen, Rojahn, Aman, & Ruedrich, 2003), and problems with reliability and/or validity may be present when used with this population. Moreover, the symptoms that an individual with ID may present with could be different than what the measure attributes as symptoms of particular disorder; thus, the ratings made with these measures may not be accurate. Additionally, dual diagnosis assessment instruments should have high face validity and content validity; the content of the items on these instruments should measure all the major areas of psychopathology as well as specific content areas (Reiss & Valenti-Hein, 1994). Many dual diagnosis instruments used to assess anxiety specifically in children, adolescents, and adults with ID vary widely in length, format, and psychometric proprieties (Hermans, van der Pas, & Evenhuis, 2011). There are a few assessment instruments that were specifically designed to measure anxiety in individuals with ID, and there is sparse research examining the psychometrics of these

measures. Even so, the following sections outline both broadband and anxiety-specific rating scales and behavioral interviews that can be utilized in the assessment of anxiety with children, adolescents, and adults with varying severity levels of ID (though readers are cautioned given the inherent limitations discussed so far). The psychometric properties of each measure are discussed.

Rating Scales

A frequently utilized method to obtain information about one's symptoms and/or experiences is rating scales. Rating scales typically supplement a behavioral interview. These measures can provide clinicians with a general overview of an individual's psychopathology for the initial assessment or can be disorder specific, and there frequent incorporation of normative comparisons for scores provides a convenient way to estimate the severity, frequency, intensity, etc. of a desired symptom relative to a studied sample. Rating scales can be completed by caregivers, teachers, and other individuals who can provide valuable insight. There are also scales that are self-report; those individuals with mild ID may be able to report their own symptomology in addition to other caregivers. The format of the self-report rating scale is important to consider when working with an individual with ID. There has been success with obtaining reliable information and utilizing self-report measures with yes/no responses or simple Likert scales that are easy to understand (Hartley & MacLean, 2006). Additionally, modifications can be made to some rating scales including supplementing the text with pictorial representations, since many individuals with ID can recognize concepts visually rather than just reading text (Hartley & MacLean, 2006), though it should be understood this may change the inherent psychometrics. There are, however, limitations to using rating scales with this population. For instance, rating scales tend to be more sensitive to atypical behaviors that are commonly associated with individuals with ID and therefore may not be truly representative of the actual symptoms one is trying to assess (Dykens, 2000).

Broadband Rating Scales

Achenbach System of Empirically Based Assessment (ASEBA; Achenbach 1991) The Achenbach System is one of the most widely used broadband questionnaire systems, and it is made up of the Childhood Behavior Checklist (CBCL), which is completed by a child's caregivers; the Teacher Report Form (TRF), which is completed by the child's teacher; and the Youth Self-Report Form (YSR). Both the CBCL and the TRF measure a child or adolescent's emotional and behavioral functioning across a variety of domains and can be completed for children 1–5½ years old and 6–18 years old. Furthermore, the YSR was created for youth between 11 and 18 years to complete on their own. All three versions have an overall internalizing problems scale and a DSM-oriented anxiety problems scale; the clinical syndrome scales also include questions that measure anxious/depressed symptomology. Research supports the use of the CBCL and YSR to discriminate between those diagnosed with anxiety disorder from non-disordered individuals (van Meter et al., 2014). T-scores greater than 69 indicate a greater likelihood that the youth is experience an anxiety disorder.

There have been multiple studies that have examined the CBCL, TRF, and YSR Anxious/Depressed subscales in children and adolescents with ID. Koskentausta, Iivanainen, and Almqvist (2004) found that children with moderate ID were reported to have higher internalizing and externalizing problems than children with profound ID. Good internal consistency has been found for the YSR Anxious/Depressed subscale among adolescents with mild-moderate ID (Douma, Dekker, Verhulst, & Koot, 2006). Multiple studies have yielded fair inter-rater reliability (caregiver and self-report, and between caregivers) for children and adolescents with mild ID (Douma et al., 2006; Embregts, 2000). Further psychometric properties such as criterion validity of the Anxious/Depressed subscale and

validation of the DSM-oriented anxiety disorder subscale need to be established with children and adolescents with varying levels of ID.

The Adult Behavior Checklist (ABCL) is a 118-item rating scale created to measure psychopathology in adults (Achenbach & Rescorla, 2003). Similar to the child and adolescent versions, the ABCL has eight syndrome scales including the Anxious/Depressed subscale, which is made up of 14 items. Tenneij and Koot (2007) tested the utility of this measure with individuals with ID (mean IQ of 70); reliability of the ABCL Anxious/Depressed scale was 0.89, and the inter-rater reliability for the scale was good (0.62). Those individuals with diagnosed anxiety disorders had higher scores on the ABCL Anxious/Depressed scale and Internalizing Problems than those without a diagnosed anxiety disorder.

Assessment for Dual Diagnosis (ADD; Matson & Bamburg, 1998) The ADD is a rating scale that was specifically designed to measure comorbid psychopathology in adults with mild to moderate ID. The ADD is a 79-item scale with an anxiety-specific subscale (11 items), and each item is rated on a 3-point Likert scale. The ADD was found to have high inter-rater reliability and internal consistency in a sample of adults with mild or moderate ID. Myrbakk and von Tetzchner (2008) found convergent validity between the ADD and the Developmental Assessment for Individuals with Severe Disabilities, Second Edition (DASH-II).

Developmental Behavior Checklist (DBC; Einfeld & Tonge, Gray, Brereton, Dekker, & Koot, 2002) The DBC is a 96-item measure of broad child psychopathology that contains five subscales including a 9-item anxiety subscale that is used to measure symptoms of children ages 4 and older. The measure has both a caregiver and a teacher version. Responses are based on a 3-point Likert scale, ranging from 0 (not true) to 2 (often true or very true). Although the internal consistently of the DBC-Anxiety subscale has been reported to be fair (0.62–0.67), the scale has been found to have excellent test-retest reliability with children and adolescents with borderline as well as more severe levels of ID (Einfeld & Tonge, 1995). Furthermore, inter-rater reliability between caregivers has been found to be relatively promising, while caregiver and teacher agreement has been found to be lower. Dekker et al. (2002) also found that children who were diagnosed with an anxiety disorder based on the Diagnostic Interview Schedule for Children, Fourth Edition (DISC-IV), had significantly higher scores on the DBC-Anxiety scale than those who did not receive an anxiety disorder diagnosis.

Developmental Assessment for Individuals with Severe Disabilities, Second Edition (DASH-II; Matson et al., 1997) The DASH-II is one of the only measures that has been validated with individuals with severe and profound ID. This measure is an 84-item rating scale with 13 subscales that measure a wide range of psychopathology including one subscale that measures anxiety. Each item is rated on a 3-point Likert scale for the severity, frequency, and duration of the symptom and/or behavior. Matson et al. (1997) found that all individuals with severe to profound ID who were given an anxiety disorder diagnosis exceed the cutoff on the DASH-II's anxiety subscale, but a descriptive analysis of specific symptom criteria of anxiety yielded that only observable behaviors related to anxiety could be reliably reported for this population.

Nisonger Child Behavior Rating Form (NCBRF; Aman, Tassé, Rojahn, & Hammer, 1996) The NCBRF is a 76-item broadband measure that includes a 15-item Insecure/Anxious subscale and a caregiver and teacher form. The Insecure/Anxious subscale was found to have good criterion validity, demonstrating significant differences between a group with an anxiety and/or mood disorder and group without an anxiety and/or mood disorder (Norris & Lecavalier, 2011). It has good internal consistency (0.83–0.89) among different samples of

children with varying levels of ID (Rojahn et al., 2010). Additionally, Norris and Lecavalier (2011) found that the Anxious subscale of the DBC correlated significantly with the NCBRF Insecure/Anxious subscale, although a statement about actual convergent validity cannot be made. Further, test-retest reliability has been found to be acceptable, but there have been mixed results regarding caregiver-teacher interrater reliability with some studies finding moderate correlations to some research finding no correlation between the two raters (Rojahn et al., 2010).

Psychopathology Inventory for Mentally Retarded Adults (PRIMA; Matson, Kazdin, & Senatore, 1984) The PRIMA was developed for primary use with adults with varying levels of ID. This rating scale has both a self-report and informant-report, and both versions contain a 7-item Anxiety Disorders subscale that is based on DSM criteria for anxiety disorders. The PRIMA has also been utilized as a semi-structured interview (Matson et al., 1984). Masi, Brovedani, Mucci, and Favilla (2002) examined the validity of the PRIMA in a sample of older adolescents with ID and found high convergent validity between the PRIMA-Anxiety Disorders subscale and the CBCL Anxious/Depressed subscale. Additionally, the anxiety disorders subscale of the PRIMA has also been found to be associated with the Zung Self-Rating Anxiety Scale (Ramirez & Lukenbill, 2008).

Reiss Scales for Children's Dual Diagnosis (RSCDD; Reiss & Valenti-Hein, 1994) The RSCDD is a 60-item broad-based measure that assesses for a variety of child behavior and emotional problems related to child psychopathology. It has a 5-item Anxiety Disorder subscale that has been found to have acceptable internal consistently (0.74–0.75) among a sample of children with mild-profound ID. There is limited research on the psychometric prosperities of this scale including its predictive validity.

Anxiety-Specific Rating Scales

The Anxiety, Depression, and Mood Scale (ADAMS; Esbensen et al., 2003) The ADAMS is an observation-based informant rating scale that includes 29 items with 5 subscales including Social Avoidance (7 items) and General Anxiety (7 items). Each item has a 4-point Likert response option. This scale was created to assess symptoms of anxiety and depression in individuals 10 years and older ranging from mild to profound ID. The Social Avoidance and General Anxiety subscales have been reported to have good internal consistency, and the ADAMS is reported to have good overall internal consistency (0.80; Esbensen et al., 2003; Rojahn, Rowe, Kasdan, Moore, & van Ingen, 2011). Rojahn and colleagues (2011) found good convergent and discriminant validity of the ADAMS subscales. Even so, further research of the utility of the ADAMS to measure anxiety is needed.

The Glasgow Anxiety Scale for People with an Intellectual Disability (GAS-ID; Mindham & Espie, 2003) The GAS-ID is a self-report rating scale that contains items measuring the cognitive, behavioral, and physical symptoms related to anxiety. The item responses are 3-point Likert scales that are paired with visual representations of the response options. The GAS-ID was created for adults in the mild to moderate range of ID; small sample psychometric studies have indicated good reliability and validity, including the GAS-ID discriminating between those with and without a diagnosed anxiety disorder (Mindham & Espie, 2003).

Revised Children's Manifest Anxiety Scale (RCMAS; Reynolds & Richmond, 1985) The RCMAS is a self-report measure of 37 items of different anxiety-related symptomology in typically developing youth 6–19 years of age. Only one study has examined the RCMAS in children with ID and found moderate correlations between this measure, the Fear Survey Schedule for Children With and Without Mental Retardation (FSCMR-R; Ramirez & Kratochwill, 1997).

Although available, the second edition of the RCMAS has yet to be validated with a sample of children with ID.

The Zung Self-Rating Anxiety Scale (Zung, 1971) The Zung Self-Rating Anxiety Scale is a 20-item self-report measure of anxiety for adults and has been modified to use with individuals with ID. The adapted version of this measure included yes/no responses for the items. There is limited research regarding the psychometric properties of this scale. Reliability coefficients for the yes/no item response presentation were acceptable (0.69) compared to the standard presentation, which was a nominal 4-item response scale ranging from "non or a little of the time" to "most of the time" and was found to have a very poor reliability coefficient (0.12; Lindsay & Michie, 1988). Masi et al. (2002) found low correlations between the modified Zung Self-Rating Anxiety Scale and the PRIMA-Anxiety Disorders Scale, demonstrating that the two measures may be capturing different aspects of anxiety in individuals with ID.

Fear Survey Schedule for Children With and Without Mental Retardation (FSCMR; Ramirez & Kratochwill, 1990) The FSCMR was adapted from the Fear Survey Schedule for Children-Revised (FSSCR-R; Ollendick, 1983) and is used to assess fear in individuals with ID, although originally intended to measure anxiety in those without intellectual disabilities. The self-report version has been modified for children with ID, and an informant report for research has been also created. The adaptation of these measures includes having the language of the original items simplified and supplementing the items with visual representations in order to improve understanding for those more impaired. The response choices were also adapted to include a visual scale made of facial expressions related to fear. The FSCMR has been found to have high test-retest reliability, and the FSSCR-R has been shown to have excellent internal consistency as well as high test-retest reliability (Ramirez & Kratochwill, 1990).

Fear Survey for Adults with Mental Retardation (FSAMR; Ramirez & Lukenbill, 2008) The FSAMR is a phobia-specific self-report measure of the intensity and frequency of specific fears. This measure has yes/no response style questions and is an adult version of the two child fear surveys that had already been developed. This measure was created to be used with adults with mild to moderate ID since research supports that adults with ID may have different fears than adults or children with more typical cognitive abilities (Dykens, 2000); the measure has been found to have good reliability (Ramirez & Lukenbill, 2008).

Behavioral Interviews

A common method to gain information about an individual with ID is a behavioral interview. These interviews can be structured, semi-structured, or unstructured interviews. Behavioral interviews tend to be more specialized and can be conducted with some individuals with ID, given the individual's level of cognitive functioning and communication capabilities. Caregivers and other informants are typically the main sources of information during behavioral interviews. If the individual with ID is capable of responding to interview questions, clinicians are encouraged to include them in the assessment process. The individual who is experiencing the anxiety could offer insight into their difficulties that informants would not be able to provide such as what they perceive is the source of the anxiety or what variables are influencing the manifestation of anxiety (Hagopian & Jennett, 2008). If the information gathered from the behavioral interview were mostly general (e.g., the informants are unable to provide what stimuli is causing the anxiety), the clinician may ask the informants to obtain behavior samples by observing the individual with ID in a variety of different situations. On the other hand, if the individual and/or the informants

provide more conclusive details, the interview may allow the clinician to begin to formulate a treatment plan.

Mood and Anxiety Semi-Structured Interview (MASS; Charlot, Deutsch, Hunt, Fletcher, & McLlvane, 2007) The MASS is a semi-structured interview that is used to assess whether or not the individual being assessed has displayed specific symptoms (related to *DSM-IV-TR* diagnostic criteria) within the last month and that these symptoms represent significant deviation from his or her normal functioning. When used to assess an individual with ID, specific behavioral descriptions are provided to demonstrate how the particular symptom being discussed manifests or presents in an individual with ID (Charlot et al., 2007). The MASS has been found to be more sensitive than specific for most diagnoses (including any anxiety disorder). More research needs to be done to determine the utility of the MASS as part of a dual diagnostic assessment.

Anxiety Disorders Interview Schedule for DSM-IV (ADIS-IV-C/P; Silverman & Albano, 1996) The ADIS-C/P is a semi-structured interview that probes for symptoms related to *DSM-IV* anxiety disorders, as well as other internalizing disorders (e.g., major depression) and externalizing disorders (e.g., attention-deficit/hyperactive disorder, oppositional defiant disorder). There is both a child and parent/caregiver version where the informant rates the intensity and interference of the symptoms on a 0–8-point Likert scale, with 4 being the clinical threshold. The interview can be given in its entirety, or select modules corresponding to particular disorders can be pulled and administered in isolation. There is currently no published data on the ADIS-C/P and its utility in diagnosing children who have ID with anxiety or another psychological disorder; however, it is being used with increasing frequency in those with other developmental disabilities (e.g., autism spectrum disorder; see Davis, White, & Ollendick, 2014). Generally, the caregiver interview is recommended for use with children as young as 6 years of age, and the child interview is recommended for children as young as 7; based on our experience, it can on rare occasions be done with astute children who are even younger. As a result, it may be that this interview, or at least selected modules, could be used as a rough guide to inquire about symptoms in those whose cognitive functioning approximates or exceeds a young child.

Behavioral Observations

A third method of obtaining information about an individual with ID with reported anxiety is through direct observations of behavior. Direct behavioral observations tend to be used to validate the findings from the behavioral interviews and rating scales. There are multiple approaches to direct observation such as behavioral avoidance tasks (BATs; Castagna, Lilly, & Davis, 2017; Dadds, Rapee, & Barrett, 1994) and naturalistic behavioral observations. Behavioral avoidance tasks are utilized when the stimulus that is causing the fear or anxiety is identified (Dadds et al., 1994). This method of observation is highly structured and involves progressively exposing the individual to the feared or anxiety-provoking stimulus along some dimension (e.g., distance) and recording when the individual displays the avoidance behavior (Dadds et al., 1994). Utilizing BATs with individuals with ID can provide relevant observations above and beyond what may be reported during interviews or through rating scales. If BATs are unable to be used in the assessment process, clinicians may conduct observations in naturalistic settings; functional behavioral assessments (FBAs) can be used in a variety of settings to determine the variables related to the anxiety. These observations typically require the clinician or caregiver to observe the individual's behavior and record antecedents and consequences surrounding that behavior. This data can be used to inform treatment and be utilized as a baseline assessment of functioning.

Physiological Measures

Lastly, physiological measures can be utilized to assess and measure anxiety during direct behavioral observations (Silverman & Lopez, 2004). Research has shown that objective, physiological measures of anxiety provide real-time data on how an individual is feeling; specifically, ambulatory heart rate monitors have been found to be the most useful (Kantor, Endler, Heslegrave, & Kocovski, 2001). It is important to note that there is generally limited research concerning the validity of these measures to aid in the diagnosis of anxiety disorders. To date, no research has examined the use of these measures with individuals with ID and anxiety. Furthermore, individuals with ID may have more difficulty using the equipment, tolerating the device, and enduring the procedures; the benefits of using physiological measures may not outweigh the negative responses that may occur (Hagopian & Jennett, 2008).

Future Directions

Assessing anxiety in individuals with ID continues to be challenging. Recognition of the presentation of symptoms and how to conceptualize them has fueled the need to conduct more research in this area. Although there are measures that have been utilized in the dual diagnosis assessment of anxiety with individuals who have ID, there are gaps in research on each instrument's utility in reliably and accurately measuring comorbid anxiety. Further studies need to use large sample sizes of both children/adolescents and adults with varying levels of ID to advance the psychometric data for these measures. Given the high rate of comorbid internalizing disorders in individuals with ID, clinicians who work with individuals with ID would greatly benefit from an established measure of anxiety that is appropriate for use with this population, is accurate in identifying individuals with anxiety, can be incorporated into their assessment battery, and can serve much-needed progress/treatment monitoring beyond mere initial identification.

References

Achenbach, T. M. (1991). Manual for the child behavior checklist 4–18 & 1991 profile. Burlington, VT: University of Vermont, Department of Psychiatry.

Achenbach, T. M., & Rescorla, L. (2003). ASEBA adult forms & profiles: for ages 18-59: Adult self-report and adult behavior checklist. ASEBA.

Aman, M. G., & Singh, N. N. (1991). Pharmacological interventions. In J. L. Matson & J. A. Mulick (Eds.), *Handbook of mental retardation*. New York: Pergamon Press.

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*, 41–57.

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

American Psychological Association. (2000). *Report of the task force on test user qualifications*. Washington, DC: Author.

Baker, B. L., Neece, C. L., Fenning, R. M., Crnic, K. A., & Blacher, 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.

Carlson, L. (2013). Research ethics and intellectual disability: Broadening the debates. *The Yale Journal of Biology and Medicine, 86*, 303.

Castagna, P. J., Lilly, M. E., & Davis, T. E. (2017). The behavioral avoidance task with anxious youth: A review of procedures, properties, and criticisms. *Clinical Child and Family Psychology Review, 20*, 162–184.

Charlot, L., Deutsch, C., Hunt, A., Fletcher, K., & McLlvane, W. (2007). Validation of the mood and anxiety semi-structured (MASS) interview for patients with intellectual disabilities. *Journal of Intellectual Disability Research, 51*, 821–834.

Chadwick O., Piroth N., Walker J., Bernard S. & Taylor E. (2000). Factors affecting the risk of behaviour problems in children with severe intellectual disability. *Journal of Intellectual Disability Research, 44*, 108–123.

Costello, E. J., Mustillo, S., Erkanli, A., Keeler, G., & Angold, A. (2003). Prevalence and development of psychiatric disorders in childhood and adolescence. *Archives of General Psychiatry, 60*, 837–844.

Dadds, M. R., Rapee, R. M., & Barrett, P. M. (1994). Behavioral observation. In *International handbook of phobic and anxiety disorders in children and adolescents* (pp. 349–364). Boston, MA: Springer.

Davis, T. E., III. (2012). Where to from here for ASD and anxiety? Lessons learned from child anxiety and the issue of DSM-5. *Clinical Psychology: Science and Practice, 19*, 358–363.

Davis, T. E., III, May, A. C., & Whiting, S. E. (2011). Evidence-based treatment of anxiety and phobia in

children and adolescents: Current status and effects on the emotional response. *Clinical Psychology Review, 31*, 592–602.

Davis, T. E., III, & Ollendick, T. H. (2005). Empirically supported treatments for specific phobia in children: Do efficacious treatments address the components of a phobic response? *Clinical Psychology: Science and Practice, 12*, 144–160.

Davis, T. E., III, Ollendick, T. H., & Öst, L. G. (Eds.). (2012). *Intensive one-session treatment of specific phobias*. New York, NY: Springer Science and Business Media, LLC.

Davis, T. E., III, Ollendick, T. H., & Öst, L.-G. (2019). One-session treatment of specific phobias in children: Recent developments and a systematic review. *Annual Review of Clinical Psychology, 15*, 233–256.

Davis, T. E., III, White, S. W., & Ollendick, T. H. (2014). *Handbook of autism and anxiety*. New York, NY: Springer.

De Los Reyes, A., & Kazdin, A. E. (2004). Measuring informant discrepancies in clinical child research. *Psychological Assessment, 16*, 330.

Dekker, M. C., & Koot, H. M. (2003). DSM-IV disorders in children with borderline to moderate intellectual disability: I. Prevalence and impact. *Journal of American Academy of Child and Adolescent Psychiatry, 42*, 915–922.

Dekker, M. C., Nunn, R., & Koot, H. M. (2002). Psychometric properties of the revised Developmental Behaviour Checklist scales in Dutch children with intellectual disability. *Journal of Intellectual Disability Research, 46*, 61–75.

Douma, J. C., 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*, 1224–1231.

Dykens, E. M. (2000). Psychopathology in children with intellectual disability. *Journal of Child Psychology and Psychiatry, 41*, 407–417.

Ehrenreich-May, J., & Remmes, C. S. (2013). Treatment of childhood anxiety in the context of limited cognitive functioning. In *Handbook of treating variants and complications in anxiety disorders* (pp. 149–161). New York, NY: Springer.

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*, 81–104.

Einfeld, S. L., Tonge, B. J., Gray, K. M., Brereton, A. V., Dekker, M. C., & Koot, H. M. (2002). *Manual for the Developmental Behaviour Checklist: Primary carer version (DBC-P) and teacher version (DBC-T)* (2nd ed.). Clayton, VIC: Australia Monash University.

Embregts, P. (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.

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

Emerson, E., & Hatton, C. (2007). Mental health of children and adolescents with intellectual disabilities in Britain. *British Journal of Psychiatry, 191*, 493–499.

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*, 617–629.

Essau, C. A., Lewinsohn, P. M., Lim, J. X., Moon-ho, R. H., & Rohde, P. (2018). Incidence, recurrence and comorbidity of anxiety disorders in four major developmental stages. *Journal of Affective Disorders, 228*, 248–253.

Eyman R. K., & Call T. (1977). Maladaptive behaviour and community placement of mentally retarded persons. *American Journal of Mental Deficiency, 82*, 137–144.

Gillberg C., Persson E., Grufman M. & Themner U. (1986). Psychiatric disorders in mildly and severely mentally retarded urban children and adolescents: epidemiological aspects. *British Journal of Psychiatry, 149*, 68–74.

Green, S. A., Berkovits, L. D., & Baker, B. L. (2015). Symptoms and development of anxiety in children with or without intellectual disability. *Journal of Clinical Child and Adolescent Psychology, 44*, 137–144.

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.

Hartley, S. L., & MacLean, W. E., Jr. (2006). A review of the reliability and validity of Likert-type scales for people with intellectual disability. *Journal of Intellectual Disability Research, 50*, 813–827.

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*, 861–870.

Jacobson, J. W. (1982). Problem behavior and psychiatric impairment within a developmentally disabled population I: Behavior frequency. Applied Research in Mental Retardation, 3, 121–139.

Kantor, L., Endler, N. S., Heslegrave, R. J., & Kocovski, N. L. (2001). Validating self-report measures of state and trait anxiety against a physiological measure. *Current Psychology, 20*, 207–215.

Koskentausta, T., Iivanainen, M., & Almqvist, F. (2004). CBCL in the assessment of psychopathology in Finnish children with intellectual disability. *Research in Developmental Disabilities, 25*, 341–354.

Koskentausta, T., Livanainen, M., & Almqvist, F. (2007). Risk factors for psychiatric disturbance in children with intellectual disability. *Journal of Intellectual Disability, 51*, 43–53.

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*, 485–490.

March, J. S., & Albano, A. M. (1996). Assessment of anxiety in children and adolescents. *American Psychiatric Press Review of Psychiatry, 15*, 405–427.

Masi, G., Brovedani, P., Mucci, M., & Favilla, L. (2002). Assessment of anxiety and depression in adolescents with mental retardation. *Child Psychiatry and Human Development, 32*, 227–237.

Matson, J. L., & Bamburg, J. W. (1998). Reliability of the assessment of dual diagnosis (ADD). *Research in Developmental Disabilities, 19*, 89–95.

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.

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., 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*, 39–44.

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*, 22–30.

Moss, S., Prosser, H., Ibbotson, B., & Goldberg, D. (1996). Respondent and informant accounts of psychiatric symptoms in a sample of patients with learning disability. *Journal of Intellectual Disability Research, 40*, 457–465.

Myrbakk, E., & von Tetzchner, S. (2008). Psychiatric disorders and behavior problems in people with intellectual disability. *Research in Developmental Disabilities, 29*, 316–332.

Norris, M., & Lecavalier, L. (2011). Evaluating the validity of the Nisonger child behavior rating form–parent version. *Research in Developmental Disabilities, 32*, 2894–2900.

Ollendick, T. H. (1983). Reliability and validity of the revised fear survey schedule for children (FSSC-R). *Behaviour Research and Therapy, 21*(6), 685–692.

Ollendick, T. H., Oswald, D. P., & Ollendick, D. G. (1993). Anxiety disorders in mentally retarded persons. In J. L. Matson & R. P. Barrett (Eds.), *Psychopathology in the mentally retarded* (pp. 41–85). Needham Heights, MA: Allyn & Bacon.

Ramirez, S. Z., & Kratochwill, T. R. (1990). Development of the fear survey for children with and without mental retardation. *Behavioral Assessment, 12*, 457–470.

Ramirez, S. Z., & Kratochwill, T. R. (1997). Self-reported fears in children with and without mental retardation. *Mental Retardation, 35*, 83–92.

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*, 573–580.

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.

Reiss, S., Levitan, G. W., & Szyszko, J. (1982). Emotional disturbance and mental retardation: Diagnostic overshadowing. *American Journal of Mental Deficiency, 86*, 567.

Reiss, S., & Valenti-Hein, D. (1994). Development of a psychopathology rating scale for children with mental retardation. *Journal of Consulting and Clinical Psychology, 62*, 28.

Reynolds, C. R., & Richmond, B. O. (1985). Revised children's manifest anxiety scale (RCMAS). Manual.

Rojahn, J., Rowe, E. W., Kasdan, S., Moore, L., & van Ingen, D. J. (2011). Psychometric properties of the aberrant behavior checklist, the anxiety, depression and mood scale, the assessment of dual diagnosis and the social performance survey schedule in adults with intellectual disabilities. *Research in Developmental Disabilities, 32*, 2309–2320.

Rojahn, J., Rowe, E. W., Macken, J., Gray, A., Delitta, D., Booth, A., & Kimbrell, K. (2010). Psychometric evaluation of the Behavior Problems Inventory-01 and the Nisonger child behavior rating form with children and adolescents. *Journal of Mental Health Research in Intellectual Disabilities, 3*, 28–50.

Schopler, E., Reichler, R. J., & Renner, B. R. (1988). *The childhood autism rating scale*. Los Angeles, CA: Western Psychological Services.

Schroeder, S. (1991). Self-injury and stereotypy. In J. L. Matson & J. A. Mulick (Eds.), *Handbook of mental retardation*. New York, NY: Pergamon Press.

Silverman, W. K., & Albano, A. M. (1996). Anxiety disorders interview schedule: Adis-IV child interview schedule (Vol. 2). Graywind Publications.

Silverman, W. K., & Lopez, B. (2004). Anxiety disorders. In M. Hersen (Ed.), *Psychological assessment in clinical practice: A pragmatic guide* (pp. 269–296). New York, NY: Brunner-Routledge.

Silverman, W. K., & Ollendick, T. H. (2005). Evidence-based assessment of anxiety and its disorders in children and adolescents. *Journal of Clinical Child and Adolescent Psychology, 34*, 380–411.

Sovner, R., & Hurley, A. D. (1983). Do the mentally retarded suffer from affective illness? *Archives of General Psychiatry, 40*, 61–67.

Stavrakaki, C. (1999). Depression, anxiety and adjustment disorders in people with developmental disabilities. In *Psychiatric & behavioural disorders in developmental disabilities & mental retardation* (pp. 175–187). Cambridge, UK: Cambridge University Press.

Stromme, P., & Diseth, T. H. (2000). Prevalence of psychiatric diagnosis in children with mental retardation: Data from a population-based study. *Developmental Medicine and Child Neurology, 42*, 266–270.

Suhr, J. (2015). Assessment as a decision-making process. In *Psychological assessment: A problem-solving approach* (pp. 13–32). New York, NY: The Guilford Press.

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*, 391–400.

Van Meter, A., Youngstrom, E., Youngstrom, J. K., Ollendick, T., Demeter, C., & Findling, R. L. (2014). Clinical decision making about child and adolescent anxiety disorders using the Achenbach system of empirically based assessment. *Journal of Clinical Child & Adolescent Psychology, 43*, 552–565.

Zung, W. W. (1971). A rating instrument for anxiety disorders. Psychosomatics: Journal of Consultation and Liaison Psychiatry.

14 Assessment of Major Depression in Dual Diagnosis

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Constructs of Depression

Depression is a mental health disorder with a wealth of cognitive variables involved in symptom presentation and diagnosis. Esbensen and Benson (2005) underscore the cognitive triad of hopelessness, attributions, and self-esteem. In their study 73 adults with intellectual disability (ID) were evaluated on cognitive constructs related to depressed mood. Additionally, 12 participants with ID and a diagnosis of depression were compared to 12 adults with ID and no mental health diagnosis. These authors found that the participants were able to reliably report on subjective feelings of depressive symptoms. Also, cognitive models of depression developed for the general population proved to be applicable for persons with ID also, with some modification.

Along the lines of the Esbensen and Benson (2005), Rees and Langdon (2016) found that there was a positive relationship between hopelessness and depression. With respect to self-harm, there was little relationship to this behavior and hopelessness or depression. And, while self-harm did not seem to be associated with depression, life events do seem to impact the condition (Hove, Assmus, & Havik, 2016). Specifically, loss, illness, and bullying did result in increased rates in depression. However, improved supports help moderate these three risk factors.

With respect to depression in a dual diagnosis population, gender has also been addressed as a risk factor. Lunsky (2003) found that women diagnosed with depression outnumbered males 2 to 1. He also found that persons with higher depression scores reported more loneliness and had higher levels of stress. Stress can also be associated with cyber bullying of persons with ID, resulting in increased rates of depression. Wright (2017) drew this conclusion from the assessment of 131 13–15 year olds. High levels of parental support were found to be a moderating factor and improved depressive symptoms.

Depression has also been linked to other potential problem. Hermans and Evenhuis (2013) evaluated 990 people with an average age of 50 with mild to profound ID. Based on questionnaires and medical and psychological records, depression symptoms were found to be related to anxiety. The increase in depressive symptoms was also associated with an increase in anxiety. Also, depression was correlated to chronic diseases and deficits in daily living skills.

Depression has also been linked to psychosocial stressors. Weiss, Ting, and Perry (2016) studied 141 parents of children and adolescents,

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4–18 years of age who had severe or profound ID. Factors addressed were child and family factors, quality of life, negative life events, and overall mental health of all members of the family unit. All of these stressors adversely affected depression of the child. Similarly, McGillivray and McCabe (2007) found that negative thoughts, less quality and frequent social support, and low self-esteem were all related to depression in adults with mild and moderate ID.

Marston, Perry, and Roy (1997) found that sleep problems were associated with depression. These authors evaluated 36 people with varying degrees of ID and depressive symptoms compared to 46 people with similar levels of ID, but no depression. The authors concluded that people with mild ID evinced symptoms of ID similar to the general population. However, they also concluded that people with severe and profound ID did not fit the same diagnostic profile as persons with ID but higher cognitive functioning. They hypothesized that an increase in disability symptoms becomes "behavioral equivalents" including aggression, screaming, and self-injurious behavior. This latter view however is very controversial with the majority of the researchers in the field disagreeing with the notion of behavioral equivalents. More on this point later.

Prevalence

The general consensus is that mental health issues in persons with ID are greater than what is observed in the general population. This position dates to Weaver (1946), who evaluated military personnel identified with ID. This researcher found that 44% of the males and 38% of females had some form of mental health problem. Along these same lines, Dewan (1948) found that 47% of Canadian Army members identified with ID also had a mental health issue. These participants would have largely been in the borderline to mild range of ID. Another of these early studies evaluated 444 people who lived in a center for persons with developmental disabilities (Pollock, 1944). He found that about 40% of the sample evinced psychosis. A population in such a facility would include persons in the mild to profound range of ID. However, at that time formal assessments of ID using standardized I.Q. were infrequently done. Thus, people with normal I.Q. and other issues such as speech or hearing problems may have been included as well. Other more recent studies have found similar results (Corbett, 1977; Philips & Williams, 1975).

Studies that look specifically at the prevalence of depression among persons with ID are of more recent origin. Ali, King, Strydom, and Hassiotis (2015), for example, evaluated the effects of stigma on 229 people with ID, but without a diagnosis of a mental disorder. These authors found that stigma was associated with psychological distress, specifically with respect to depression and anxiety symptoms.

Hermans, Beekman, and Evenhuis (2013) also addressed depression and anxiety. They looked at 990 older adults (over age 50) with ID. Levels of ID ranged from borderline to profound. These participants were evaluated with self and other report measures. Depression symptoms were evident in 16.8% of the sample and were most highly associated with older age. Conversely, a diagnosable major depressive disorder was present for 7.6% of the population. No relationship was noted based on level of ID, gender, or age.

Maïano et al. (2018) look at depression disorders among children and adolescents with ID. This was a review where 21 studies were assessed. Dysthymic disorder was reported in 3.4% of the sample, while 2.5% of the participants studied evinced major depressive disorder. Special issues can also contribute to the rates of depression in ID populations. Hryniewiecka-Jaworska, Foden, Kerr, Felce, and Clarke (2016) looked at 56 women with Rett syndrome. Caregivers were assessed, since persons with Rett syndrome have severe levels of ID and limited language. Based on these authors, assessment using standardized mental health measures, eight individuals (14.3%) met criteria on an affective (depression/neurosis) scale. Finally, prepartum depression and other stress-related problems were assessed in women with ID. McConnell, Mayes, and Llewellyn (2008)

conducted their study in Australia with 878 women while attending their first antenatal visit. Fifty-seven of the patients assessed met criteria for ID. They all were invited to complete two prepartum and one postpartum interview. Over a third of the persons interviewed evinced depression, anxiety, and stress.

Due to the high rates of depression in persons with ID (estimated 3–6%), continued efforts to better understand this phenomenon are needed (Cooper, 1997; Hurley, 2008). Also, these rates may indeed be low. Paykel and Priest (1992) report that depressive disorder tends to be underreported in the larger general population. This phenomenon may also exist among persons with ID and may be further compounded by poor self-awareness and inadequacies in expressing one's emotions (Levitas, Hurley, & Pary, 2001).

Patterns of Symptom Presentation

For many years the focus of research on psychopathology both within and outside the field of ID had been on core symptoms. However, as the evidence has mounted, it became prudent to look at the effects of dual diagnosis on related emotional constructs. Smith and Matson (2010), for example, compared 4 matched groups of 25 people each: ID only, epilepsy and ID, autism spectrum disorder and ID, and epilepsy, autism spectrum disorder, and ID. The latter group proved to be significantly impaired on subscales measuring other mental health problems than the ID only group or groups with ID and epilepsy or autism spectrum disorders. Areas of concern were anxiety/repetitive behavior, irritability, attention/hyperactivity, and depression.

Austin, Hunter, Gallogher, and Campbell (2018) also looked at factors that increase the likelihood of increased depression. They assessed 55 young adults with ID and 55 typically developing matched controls. Depression for both groups was related to maladaptive coping skills, while the typically developing group also had better insight into their problem, and this variable was also related to increases in depressive symptoms.

Limited coping skills along with more varied life events due to different living circumstances have been associated with depression in older persons with ID as well. Hermans and Evenhuis (2012) used self and informant report on a 28-item measure that addressed depression and anxiety with 988 people with ID who were 65 or over. Also, a psychiatric interview was given to 286 participants. All the people evaluated were in the mild to moderate range of ID. Factors associated with depression were minor physical illness, social difficulties, loss of mobility, and decreasing leisure time activities.

Earlier in this chapter, a study was reviewed focusing on aggression as a behavioral equivalent to depression. Davies and Oliver (2014) conclude that using aggression in this way is not supported by robust literature on the topic. These authors reviewed 15 papers on the topic in their efforts to address this issue. The reason for this diagnostic confusion may in large part be due to the fact that aggression and other challenging behaviors occur at high rates among persons with ID. Reiss and Rojahn (1993), for example, found that high levels of depression symptoms were evident in about four times as many aggressive individuals compared to persons with ID who were not aggressive. These authors conclude that anger may play a role in mediating between the constructs of depression and aggression.

Sturmey, Laud, Cooper, Matson, and Fodstad (2010) present one of the more forceful arguments against the notion of depression equivalents. They tested 693 institutionalized adults with severe or profound ID. The Diagnostic Assessment for the Severely Handicapped-II which has a depression and a manic subscale was administered to care workers who had provided services to a participant for at least 6 months. Depression subscale scores were compared to subscale scores on stereotypy, impulse control, and miscellaneous behavior problems and self-injurious behavior. Challenging behaviors were not associated with depression in this study. These data in large part replicate the findings of Tsiouris, Mann, Patti, and Sturmey (2003). They administered the Clinical Behavior Checklist for Persons with

Intellectual Disabilities (CBCPID) to staff familiar with each of the 92 participants, who range from mild to profound ID. The depression group was defined as a DSM-IV diagnosis of major depression, bipolar II disorder-depressed phase, or schizoaffective disorder-depressed phase and included 35 people. The 57 people in the nondepressed group included all other diagnoses. Their general conclusion was that challenging behaviors were not indicators of dysfunction or distress and thus should not be viewed as indicators of potential underlying psychopathology.

Bramston and Fogarty (2000) studied 147 people with mild or moderate ID. Their goal was to establish the relationship between anger, stress, and depression. The reader will recall that the two former constructs had been suggested as possible moderators of depression. These authors report that the overlap between all three constructs was consistently low. Data of this sort further underscore that depression is an independent construct with similar core symptoms across all levels of ID. Scott and Havercamp (2015) warn us however that where marked limitations in communication are present diagnosis may be more difficult. This issue is particularly common among persons with severe and profound ID. Identification is nonetheless very important, since depression often emerges in childhood or adolescents but persists into adulthood (Foley et al., 2015).

Diagnostic Instruments and Systems

In this section, a review of some instruments used to identify depression in persons with ID will be reviewed. Some measures are adaptations of instruments designed for the general population, while others are broad-based measure designed for and normed on an ID population where depression is one subscale. A number of these instruments have good psychometrics (Lunsky & Palucka, 2004). While not optimal, diagnostic system alone has also been used (e.g., DSM).

Interviews

Einfeld et al. (2007) describe a mental health interview using a DSM-IV checklist. Experienced psychiatrist and psychologists with expertise in dual diagnosis interviewed 52 participants suspected of having depression or psychosis. Two professionals independently assessed each participant. The mental health experts were blind to the other persons' results.

Evans, Cotton, Einfeld, and Florio (1999) also used a DSM checklist (DSM-IV-R). Two nurses rated each participant independently for depression. Clients were 89 adults with severe or profound ID.

Cooper, Smiley, Morrison, Williamson, and Allan (2007) employed individual assessment with a diagnostic system for 1023 people with ID. They used criteria from the DC-LD: Diagnostic Criteria for Psychiatric Disorders for use with Adults with Learning Disabilities/Mental Retardation. This set of criteria was established by the Royal College of Psychiatrists for use with persons who have moderate to profound ID. The authors also suggest it can be used along with the ICD-10 for persons with mild ID. Cooper et al. (2007) identified depression in about 4% of their sample. The argument for using these approaches is that they are more "in-depth" assessments and more flexible methods of evaluation. However, few studies have demonstrated psychometrics, and this approach can be used but augmented with standardized tests.

Adapting Existing Measures

Rather than start from scratch and thus develop a new measure, well-established instruments have also been employed. One of the best known of the depression measures is the Beck Depression Inventory II. Lindsay and Skene (2007) employed the scale with persons with ID. This study focused on the psychometrics of the test. Using factor analysis, three subscales were established: cognitive self, cognitive-affective loss of functioning, and somatic balance symptoms.

Meins (1993) also tested a well-established measure of depression for the general population among persons with ID. He administered the Children's Depression Inventory (CDI) to a sample of 798 adults, 19 years of age and older. Participants ranged in cognitive functioning from mild to profound. The author concludes that the CDI is a suitable measure for diagnostic screening among adults with ID and depressive symptoms. Other scales have good psychometrics for the general population. They include the Reynolds Child Depression Scale, Beck Anxiety Inventory, Automatic Thoughts Questionnaire, Cognitive Checklist (Glenn, Bihm, & Lammers, 2003), and Youth Self-Report and Child Behavior Checklist (Douma, Dekker, Verhulst, & Koot, 2006). The research that is available using this approach is limited relative to studies designed to address persons with dual diagnosis. A rundown of some of these measures follows next.

ID-Specific Measures

Psychopathology Instrument for Mentally Retarded Adults (PIMRA) The first scale specifically designed to assess for dual diagnosis was the PIMRA (Matson, Kazdin, & Senatore, 1984). This measure was based on the DSM-III criteria and has been primarily used with mild and moderate ID adults. The initial study involved 110 adults ranging in age from 18 to 71 years and in cognitive functioning from mild to severe. Factors assessed on the scale include schizophrenia disorder, affective disorder, psychosexual disorder, adjustment disorder, anxiety disorder, somatoform disorder, personality disorder, and inappropriate mental adjustment. With respect to depression, the affective disorder subfactor of the PIMRA has proven to be highly correlated with the Zung Self-Rating Depression Scale and the Beck Depression Scale. Kazdin, Matson, and Senatore (1983) conclude that this finding suggests that all four of these measures are addressing a unitary construct.

Psychopathology Checklists for Adults with Intellectual Disabilities Hove et al. (2016) assessed the relationship between depression symptoms and life traumas. They tested 593 adults with ID. Using the Psychopathology Checklists for Adults with Intellectual Disabilities, they found that increased intensity of loss, illness, and bullying exacerbated symptoms of depression.

Assessment of Dual Diagnosis (ADD) The ADD was designed to identify mental health issues in persons with mild and moderate ID. Using DSM-IV criteria for guidance, 79 items were proposed across 13 subscales. Among these subscales were depression, mania, and anxiety. The ADD covers a much broader range of mental health disorders than the PIMRA. Good internal consistency was established as well as stability across disorders and time for 101 participants (Matson & Bamburg, 1998).

Reiss Screen for Maladaptive Behavior Reiss and Rojahn (1993) evaluated 528 adults with ID. They tested people using an adult and child form. Depression and aggression were assessed and proved to be highly correlated on this scale. Later Sturmey and Bertman (1994) demonstrated good validity on this measure for measuring depression.

Anxiety, Depression, and Mood Scale Esbensen, Rojahn, Aman, and Ruedrich (2003) established the reliability and validity of the Anxiety, Depression, and Mood Scale (ADAMS). The scale consists of 55 items. Subscales include depressed mood, manic/hyperactive behavior, social avoidance, general anxiety, and obsessive/compulsive behavior. These five factors were established along with good reliability in an evaluation of 265 participants. Also, Rojahn, Rowe, Kasdan, Moore, and van Ingen (2011) in a related study tested 263 adults with mild to profound ID. They found high factor loadings on a confirmatory factor analysis. Finally, Hermans, Jelluma, van der

Pas, and Evenhuis (2012) evaluated the psychometrics of a Dutch version of the scale. A total of 1050 adults over 50 years of age who experienced an ID were studied. Based on their results, the authors concluded that the Dutch version of the ADAMS was reliable and valid to screen for anxiety and depression in this population.

Diagnostic Assessment for the Severely Handicapped-II (DASH-II) Matson, Gardner, Coe, and Sovner (1991) describe a scale to measure psychopathology in persons with severe and profound ID. Keyed to DSM-IV they report norms, reliability, and cutoff scores. Subscales for the DASH-II include self-injurious behavior, stereotypies and tics, anxiety, depression/mood, pervasive developmental disorders, eating disorders, organic syndromes, schizophrenia, sleep disorders, sexual disorders, elimination disorders, mania, and impulse control and other miscellaneous behaviors. Reliability data were also found to be good in a study by Sevin, Matson, Williams, and Kirkpatrick-Sanchez (1995).

Matson et al. (1999) looked specifically at the depression subscale. Their sample consisted of 57 people with severe or profound ID who ranged in age from 22 to 79 years of age. Using DSM-IV criteria, licensed psychiatrists diagnosed 18 people with major depression, 19 with autism, and 20 with no DSM-IV diagnosis. All participants in the three groups were assessed by a professional (psychologist or Qualified Mental Retardation Professional) on the DASH-II. The symptoms of depression most commonly identified were nonverbal behaviors such as disruptions in sleep and eating, psychomotor problems, and irritability. It was concluded that the DASH-II was a valid indicator of depression. The authors also found that the most common comorbidities were mania and impulse control. In another paper using the DASH-II, 693 adults with severe or profound ID were assessed for depression symptoms and for behavioral equivalents of depression (Sturmey et al., 2010). Items on the DASH-II subscale were compared to 15 additional depression items not on the DASH-II and subscales from the DASH-II that had items described as behavioral equivalents in the literature. These three subscales were stereotypy, self-injury, and impulse control and miscellaneous. The two depression measures correlated strongly with each other but not with the three subscales of the DASH-II designated as behavioral equivalents. In a related study, the irritability and hyperactivity subscales of the Aberrant Behavior Checklist (ABC) correlated highly with the depression subscale of the DASH-II, while facts of the ABC such as sleep disorders and impulse control had much weaker correlations.

Mood, Interest and Pleasure Questionnaire (MIPQ) The initial psychometrics of the MIPQ were reported by Ross and Oliver (2003). This measure is a 25-item questionnaire and has two subscales, mood, and interest and pleasure. They tested 53 people across the range of ID. On the self-report, 25 individuals with severe or profound ID were unable to complete the measure. The authors report good reliability on the inter-rater and test-retest reliability.

Hayes, McGuire, O'Neill, Oliver, and Morrison (2011) also describe a study using the MIPQ. Direct care staff for 52 individuals with severe or profound ID filled out scales that addressed depression, communication, and challenging behaviors. They concluded that the MIPQ was able to detect low mood in the population studied.

Clinical Behavior Checklist for Persons with Intellectual Disabilities (CBCPID) The CBCPID was administered to 57 people without depression and 35 individuals diagnosed with depression. All of the participants had ID which ranged from mild to profound. This scale consisted of 30 items adopted from the depression criteria of the ICD-10 and Holmes, Shah, and Wing's (1982) Disability Assessment Schedule.

The authors of this study found that depressive equivalents such as depression had no support and that core symptoms of depression as reflected in the DSM should be the focus for diagnosis (Tsiouris et al., 2003).

Marston 30 Symptoms Checklist The Marston 30 Symptoms Checklist has been used to detect depression in persons with severe and profound ID (Tsiouris, 2001). Twenty-two people with bipolar I, bipolar II, or major depression were selected from a larger sample (n = 150). A psychiatrist using DSM-III-R criteria, history, and observations made the diagnosis. This scale was helpful in detecting depression.

Brief Symptoms Inventory (BSI) Wieland, Wardenaar, Fontein, and Zitman (2012) normed the BSI on people with borderline and mild ID. The study was conducted in the Netherlands, and the authors concluded that the BSI was useful. They report high internal consistency. Also, this measure proved helpful in discriminating between depression, anxiety, and phobic anxiety.

Self-Report Depression Questionnaire (SRDQ) This measure is a 32-item test which maps on to the DSM-IV. The scale was designed to measure depression in persons with mild or moderate ID (Esbensen, Seltzer, Greenberg, & Benson, 2005). They found good to excellent reliability. Also, strong convergent, discriminant, and predictive validity were noted.

Glasgow Depression Scale Cuthill, Espie, and Cooper (2003) initially studied a focus group of 12 adults with mild or moderate ID. The purpose of the evaluation was to determine how these persons described affect. Items most commonly used to describe depression were included in the scales items. Criteria for scoring items was a three-point Likert scale. The measure was then piloted on six persons with ID, and the original 28 items were paired down to 20. Using the DSM-IV criteria for depression, the experimental sample was divided into three groups: ID plus depression, ID only, and depression only. The scale proved to have good reliability both internally and via test-retest. Additionally, good content, discriminant, and criterion validity were reported.

Overview

The purpose of this chapter was to provide a summary of issues in the assessment of depression among persons with ID. At present there are a number of measures available for self-report and especially other report. Major test companies for the most part have not gotten involved in test development with respect to depression among persons with ID. As a result, test development has been sporadic leading those who have looked at the area as a whole, to be promising but in need of further development (Hermans & Evenhuis, 2010). These authors single out the Glasgow Depression Scale for self-report and Assessment for Dual Diagnosis, Reiss Screen for Maladaptive Behavior, and Children's Depression Inventory for informant report as particularly noteworthy. Their paper does not take into account persons with severe and profound ID. Additionally, a mature and robust set of depression scales may still be a ways off, given the hit and miss nature of the research. Other reviews the reader may wish to consult include McBrien's (2003) overview of depression measures for people with ID and Cassidy, Bradley, Bowen, Wigham, and Rodgers' (2018) review of depression measures for adults with autism. Given the high rate of ID in persons with autism, the paper should be of interest to the readers of this chapter.

An important aspect of assessment and differential diagnosis of depression among persons with ID is how this information can be best linked to intervention (Cooper & Collacott, 1996). While the need to identify symptom profiles that might benefit most from specific treatments has been recognized for decades, little has been done to advance knowledge on this critically important topic. Thus, moving forward, along with growing

the depth and maturity of the assessment methods themselves, linking them to interventions should be a top priority.

References

Ali, A., King, M., Strydom, A., & Hassiotis, A. (2015). Self-reported stigma and symptoms of anxiety and depression in people with intellectual disabilities: Findings from a cross sectional study in England. *Journal of Affective Disorders, 187*, 224–231.

Austin, K. L., Hunter, M., Gallogher, E., & Campbell, L. E. (2018). Depression and anxiety symptoms during the transition to early adulthood for people with intellectual disabilities. *Journal of Intellectual Disability Research, 62*, 407–421.

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*, 487–500.

Cassidy, S. A., Bradley, L., Bowen, E., Wigham, S., & Rodgers, J. (2018). Measurement properties of tools used to assess depression in adults with and without autism spectrum conditions: A systematic review. *Autism Research, 11*, 738–754.

Cooper, S. A. (1997). Epidemiology of psychiatric disorders in elderly compared with younger adults with learning disabilities. *The British Journal of Psychiatry, 170*(4), 375–380.

Cooper, S. A., & Collacott, R. A. (1996). Depressive episodes in adults with learning disabilities. *Irish Journal of Psychological Medicine, 13*, 105–113.

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*, 27–35.

Corbett, J. A. (1977). Mental retardation- psychiatric aspects. In M. Rutter & L. Hersov (Eds.), *Child psychiatry: Modern approaches*. Oxford, UK: Blackwell Scientific Publications.

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. *The British Journal of Psychiatry: the Journal of Mental Science, 182*, 347–353.

Davies, L. E., & Oliver, C. (2014). The purported association between depression, aggression, and self-injury in people with intellectual disability: A critical review of the literature. *American Journal on Intellectual and Developmental Disabilities, 119*, 452–471.

Dewan, J. G. (1948). Intelligence and emotional stability. *The American Journal of Psychiatry, 104*, 548–554.

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 and Adolescent Psychiatry, 45*, 1224–1231.

Einfeld, S., Tonge, B., Chapman, L., Mohr, C., Taffe, J., & Horstead, S. (2007). Inter-rater reliability of the diagnoses of psychosis and depression in individuals with intellectual disabilities. *Journal of Applied Research in Intellectual Disabilities, 20*, 384–390.

Esbensen, A. J., & Benson, B. A. (2005). Cognitive variables and depressed mood in adults with intellectual disability. *Journal of Intellectual Disability Research, 49*, 481–489.

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*, 617–629.

Esbensen, A. J., Seltzer, M. M., Greenberg, J. S., & Benson, B. A. (2005). Psychometric evaluation of a self-report measure of depression for individuals with mental retardation. *American Journal of Mental Retardation, 110*, 469–481.

Evans, K. M., Cotton, M. M., Einfeld, S. L., & Florio, T. (1999). Assessment of depression in adults with severe or profound intellectual disability. *Journal of Intellectual and Developmental Disability, 24*, 147–160.

Foley, K. R., Bourke, J., Einfeld, S. L., Tonge, B. J., Jacoby, P., & Leonard, H. (2015). Patterns of depressive symptoms and social relating behaviors differ over time from other behavioral domains for young people with down syndrome. *Medicine, 94*, e710.

Glenn, E., Bihm, E. M., & Lammers, W. J. (2003). Depression, anxiety, and relevant cognitions in persons with mental retardation. *Journal of Autism and Developmental Disorders, 33*, 69–76.

Hayes, S., McGuire, B., O'Neill, M., Oliver, C., & Morrison, T. (2011). Low mood and challenging behaviour in people with severe and profound intellectual disabilities. *Journal of Intellectual Disability Research, 55*, 182–189.

Hermans, H., Beekman, A. T. F., & Evenhuis, H. M. (2013). Prevalence of depression and anxiety in older users of formal Dutch intellectual disability services. *Journal of Affective Disorders, 144*, 94–100.

Hermans, H., & Evenhuis, H. M. (2010). Characteristics of instruments screening for depression in adults with intellectual disabilities: Systematic review. *Research in Developmental Disabilities, 31*, 1109–1120.

Hermans, H., & Evenhuis, H. M. (2012). Life events and their associations with depression and anxiety in older people with intellectual disabilities: Results of the HA-ID study. *Journal of Affective Disorders, 138*, 79–85.

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*, 691–699.

Hermans, H., Jelluma, N., van der Pas, F. H., & Evenhuis, H. M. (2012). Feasibility, reliability and validity of the

Dutch translation of the anxiety, depression and mood scale in older adults with intellectual disabilities. *Research in Developmental Disabilities, 33*, 315–323.

Holmes, N., Shah, A., & Wing, L. (1982). The disability assessment schedule: A brief screening device for use with the mentally retarded. *Psychological Medicine, 12*, 879–890.

Hove, O., Assmus, J., & Havik, O. E. (2016). Type and intensity of negative life events are associated with depression in adults with intellectual disabilities. *American Journal on Intellectual and Developmental Disabilities, 121*, 419–431.

Hryniewiecka-Jaworska, A., Foden, E., Kerr, M., Felce, D., & Clarke, A. (2016). Prevalence and associated features of depression in women with Rett syndrome. *Journal of Intellectual Disability Research, 60*, 564–570.

Hurley, A. D. (2008). Depression in adults with intellectual disability: Symptoms and challenging behaviour. *Journal of Intellectual Disability Research, 52*, 905–916.

Kazdin, A. E., Matson, J. L., & Senatore, V. (1983). Assessment of depression in mentally retarded adults. *The American Journal of Psychiatry, 140*, 1040–1043.

Levitas, A. S., Hurley, A. D., & Pary, R. (2001). The mental status examination in patients with mental retardation and developmental disabilities. *Mental Health Aspects of Developmental Disabilities, 4*, 2–16.

Lindsay, W. R., & Skene, D. D. (2007). The Beck depression inventory II and the Beck anxiety inventory in people with intellectual disabilities: Factor analyses and group data. *Journal of Applied Research in Intellectual Disabilities, 20*, 401–408.

Lunsky, Y. (2003). Depressive symptoms in intellectual disability: Does gender play a role? *Journal of Intellectual Disability Research, 47*, 417–427.

Lunsky, Y., & Palucka, A. M. (2004). Depression in intellectual disability. *Current Opinion in Psychiatry, 17*, 359–363.

Maïano, C., Coutu, S., Tracey, D., Bouchard, S., Lepage, G., Morin, A. J. S., & Moullec, G. (2018). Prevalence of anxiety and depressive disorders among youth with intellectual disabilities: A systematic review and meta-analysis. *Journal of Affective Disorders, 236*, 230–242.

Marston, G. M., Perry, D. W., & Roy, A. (1997). Manifestations of depression in people with intellectual disability. *Journal of Intellectual Disability Research, 41*(6), 476.

Matson, J. L., & Bamburg, J. W. (1998). Reliability of the Assessment of Dual Diagnosis (ADD). *Research in Developmental Disabilities, 19*, 89–95.

Matson, J. L., Gardner, W. I., 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. *The British Journal of Psychiatry, 159*, 404–409.

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., Rush, K. S., Hamilton, M., Anderson, S. J., Bamburg, J. W., Baglio, C. S., … Kirkpatrick-Sanchez, S. (1999). Characteristics of depression as assessed by the diagnostic assessment for the severely handicapped-II (DASH-II). *Research in Developmental Disabilities, 20*, 305–313.

McBrien, J. A. (2003). Assessment and diagnosis of depression in people with intellectual disability. *Journal of Intellectual Disability Research, 47*, 1–13.

McConnell, D., Mayes, R., & Llewellyn, G. (2008). Prepartum distress in women with intellectual disabilities. *Journal of Intellectual & Developmental Disability, 33*, 177–183.

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.

Meins, W. (1993). Assessment of depression in mentally retarded adults: Reliability and validity of the Children's Depression Inventory (CDI). *Research in Developmental Disabilities, 14*, 299–312.

Paykel, E. S., & Priest, R. G. (1992). Recognition and management of depression in general practice: Consensus statement. *British Medical Journal, 305*, 1198–1202.

Philips, I., & Williams, N. (1975). Psychopathology and mental retardation: A study of 100 mentally retarded children: I. Psychopathology. *The American Journal of Psychiatry, 132*, 1265–1271.

Pollock, H. M. (1944). Mental disease among mental defectives. *American Journal of Psychiatry, 101*, 361–363.

Rees, J., & Langdon, P. E. (2016). The relationship between problem-solving ability and self-harm amongst people with mild intellectual disabilities. *Journal of Applied Research in Intellectual Disabilities, 29*, 387–393.

Reiss, S., & Rojahn, J. (1993). Joint occurrence of depression and aggression in children and adults with mental retardation. *Journal of Intellectual Disability Research, 37*, 287–294.

Rojahn, J., Rowe, E. W., Kasdan, S., Moore, L., & van Ingen, D. J. (2011). Psychometric properties of the "aberrant behavior checklist," the "anxiety, depression and mood scale," the "assessment of dual diagnosis" and the "social performance survey schedule" in adults with intellectual disabilities. *Research in Developmental Disabilities: A Multidisciplinary Journal, 32*, 2309–2320.

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, 42*, 81–93.

Scott, H., & Havercamp, S. M. (2015). The diagnosis of depression in people with severe limitations in intellectual functioning. *Journal of Mental Health Research in Intellectual Disabilities, 8*, 168–185.

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). *The British Journal of Clinical Psychology, 34*, 93–94.

Smith, K. R. M., & Matson, J. L. (2010). Psychopathology: Differences among adults with intellectually disabled, comorbid autism spectrum disorders and epilepsy. *Research in Developmental Disabilities, 31*, 743–749.

Sturmey, P., & Bertman, L. J. (1994). Validity of the Reiss screen for maladaptive behavior. *American Journal of Mental Retardation, 99*, 201–206.

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*, 1002–1007.

Tsiouris, J. A. (2001). Diagnosis of depression in people with severe/profound intellectual disability. *Journal of Intellectual Disability Research, 45*, 115–120.

Tsiouris, J. A., Mann, R., Patti, P. J., & Sturmey, P. (2003). Challenging behaviours should not be considered as depressive equivalents in individuals with intellectual disability. *Journal of Intellectual Disability Research, 47*, 14–21.

Weaver, T. R. (1946). The incident of maladjustment among mental defectives in military environment. *American Journal of Mental Deficiency, 51*, 238–246.

Weiss, J. A., Ting, V., & Perry, A. (2016). Psychosocial correlates of psychiatric diagnoses and maladaptive behaviour in youth with severe developmental disability. *Journal of Intellectual Disability Research, 60*, 583–593.

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*, 843–853.

Wright, M. F. (2017). Cyber victimization and depression among adolescents with intellectual disabilities and developmental disorders: The moderation of perceived social support. *Journal of Mental Health Research in Intellectual Disabilities, 10*, 126–143.

Severe Psychopathology

15

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Introduction

In the general population, schizophrenia and bipolar disorder are considered the most costly and arguably the most debilitating of all of the major psychological conditions (Goeree et al., 2005; Wu et al., 2005). Both conditions have high rates of personal, social, and occupational disability. The cost of treatment usually entails a lifetime of medication management, case management, and psychosocial therapies. Sadly, many have recurrent inpatient treatments to manage their symptom episodes. Even when persons with schizophrenia and bipolar disorder receive optimal treatments, many continue to experience substantial residual symptoms and impairments throughout most of their lives. For persons with dual diagnoses (co-occurring intellectual and psychiatric conditions), the social and occupational consequences are often more severe (Knapp, Mangalore, & Simon, 2004; Meadows et al., 1991). The cost of inpatient treatments for persons with dual diagnosis is often double compared to the general population (Lai, Hung, Lin, Chien, & Lin, 2011). One of the major challenges in working with dual diagnosis individuals is conducting a proper assessment for schizophrenia and/or bipolar disorder. Difficulties in language, insight, attention, and memory are barriers to a thorough diagnostic assessment, which makes many of the common measures ineffective. This chapter discusses relevant issues in the assessment of persons with intellectual deficits and symptoms of schizophrenia and bipolar disorder. Recommendations on useful scales and measures will be highlighted. First, we will examine how common these disorders are in the general population, diagnostic criteria for each disorder, and then how these are displayed in persons with dual diagnoses.

Prevalence Rates and Risk Factors

Similarly, across a number of population studies, the rate of schizophrenia and bipolar disorder is around 1% of the general population (as reviewed in Combs, Mueser, Morales, & Smith, 2018; Johnson & Miklowitz, 2018; Merikangas et al., 2012). In persons with intellectual disability, there is an increased risk of developing psychological conditions such as schizophrenia and bipolar disorders (Turner, 1989). Specifically, among persons with intellectual disability, between 15% and 40% show signs or symptoms of schizophrenia and/or bipolar disorder over their lifespan (Helps, 2015). Among this population, about 3.8% meet DSM diagnostic criteria

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for schizophrenia and 5.7% for bipolar disorder (Kendall & Owen, 2015). For both conditions, the presence of poverty, brain abnormalities, chronic stress, trauma and neglect, and lack of coping resources (mental health, educational, etc.) are believed to be major factors in the elevated risk for developing both of these conditions. Accurate statistics on psychosis in the intellectual disability population are scarce and have been clouded by the overlap between autism spectrum disorder, which is often interpreted as psychosis. Mood symptoms and especially mania are often misinterpreted as behavioral disorders related to intellectual disability. The presence of intellectual disability is often identified at birth or in early childhood, while the assessment of psychosis and bipolar often occurs between the ages of 15 and 24 in most cases. This time delay often leads to encompassing psychiatric symptoms as part of the intellectual disability condition.

General Symptoms of Schizophrenia and Bipolar Disorder

The diagnostic criteria for schizophrenia are similar across a variety of different diagnostic systems. In general, the diagnostic criteria specify some degree of impairment in work, social, or self-care, combined with psychotic lasting a significant duration (e.g., 6 months or more). According to the *DSM-5* (APA, 2013) a diagnosis of schizophrenia must include the presence of two or more of the following five symptoms: delusions, hallucinations, disorganized speech, grossly disorganized behavior, or negative symptoms. One of the symptoms must be delusions, hallucinations, or disorganized speech. The symptoms must have been present at least for a month, unless successfully treated.

The diagnosis of schizophrenia requires a clinical interview with the patient, a thorough review of all available records, and standard medical evaluations to rule out the possible biological factors. In addition, because many persons with schizophrenia are poor historians or may not provide accurate accounts of their behavior, information from significant others, such as family members, is often critical to establish a diagnosis of schizophrenia. The use of family and other informants is especially important in the assessment of prodromal or pre-psychotic states.

The diagnostic criteria for bipolar disorder (previously called manic depression) regardless of type consist of symptoms of mania or hypomania (i.e., elevated, expansive mood, and/or irritable mood) along with episodes of major depression. The presence of mania or hypomania symptoms was found to occur in about 25–30% of community-based samples (Johnson & Miklowitz, 2018). The DSM-5 describes two types of bipolar disorder. Bipolar type 1, the most severe, consists of manic and major depressive episodes with significant impairment in social and occupational functioning. Bipolar disorder, type 1 is often associated with inpatient treatment to control mood episodes and increased goal-directed activities and behaviors. Bipolar disorder, type 2 consists of hypomanic episodes with recurrent episodes of major depression. Persons with bipolar disorder, type 2 can often work and function socially and may not need inpatient treatment. Manic episodes last for 1 week or more while hypomanic episodes last at least 4 days. Significant irritability can be part of bipolar disorder, but the person must meet more symptoms for this to apply. Having one manic episode over the person's lifetime is sufficient to diagnose the person with bipolar disorder type 1, and it should be noted that current DSM-5 criteria do not require an episode of depression for this diagnosis. Associated symptoms of mania are increased self-esteem, decreased need for sleep, changes in appetite, hyperactivity, distractibility, rapid speech, flight of ideas, and reckless, impulsive behaviors. Major depressive episodes can last from 2 weeks to several months. Associated symptoms of depression include sadness, lack of interest in activities, psychomotor slowing, sleep problems, fatigue, changes in weight and/or appetite, guilt, poor concentration, and suicidal ideation.

Symptoms in Persons with Intellectual Disability

As mentioned previously, it is more difficult to derive a diagnosis in persons with intellectual disability due to the cognitive and language impairments often found in the condition (Kendall & Owen, 2015). Persons with mild intellectual disability who can verbalize some of their internal experiences can be assessed using common diagnostic interviews such as the SCID (Ryan, 1994). However, for persons with moderate to severe levels of intellectual disability, it is best to carefully document and observe their behaviors. A common diagnostic problem is determining what behaviors are part of the intellectual disability condition and what behaviors are part of the psychosis or bipolar condition. A study by Cherry, Penn, Matson, and Bamburg (2000) found that in persons with intellectual disability and schizophrenia, the psychotic symptoms exhibited were mainly behavioral disorganization, reality distortion, and there was less evidence for negative symptoms. Behaviorally, signs of hallucinations may include frequent staring, talking, or gesturing to people who are not present, covering their eyes and ears, brushing of their body (tactile), and facial grimaces when eating (gustatory). Of course, these may also be symptoms of autism spectrum disorder in the form of self-stimulation behaviors as well. Delusions such as paranoia may not be easily verbalized but may appear as hiding from certain people, avoidance of some situations, and odd emotional reactions to the presence of others. Disorganization may appear as wearing multiple layers of clothing, odd behaviors exhibited with no clear purpose, pacing, and poor grooming and hygiene. One of the most important issues to address is the presence of comorbid medical conditions in the intellectual disability population, as conditions such as partial complex seizures can appear similar to schizophrenia. For bipolar disorders, research and clinical observation has identified several key behaviors to note. These include sleep disturbance, agitation, and increased levels of activity. Again, the presence of intact language may facilitate a discussion of mood and behaviors, but this may not be possible in all cases.

General Assessment Measures

For persons with mild levels of intellectual disability who have intact language function, clinicians can use many of the common symptom and diagnostic measures. For persons with intellectual disability, it is more useful to do interview methods than to use paper and pencil tests. Most of these cover a specific period of time (e.g., last 2 weeks or last month) and ask standardized questions about symptom severity and presence. The Structured Clinical Interview for DSM-5 (SCID) is based on DSM-5 criteria and is the most useful in terms of diagnosing schizophrenia. To assess severity of psychotic symptoms, measures such as the Positive and Negative Syndrome Scale (PANNS; Kay, Fiszbein, & Opler, 1987), the Brief Psychiatric Rating Scale (BPRS; Overall & Gorham, 1962), and the Psychotic Rating Scale (PSYRATS; Haddock, McCarron, Tarrier, & Faragher, 1999) have good psychometric properties (As reviewed in Combs et al., 2018). Scales specific to positive (Scale for the Assessment of Positive Symptoms; Andreasen & Olsen, 1982) and negative symptoms (Scale for the Assessment of Negative Symptoms; Andreasen, 1982) can be used for a more in-depth assessment of these areas. Finally, the assessment of social skills and social/community functioning are important areas to examine in persons with schizophrenia. The Maryland Assessment of Social Competence (MASC; Bellack & Thomas-Lohrman, 2003) and the Social Functioning Scale (Birchwood, Smith, Cochrane, Wetton, & Copstake, 1990) and UCSD Performance-Based Skills Assessment (UPSA; Patterson, Goldman, McKibbin, Hughs, & Jeste, 2001) are widely used measures of social skills and community functioning.

For the assessment of bipolar disorders, the use of a structured clinical interview is the most reliable and valid method to examine this condition (Miller, Johnson, & Eisner, 2009). The SCID is the most common measure due to its link to the

current DSM-5 criteria. The Schedule for Affective Disorders and Schizophrenia (SADS; Endicott & Spitzer, 1978) has been used in many studies, but is based on the older Research Diagnostic Criteria (RDC) and may be less useful today. Once the clinical interview is conducted, there are several self-report scales that are commonly used to assess the manic and depression phases of the condition. Self-report instruments such as the General Behavior Inventory (Depue et al., 1981), Mood Disorder Questionnaire (MDQ; Hirschfeld et al., 2000), Altman Self-Rating Mania Scale (Altman, Hedeker, Peterson, & Davis, 1997), and the Brief Symptom Inventory (Derogatis & Melisaratos, 1983) may be useful. It may be more important to focus on the presence of mania and hypomania than depression to properly diagnose bipolar conditions. Clearly, the reliance on self-report measures is less than ideal when dealing with persons with intellectual disability as these persons may have problems completing the rating scales. We will now review several scales for the assessment of schizophrenia and bipolar that were specifically designed for persons with intellectual disability.

Assessment Measures Specific to Persons with Intellectual Disability and Schizophrenia

In general, there is a need to develop valid and useful assessment measures for persons with mental health conditions and intellectual disability (Beail, Mitchell, Vlissides, & Jackson, 2015). These measures are often behavioral in nature and can be completed using simple rating scales or with information from a knowledgeable informant.

Diagnostic Assessment for the Severely Handicapped-Revised (DASH-II)

The DASH-II (Matson, 1995) is an informant-based measure of psychopathology in people with severe and profound intellectual disabilities. This 84-item measure consists of 13 subscales: (1) Anxiety, (2) Depression, (3) Mania, (4) PDD/Autism, (5) Schizophrenia, (6) Stereotypies, (7) Self-Injury, (8) Elimination, (9) Eating, (10) Sleeping, (11) Sexual, (12) Organic, and (13) Impulse Control. The Schizophrenia subscale includes seven items focusing on behaviors consistent with psychotic disorders: (1) mood unrelated to surroundings; (2) talking to imaginary people or inanimate objects; (3) speech making no sense; (4) hearing things that are imaginary; (5) standing or sitting in bizarre positions; (6) experiencing touch or other sensations on the skin that are imaginary; and (7) seeing things that are imaginary (Bamburg, Cherry, Matson, & Penn, 2001). Each item on the DASH-II is scored as a 0, 1, or 2, and there are ratings for frequency over the previous 2 weeks, duration, and severity of behavioral symptoms (Bamburg et al., 2001; Thorson, Matson, Rojahn, & Dixon, 2008). Caretakers familiar with the individual rate each item. Previous studies have shown good levels of reliability and validity with the intellectual disability population (Bamburg et al., 2001; Matson & Malone, 2006; 1998; Myrbakk & Von Tetzchner, 2008a, 2008b; Paclawskyj, Matson, Bamburg, & Baglio, 1997; Sturmey, Matson, & Lott, 2004), and with intellectual disability and schizophrenia comorbidity (Bamburg et al., 2001; Cherry et al., 2000). Limitations of the DASH-II include that it is derived from the DSM-IV-R and is useful primarily with the severe and profound intellectual disability population. Another limitation is that some items require verbal information and nonverbal individuals' answers are scored as "0" (Thorson et al., 2008). Further, the standardization sample is limited and administration requires a trained interviewer (Aman, 1991; Mohr, Tonge, & Einfeld, 2005).

Assessment of Dual Diagnosis (ADD)

The ADD (Matson & Bamburg, 1998) is an informant-based measure of psychopathology in people with mild and moderate intellectual

disabilities. The ADD is a 79-item psychopathology screening instrument representing 13 diagnostic categories, which include (1) Mania, (2) Depression, (3) Anxiety, (4) PTSD, (5) Substance abuse, (6) Somatoform disorders, (7) Dementia, (8) Conduct disorder, (9) Pervasive developmental disorder, (10) Schizophrenia, (11) Personality disorders, (12) Eating disorders, and (13) Sexual disorders. Items are scored as a 0, 1, or 2 with ratings for frequency, duration, and severity of symptoms (Belva & Matson, 2015). Similar to the DASH-II, caretakers familiar with the individual rate each item. Previous studies have shown good reliability and validity with this population (Matson & Bamburg, 1998; Rojahn, Rowe, Kasdan, Moore, & Van Ingen, 2011). The ADD has been found to have high convergent validity with other psychopathology rating scales, specifically the Mini PAS-ADD, DASH-II, and RSMB (Myrbakk & Von Tetzchner, 2008a, 2008b). However, according to Matson, Belva, Hattier, and Matson (2012), research with the ADD is limited compared to the other measures of psychopathology in the population with intellectual disability and additional research is necessary. Rojahn et al. (2011) found adequate concurrent validity for the schizophrenia subscale compared with the ABC, but additional research is needed to further support the reliability and validity of the Schizophrenia subscale.

Psychopathology Instrument for Adults with Mental Retardation (PIMRA)

The PIMRA (Matson, 1988) is an informant-based measure of psychopathology in individuals with mild, moderate, or severe intellectual disabilities. The PIMRA was the first scale designed to assess dual diagnosis and is currently used as a screening instrument to aid in differential diagnosis of individuals with intellectual disability. Caretakers familiar with the individual rate each item and respond using a "yes" or "no" format. The PIMRA contains 56 items representing 7 classes of psychopathology based on DSM-III criteria: (1) Schizophrenia, (2) Affective disorder, (3) Psychosexual disorder, (4) Adjustment disorder, (5) Anxiety disorder, (6) Somatoform disorder, (7) Personality disorder, and one additional subscale measuring inappropriate adjustment. Studies have shown good levels of reliability and validity for the PIMRA (Balboni, Battagliese, & Pedrabissi, 2000; Belva & Matson, 2015; Gustafsson & Sonnander, 2005; Iverson & Fox, 1989; La Malfa, Notarelli, Hardoy, Bertelli, & Cabras, 1997; Masi, Brovedani, Mucci, & Favilla, 2002; Matson, Kazdin, & Senatore, 1984; Van Minnen, Savelsberg, & Hoogduin, 1994; Watson, Aman, & Singh, 1988), and the Schizophrenia subscale (Linaker & Helle, 1994; Swiezy, Matson, Kirkpatrick-Sanchez, & Williams, 1995). Limitations include that the PIMRA is based on DSM-III criteria and it contains fewer subscales compared to other measures of psychopathology. Along with the DASH-II, the standardization sample is limited and administration requires a trained interviewer (Aman, 1991; Mohr et al., 2005).

Psychiatric Assessment Schedule for Adults with Developmental Disability (PAS-ADD)

The PAS-ADD (Moss et al., 1993) is a screening measure to assess psychopathology in people with mild, moderate, or severe intellectual disabilities. There are three versions of the PAS-ADD which can be used depending on the clinical situation. The PAS-ADD checklist is often used as a screening tool, the PAS-ADD semi-structured clinical interview is used to derive a diagnosis, and the Mini PAS-ADD is used in situations requiring a briefer assessment. This family of scales contains 29 items encompassing 7 broad areas: (1) appetite and sleep, (2) tension and worry, (3) phobias and panics, (4) depression and hypomania, (5) obsessions and compulsions, (6) psychoses, and (7) autism (Moss et al., 1998). The PAS-ADD checklist and the Mini PAS-ADD are rated by a caretaker familiar with the individual, and the PAS-ADD semi-structured clinical interview utilizes parallel interviewing of client and an informant. It is designed to provide a

diagnosis under both ICD-10 and the DSM-IV-TR. Items are rated on a four-point Likert scale; if the symptom is not present a "1" is given, if the symptom is present to a mild degree a "2" is given, if the symptom is present to a moderate degree or severe for less than half the rating period a "3" is given, and if the symptom is severe for more than half the rating period a "4" is given. The Mini PAS-ADD is designed to be used by staff who do not have a background in psychiatry or psychology. The PAS-ADD was first described by Moss et al. (1993) and since then, research has demonstrated reliability and validity for the PAS-ADD (Beail et al., 2015; Moss et al., 1998; Sturmey, Newton, Cowley, Bouras, & Holt, 2005; Zeilinger, Weber, & Haveman, 2011), including the psychosis subscale (Cooper et al., 2007; Moss, Prosser, Ibbotson, & Goldberg, 1996) and the Mini PAS-ADD (Deb, Thomas, & Bright, 2001; Janssen & Maes, 2013; Myrbakk & Von Tetzchner, 2008a, 2008b; Prosser et al., 1998). According to Matson et al. (2012), the PAS-ADD has a large amount of supporting research and is the measure of choice for psychopathology in individuals with intellectual disability in the United Kingdom. However, has discussed issues with the PAS-ADD, such as (1) the sensitivity and specificity of the checklists and rating scales are largely unknown, (2) the diagnostic accuracy is unknown, and (3) standardization sample is mostly inadequate. Further, according to Mohr et al. (2005) there is disagreement in how to compute and apply cutoff scores for diagnosis.

Reiss Screen for Maladaptive Behavior (RSMB)

The RSMB (Reiss, 1988) is an informant-based measure of psychopathology in people with mild, moderate, severe, or profound intellectual disabilities. There are 36 items consisting of 8 subscales on the RSMB: (1) Aggressive disorder, (2) Autism, (3) Avoidant disorder, (4) Dependent personality disorder, (5) Depression-behavioral signs, (6) Depression-physical signs (7) Paranoia, and (8) Psychosis. Each item is scored as no problem, a problem, or a major problem in the individual's life, and the rater is instructed to consider severity, frequency, and the consequences of the behavior when making those ratings (Havercamp & Reiss, 1997). Caretakers familiar with the individual rate each item. The RSMB is one of the older, more established scales for measuring psychopathology in individuals with intellectual disability (Matson et al., 2012). Research has shown very good levels of reliability and validity for the RSMB (Gustafsson & Sonnander, 2002; Havercamp & Reiss, 1997; Kishore, Nizamie, & Nizamie, 2010; Prout, 1993; Reiss, 1997; Straccia, Tasse, Ghisletta, & Barisnikov, 2013; Sturmey & Bertman, 1994; Sturmey, Burcham, & Perkins, 1995; Van Minnen, Savelsberg, & Hoogduin, 1995; Walsh & Shenouda, 1999). However, Sturmey and Bertman (1994) noted limitations in the internal consistency of the RSMB and little research on the validity and reliability of the psychosis subscale. Noted the same limitations as with the PAS-ADD, DASH-II, PIMRA, and the ABC, namely that standardization is mostly inadequate, the sensitivity and specificity of the measure is unknown, and the diagnostic accuracy is unknown.

Aberrant Behaviors Checklist (ABC)

The ABC is an informant-based scale designed to measure treatment effects in individuals with moderate to profound intellectual disability (Aman, Singh, Stewart, & Field, 1985a, 1985b). The ABC has 58 items that are rated on a four-point Likert scale (0–3), from "not at all a problem" to, "the problem is severe in degree." The items are measured on five subscales: (1) irritability, agitation, and crying; (2) lethargy and social withdrawal; (3) stereotypic behavior; (4) hyperactivity and noncompliance; and (5) inappropriate speech, and only several items reflect possible psychosis symptoms. The ABC has been frequently studied, and Aman (2012b, June Update) listed an annotated bibliography of over 300 studies using the ABC. In addition, Shedlack, Hennen, Magee, and Cheron (2005) used the

ABC to assess treatment effects of atypical antipsychotic medication in individuals with schizophrenia and found that antipsychotics are effective for some symptoms in patients with intellectual disability and comorbid psychiatric disorders. Subscale scores are considered more useful in the intellectual disability population than the total score (Aman, 2012a).

Assessment Measures Specific to Persons with Intellectual Disability and Bipolar Disorder

Diagnostic Assessment for the Severely Handicapped-II (DASH-II)

The DASH-II contains two subscales specific to persons with bipolar disorder, depression, and mania. The scale allows the measurement of frequency, duration and severity over the last 2 weeks of time. Items from the mania subscale include "is restless or agitated," "has a decreased need for sleep," "is cranky or irritable," "is extremely happy or cheerful for no obvious reason," "talks loudly," and "talks quickly." Items from the depression subscale include "has difficulty getting to sleep," "wakes up frequently during the night," "is cranky or irritable," "is restless or agitated," lacks interest in a favorite activity or object," and "speech or sound production is slow or lacks emotion." Specific to persons with bipolar disorder, the DASH-II mania subscale correctly identified 90.0% of manic individuals using DSM-IV criteria (Matson & Smiroldo, 1997).

Assessment of Dual Diagnosis (ADD)

The ADD contains two subscales that can assist in the evaluation of bipolar disorder – mania and depression. Items from the mania subscale include "unusual weight loss" and "afraid of disease" Rojahn et al., 2011). Items from the depression subscale include "decreased energy," "appears sad," and "unhappy" (Rojahn et al., 2011). Matson and Bamburg (1998) found the ADD had good levels of internal consistency for the subscales ranging from 0.77 to 0.95. They also found interrater reliability ranging from 0.82 to 1.00 and test-retest reliability, with 2 weeks between assessments, above 0.80. A limitation of the scale is there have not been studies looking at mania or depression in samples of persons with intellectual disability and bipolar disorders. Also, the items in the mania subscale do not reflect the core features of mania, namely expansive or elevated moods.

Psychopathology Instrument for Mentally Retarded Adults

The PRIMA has an affective disorder subscale that can be used to help evaluate bipolar disorders in individuals with intellectual disability. Items from affective disorder subscale include "Mood swings and moodiness," "decreased energy; mental and/or physical fatigue," "unusual weight loss in the last four months," "statements or appearance of sadness, loneliness, unhappiness, hopelessness and/or pessimism," "social withdrawal evidence by the person being less outgoing and evidencing less group participation," and "initial insomnia and restless sleep." The PRIMA has two versions: informant based and self-report, both of which should be administered in an interview format (Gustafsson & Sonnander, 2005). Matson et al. (1984) found internal consistency on PRIMA self-report and informant-report with a coefficient alpha 0.85 and 0.83.

Anxiety, Depression, and Mood Scale (ADAMS)

The ADAMS (Esbensen, Rojahn, Aman, & Ruedrich, 2003) is an informant-based behavioral rating scale that assesses affective/emotional symptoms of individuals with intellectual disability that are ages 10 and older. The ADAMS contains 29 items that assesses frequency and severity ratings over the past 6 months. The scale contains a manic/hyperactive behavior and

depressed mood subscales which may be useful in assessing bipolar disorder. Items from the manic/hyperactive subscale include, "does not relax" and "distracted". Items from the depressed Mood subscale include, "sad" and "lacks energy". The manic/hyperactive and depressed mood subscales have good internal consistency ranging from 0.75 to 0.80 (Esbensen et al., 2003). The ADAMS and the ADD depressed mood subscales were found to be highly correlated (r = 0.72), suggesting good concurrent validity (Esbensen & Benson, 2006; see Rojahn et al., 2011 as well).

Young Mania Rating Scale (Y-MRS)

The Y-MRS (Gracious, Youngstrom, Findling, & Calabrese, 2002) is an interview-based scale designed to assess for the symptoms of mania in children and adolescents. The Y-MRS contains 11 items that are scored from 0 to 4; the higher the score the more severe the symptom. The Y-MRS typically takes 15–30 minutes to complete and is administered in a clinical interview format, and symptoms are rated based on the last 48 hours (Gracious et al., 2002). If the client cannot verbalize their answers, the Parent version of Young Mania Rating Scale can be used (Youngstrom, Gracious, Danielson, Findling, & Calabrese, 2003). Items from the Young Mania Rating Scale and the Parent version of the Young Mania Rating Scale include "Elevated mood," "increased motor activity/energy," "sleep disturbance," "irritability," "speech (rate and amount)," and "disruptive/aggressive behavior." Matson, Gozalez, Terlinge, Thorson, and Laud (2007) found that the mania items from the scale significantly predicted a diagnosis of bipolar disorder in a sample of persons with intellectual disability.

Conclusions and Future Directions

The assessment of schizophrenia and bipolar disorder in persons with comorbid intellectual deficiency is challenging on many levels. First, cognitive impairments in speech, memory, and attention may preclude the use of many of the most common assessment measures. In fact, there seems to be a dichotomy between measures that can be used with persons with mild levels of intellectual disability compared to those having moderate to severe levels. The lower the level of functioning, the more behavioral the measures need to be and the more important informants and direct observation become. Also, there are differences in how these disorders are expressed clinically in this population which makes using current DSM-5 criteria more difficult. Second, there are a number of assessment measures that can be used in this population, but many have limited sample sizes and were developed using previous editions of the DSM. Some measures have fewer items and may not reflect the symptoms expressed in this population. Moving forward, it would be useful to carefully document what types of symptoms and behaviors are expressed in these clinical populations and update the current scales with the newer DSM-5 criteria. In the end, improving these assessment measures will enhance treatment options for these individuals which will ultimately lead to a better quality of life.

References

Altman, E. G., Hedeker, D., Peterson, J. L., & Davis, J. M. (1997). The Altman self-rating mania scale. *Biological Psychiatry, 42*(10), 948–955.

Aman, M. G. (1991). *Assessing psychopathology and behaviour problems in persons with mental retardation*. Rockville, MD: US Department of Health and Human Services.

Aman, M. G. (2012a). Aberrant Behavior Checklist: Current identity and future developments. *Clinical and Experimental Pharmacology, 2*(3), 2161–1459.

Aman, M. G. (2012b, June Update). *Annotated biography on the Aberrant Behavior Checklist* (*ABC*). Unpublished manuscript. Columbus, OH: The Ohio State University.

Aman, M. G., Singh, N. N., Stewart, A. W., & Field, C. J. (1985a). Psychometric characteristics of the Aberrant Behavior Checklist. *American Journal of Mental Deficiency, 89*(5), 492–502.

Aman, M. G., Singh, N. N., Stewart, A. W., & Field, C. J. (1985b). The Aberrant Behavior Checklist: A behavior rating scale for the assessment of treatment effects. *American Journal of Mental Deficiency, 89*(5), 485–491.

American Psychiatric Association. (2013). *Diagnostic and statistical manual of mental disorders* (5th ed.). Arlington, VA: American Psychiatric Publishing.

Andreasen, N. C. (1982). Negative symptoms in schizophrenia: Definition and reliability. *Archives of General Psychiatry, 39*, 784–788.

Andreasen, N. C., & Olsen, S. (1982). Negative versus positive schizophrenia: Definition and validation. *Archives of General Psychiatry, 39*, 784–788.

Balboni, G., Battagliese, G., & Pedrabissi, L. (2000). The psychopathology inventory for mentally retarded adults: Factor structure and comparisons between subjects with or without dualdiagnosis. *Research in Developmental Disabilities, 21*(4), 311–321.

Bamburg, J. W., Cherry, K. E., Matson, J. L., & Penn, D. (2001). Assessment of schizophrenia in persons with severe and profound mental retardation using the Diagnostic Assessment for the Severely Handicapped-II (DASH-II). *Journal of Developmental and Physical Disabilities, 13*(4), 319–331.

Beail, N., Mitchell, K., Vlissides, N., & Jackson, T. (2015). Concordance of the Mini-Psychiatric Assessment Schedule for Adults who have Developmental Disabilities (PASADD) and the brief symptom inventory. *Journal of Intellectual Disability Research, 59*(2), 170–175.

Bellack, A. S., & Thomas-Lohrman, S. (2003). *Maryland assessment of social competence*. Unpublished assessment manual. Baltimore, MD.

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*(C), 291–302.

Birchwood, M., Smith, J., Cochrane, R., Wetton, S., & Copstake, S. (1990). The social functioning scale: The development and validation of a new scale of social adjustment for the use in family intervention programmes with schizophrenic patients. *British Journal of Psychiatry, 157*, 853–859.

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, 51*(7), 922–924.

Combs, D. R., Mueser, K. T., Morales, S., & Smith, C. (2018). Schizophrenia spectrum and other psychotic disorders. In D. Beidel & C. Frueh (Eds.), *Adult psychopathology & diagnosis* (8th ed., pp. 159–208). New York, NY: Wiley Press.

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.

Deb, S., Thomas, M., & Bright, C. (2001). Mental disorder in adults with intellectual disability I: Prevalence of functional psychiatric disorder among a community-based population aged between 16 and 64 years. *Journal of Intellectual Disability Research, 45*, 495–505.

Depue, R. A., Slater, J. F., Wolfstetter-Kausch, H., Klein, D., Goplerud, E., & Farr, D. (1981). A behavioral paradigm for identifying persons at risk for bipolar depressive disorder: A conceptual framework and five validation studies. *Journal of Abnormal Psychology, 90*(5), 381.

Derogatis, L. R., & Melisaratos, N. (1983). The brief symptom inventory: An introductory report. *Psychological Medicine, 13*(3), 595–605.

Endicott, J., & Spitzer, R. L. (1978). A diagnostic interview: The schedule for affective disorders and schizophrenia. *Archives of General Psychiatry, 35*(7), 837–844.

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.

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 & Developmental Disorders, 33*(6), 617–629.

Goeree, R., Farahati, F., Burke, N., Blackhouse, G., O'Reilly, D., Pyne, J., & Tarride, J. E. (2005). The economic burden of schizophrenia in Canada in 2004. *Current Medical Research & Opinion, 21*, 2017–2028.

Gracious, B. L., Youngstrom, E. A., Findling, R. L., & Calabrese, J. R. (2002). Discriminative validity of a parent version of the Young Mania Rating Scale. *Journal of the American Academy of Child & Adolescent Psychiatry, 41*(11), 1350.

Gustafsson, C., & Sonnander, K. (2002). Psychometric evaluation of a Swedish version of the Reiss Screen for Maladaptive Behavior. *Journal of Intellectual Disability Research, 46*(3), 218–229.

Gustafsson, C., & Sonnander, K. (2005). A psychometric evaluation of a Swedish version of the Psychopathology Inventory for Mentally Retarded Adults (PIMRA). *Research in Developmental Disabilities: A Multidisciplinary Journal, 26*(2), 183–201.

Haddock, G., McCarron, J., Tarrier, N., & Faragher, E. B. (1999). Scales to measure dimensions of hallucinations and delusions: The psychotic symptom rating scales (PSYRATS). *Psychological Medicine, 29*(4), 879–889.

Havercamp, S. M., & Reiss, S. (1997). The Reiss Screen for Maladaptive Behavior: Confirmatory factor analysis. *Behaviour Research and Therapy, 35*(10), 967–971.

Helps, S. (2015). Psychopathology: Anxiety, depression, and schizophrenia. In J. L. Matson & M. L. Matson (Eds.), *Comorbid conditions in individuals with intellectual disability* (pp. 85–107). Cham, Switzerland: Springer International Publishing.

Hirschfeld, R. M., Williams, J. B., Spitzer, R. L., Calabrese, J. R., Flynn, L., Keck, P. E., Jr., … Russell, J. M. (2000). Development and validation of a screening instrument for bipolar spectrum disorder: The Mood Disorder Questionnaire. *American Journal of Psychiatry, 157*(11), 1873–1875.

Iverson, J. C., & Fox, R. A. (1989). Prevalence of psychopathology among mentally retarded adults. *Research in Developmental Disabilities, 10*(1), 77–83.

Janssen, R., & Maes, B. (2013). Psychometric evaluation of a Dutch version of the Mini PAS-ADD for assessing psychiatric disorders in adults with different levels of intellectual disability. *Journal of Intellectual Disability Research, 57*(8), 689–702.

Johnson, S. L., & Miklowitz, D. J. (2018). Bipolar and related disorders. In D. Beidel & C. Frueh (Eds.), *Adult psychopathology & diagnosis* (8th ed., pp. 209–246). New York, NY: Wiley Press.

Kay, S. R., Fiszbein, A., & Opler, L. A. (1987). The positive and negative syndrome scale (PANSS) for schizophrenia. *Schizophrenia Bulletin, 13*, 261–276.

Kendall, K., & Owen, M. J. (2015). Intellectual disability and psychiatric comorbidity: Challenges and clinical issues. *Psychiatric Times, 32*(5), 60–63.

Kishore, M. T., Nizamie, S. H., & Nizamie, A. (2010). Utility of Reiss Screen in identifying psychiatric problems in persons with mental retardation. *Indian Journal of Psychological Medicine, 32*(1), 38–41.

Knapp, M., Mangalore, R., & Simon, J. (2004). The global costs of schizophrenia. *Schizophrenia Bulletin, 30*, 279–293.

La Malfa, G., Notarelli, A., Hardoy, M. C., Bertelli, M., & Cabras, P. L. (1997). Psychopathology and mental retardation: An Italian epidemiological study using the PIMRA. *Research in Developmental Disabilities, 18*(3), 179–184.

Lai, C.-I., Hung, W.-J., Lin, L.-P., Chien, W.-C., & Lin, J.-D. (2011). A retrospective population-based data analyses of inpatient care use and medical expenditure in people with intellectual disability co-occurring schizophrenia. *Research in Developmental Disabilities: A Multidisciplinary Journal, 32*(3), 1226–1231.

Linaker, O. M., & Helle, J. (1994). Validity of the schizophrenia diagnosis of the psychopathology instrument for mentally retarded adults (PIMRA): A comparison of schizophrenic patients with and without mental retardation. *Research in Developmental Disabilities, 15*(6), 473–486.

Masi, G., Brovedani, P., Mucci, M., & Favilla, L. (2002). Assessment of anxiety and depression in adolescents with mental retardation. *Child Psychiatry and Human Development, 32*(3), 227–237.

Matson, J. L. (1988). *The PIMRA manual*. Orland Park, IL: International Diagnostic Systems.

Matson, J. L. (1995). *The diagnostic assessment for the severely handicapped-revised (DASH-II)*. Baton Rouge, LA: Disability Consultants, LLC.

Matson, J. L., & Bamburg, J. W. (1998). Reliability of the assessment of dual diagnosis (ADD). *Research in Developmental Disabilities, 19*(1), 89–95.

Matson, J. L., Belva, B., Hattier, M., & Matson, M. (2012). Scaling methods to measure psychopathology in persons with intellectual disabilities. *Research in Developmental Disabilities: A Multidisciplinary Journal, 33*(2), 549–562.

Matson, J. L., Gozalez, M. L., Terlinge, R. T., Thorson, R. T., & Laud, R. B. (2007). What symptoms predict the diagnosis of mania in persons with severe/profound intellectual disability in clinical practice? *Journal of Intellectual Disability Research, 51*(1), 25–31.

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.

Matson, J. L., & Malone, C. J. (2006). Validity of the sleep subscale of the Diagnostic Assessment for the Severely Handicapped–II (DASH-II). *Research in Developmental Disabilities, 27*(1), 85–92.

Matson, J. L., & Smiroldo, B. B. (1997). Validity of the mania subscale of the Diagnostic Assessment for the Severely Handicapped-II (DASH-II). *Research in Developmental Disabilities, 18*(3), 221–225.

Matson, J. L., Smiroldo, B. B., & Hastings, T. L. (1998). Validity of the autism/pervasive developmental disorder subscale of the Diagnostic Assessment for the Severely Handicapped–II (DASH-II). *Journal of Autism and Developmental Disorders, 28*(1), 77–81.

Meadows, G., Turner, T., Campbell, L., Lewis, S. W., Reveley, M. A., & Murray, R. M. (1991). Assessing schizophrenia in adults with mental retardation: A comparative study. *The British Journal of Psychiatry, 158*(1), 103–105.

Merikangas, K. R., Cui, L., Kattan, G., Carson, G. A., Youngstrom, E. A., & Angst, J. (2012). Mania with and without depression in a community sample of US adolescents. *Archives of General Psychiatry, 68*, 241–251.

Miller, C. J., Johnson, S. L., & Eisner, L. (2009). Assessment tools for adult bipolar disorder. *Clinical Psychology: Science and Practice, 16*(2), 188–201.

Mohr, C., Tonge, B. J., & Einfeld, S. L. (2005). The development of a new measure for the assessment of psychopathology in adults with intellectual disability. *Journal of Intellectual Disability Research, 49*(7), 469–480.

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). *The British Journal of Psychiatry, 163*(4), 471–480.

Moss, S., Prosser, H., Costello, H., Simpson, N., Patel, P., Rowe, S., … Hatton, C. (1998). Reliability and validity of the PAS-ADD Checklist for detecting psychiatric disorders in adults with intellectual disability. *Journal of Intellectual Disability Research, 42*(2), 173–183.

Moss, S., Prosser, H., Ibbotson, B., & Goldberg, D. (1996). Respondent and informant accounts of psychiatric symptoms in a sample of patients with learning disability. *Journal of Intellectual Disability Research, 40*(5), 457–465.

Myrbakk, E., & Von Tetzchner, S. (2008a). Psychiatric disorders and behavior problems in people with intellectual disability. *Research in Developmental Disabilities: A Multidisciplinary Journal, 29*(4), 316–332.

Myrbakk, E., & von Tetzchner, S. (2008b). Screening individuals with intellectual disability for psychiatric disorders: Comparison of four measures. *American Journal on Mental Retardation, 113*(1), 54–70.

Overall, J. E., & Gorham, D. R. (1962). The brief psychiatric rating scale. *Psychological Reports, 10*, 799–812.

Paclawskyj, T. R., Matson, J. L., Bamburg, J. W., & Baglio, C. S. (1997). A comparison of the Diagnostic Assessment for the Severely Handicapped-II (DASH-II) and the Aberrant Behavior Checklist (ABC). *Research in Developmental Disabilities, 18*(4), 289–298.

Patterson, T. L., Goldman, S., McKibbin, C. L., Hughs, T., & Jeste, D. (2001). UCSD performance- based skills assessment: Development of a new measure of everyday functioning for severely mentally ill adults. *Schizophrenia Bulletin, 27*, 235–245.

Prosser, H., Moss, S., Costello, H., Simpson, N., Patel, P., & Rowe, S. (1998). Reliability and validity of the Mini PAS-ADD for assessing psychiatric disorders in adults with intellectual disability. *Journal of Intellectual Disability Research: JIDR, 42*(Pt 4), 264–272.

Prout, H. (1993). Reviews and critiques of school psychology materials Douglas T. Brown, Associate Editor: Assessing psychopathology in persons with mental retardation: A review of the Reiss Scales. *Journal of School Psychology, 31*(4), 535–540.

Reiss, S. (1988). *The Reiss screen for maladaptive behavior test manual*. Worthington, OH: IDS Publishing Corporation.

Reiss, S. (1997). Comments on the Reiss screen for maladaptive behaviour and its factor structure. *Journal of Intellectual Disability Research, 41*(4), 346–354.

Rojahn, J., Rowe, E. W., Kasdan, S., Moore, L., & Van Ingen, D. J. (2011). Psychometric properties of the "Aberrant Behavior Checklist," the "Anxiety, Depression and Mood Scale," the "Assessment of Dual Diagnosis" and the "Social Performance Survey Schedule" in adults with intellectual disabilities. *Research in Developmental Disabilities: A Multidisciplinary Journal, 32*(6), 2309–2320.

Ryan, R. M. (1994). Recognition of psychosis in persons who do not use spoken communication. In R. J. Ancill, S. Holliday, & J. Higenbottam (Eds.), *Schizophrenia: Exploring the spectrum of psychosis* (pp. 339–344). New York, NY: Wiley Press.

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.

Straccia, C., Tasse, M. J., Ghisletta, P., & Barisnikov, K. (2013). The French version of the Reiss Screen for Maladaptive Behavior: Factor structure, point prevalence and associated factors. *Research in Developmental Disabilities, 34*(11), 4052–4061.

Sturmey, P., & Bertman, L. J. (1994). Validity of the Reiss Screen for Maladaptive Behavior. *American journal of mental retardation: AJMR, 99*(2), 201–206.

Sturmey, P., Burcham, K. J., & Perkins, T. S. (1995). The Reiss Screen for Maladaptive Behaviour: Its reliability and internal consistencies. *Journal of Intellectual Disability Research, 39*(3), 191–195.

Sturmey, P., Newton, J. T., Cowley, A., Bouras, N., & Holt, G. (2005). The PAS–ADD Checklist: Independent replication of its psychometric properties in a community sample. *The British Journal of Psychiatry, 186*(4), 319–323.

Sturmey, P. L., Matson, J. D., & Lott, J. (2004). The factor structure of the DASH-II. *Journal of Developmental and Physical Disabilities, 16*(3), 247–255.

Swiezy, N. B., Matson, J. L., Kirkpatrick-Sanchez, S., & Williams, D. E. (1995). A criterion validity study of the schizophrenia subscale of the Psychopathology Instrument for Mentally Retarded Adults (PIMRA). *Research in Developmental Disabilities, 16*(1), 75–80.

Thorson, R., Matson, J., Rojahn, J., & Dixon, D. (2008). Behaviour problems in institutionalised people with intellectual disability and schizophrenia spectrum disorders. *Journal of Intellectual and Developmental Disability, 33*(4), 316–322.

Turner, T. H. (1989). Schizophrenia and mental handicap: An historical review, with implications for further research. *Psychological Medicine, 19*(2), 301–314.

Van Minnen, A., Savelsberg, P. M., & Hoogduin, K. (1994). A Dutch version of the Psychopathology Inventory for Mentally Retarded Adults (PIMRA). *Research in Developmental Disabilities, 15*(4), 269–278.

Van Minnen, A., Savelsberg, P. M., & Hoogduin, K. (1995). A Dutch version of the Reiss Screen of Maladaptive Behavior. *Research in Developmental Disabilities, 16*(1), 43–49.

Walsh, K. K., & Shenouda, N. (1999). Correlations among the Reiss Screen, the adaptive behavior scale Part II, and the aberrant behavior checklist. *American Journal on Mental Retardation, 104*(3), 236–248.

Watson, J. E., Aman, M. G., & Singh, N. N. (1988). The psychopathology instrument for mentally retarded adults: Psychometric characteristics, factor structure, and relationship to subject characteristics. *Research in Developmental Disabilities, 9*(3), 277–290.

Wu, E. Q., Birnbaum, H. G., Shi, L., Ball, D. E., Kessler, R. C., Moulis, M., & Aggarwal, J. (2005). The economic burden of schizophrenia in the United States in 2002. *Journal of Clinical Psychiatry, 66*, 1122–1129.

Youngstrom, E. A., Gracious, B. L., Danielson, C. K., Findling, R. L., & Calabrese, J. (2003). Toward an integration of parent and clinician report on the Young Mania Rating Scale. *Journal of Affective Disorders, 77*(2), 179.

Zeilinger, E. L., Weber, G., & Haveman, M. J. (2011). Psychometric properties and norms of the German ABC-Community and PAS-ADD Checklist. *Research in Developmental Disabilities: A Multidisciplinary Journal, 32*(6), 2431–2440.

16 Assessing Autism in Dual Diagnosis

Johnny L. Matson and Joshua Montrenes

One of the most common forms of dual diagnosis is persons with ID who also experience autism. This situation exists due to the high overlap in the two conditions. Also, autism is now considered one of the more common neurodevelopmental disorders, along with ADHD (Johnson, Meyers, & the Council on Children with Disabilities, 2007). With this specific form of dual diagnosis, a number of other serious comorbidities are evident. For example, Minshawi, Hurwitz, Morriss, and McDougle (2015) describe the phenomenon of self-injurious behavior (SIB) in persons with autism and ID.

"Gold standard" diagnosis is a time-consuming process that involves history, observation, and use of one or more assessment scales (Falkmer, Anderson, Falkmer, & Horlin, 2013). To establish a gold standard for a specific scale, direct comparisons between measures are needed. To date, these studies have not been conducted. Furthermore, given the heterogeneity in core symptoms, the wide range of I.Q. scores, and the lifelong nature of the condition, it is highly likely that a one-size-fits-all measure will not emerge. Szatmari and colleagues (2002) further underscore the need to address cognitive functioning, since persons with autism may be classified as high or low functioning on I.Q. As a result, these authors and others have suggested that measures such as the Vineland Adaptive Behavior Scales be used in addition to measures core autism symptoms. They also note that more severe symptoms of autism are not necessarily linked to level of I.Q.

In the USA, most states are attempting to put assessment systems in place to push down the age of first diagnosis, often this means working through local pediatricians who typically serve as the entry point for at risk children. Identification of ASD can typically be done between 2 and 3 years of age (Matson, 2007). Thus, measures normed on very young children should be done with scales with a small number of items which are referred to as screeners. Where positive scores are reported, the screener would be followed by more detailed, lengthy assessments. This latter group of scales are called level two assessments. It is also the case that children are misdiagnosed or not diagnosed at all when they are very young. Thus, measurement still takes place at older ages as well. Milder symptoms of ASD, autistic regression, and Asperger's syndrome also can result in a diagnosis at older ages. All these factors have resulted in a proliferation of measures. In one review, for example, 21 tests were identified (Matson, Nebel-Schwalm, & Matson, 2007). Given the focus of this book, it should be underscored that I.Q. is a very important factor in the evaluation of symptoms of ASD. More intellectually impaired persons may have considerable limitations in following instruction and in verbalizing their thoughts and feelings (Prior, 1979).

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Symptom Presentation and Prevalence

I.Q. appears to be an important moderator of ASD symptoms. Matson, Dempsey, LoVullo, and Wilkins (2008) evaluated three groups: one with ASD and ID and one with ID only and one with Pervasive Developmental Disorder-Not Otherwise Specified (PDD-NOS) and ID. Each of these three groups was then split in half based on high or low I.Q. For the ASD and PDD-NOS groups, autism symptoms were high irrespective of high or low I.Q. However, for the ID only group, autism symptoms were almost as high for the low I.Q. group as for the ASD and PDD-NOS groups. The higher cognitive functioning ID group had negligible autism symptoms. This finding in group 3 may be related to the overlap of some symptoms of ID and ASD such as decreased communication and social skills. Also, the type of qualitative repetitive behaviors may vary from ASD to ID, but both groups do display these behaviors.

Along the lines noted above, Matson, Wilkins, and Ancona (2008) were among the first to look at how autism symptoms present in adults with very severe ID. Marked differences from previous studies of persons with mild ID and the core symptoms of ASD were noted. Specifically, among the 57 people who were evaluated, greater impairments in social interactions and more restricted and repetitive behaviors were evident in persons with severe versus mild ID. Additionally, more severe symptoms of ID and of ASD were major risk factors for greater deficits in adaptive behavior, and a greater likelihood of challenging behaviors, and comorbid psychopathology (Matson & Shoemaker, 2009).

Comorbidity of ASD to other conditions has been a fruitful area of study. La Malfa, Lassi, Bertelli, Salvini, and Placidi (2004) noted in an Italian sample that 40% of the 166 people they studied with ID also presented with autism. In other reviews, rates as high as 70% of people with ASD also presented with ID (Matson & Shoemaker, 2009). ID has been found to be a risk factor for a host of mental health conditions (Emerson, 2003). He found that children with anxiety-related conditions, conduct disorder, depression, and ASD were much more likely to occur in persons with ID when compared to the general population. This finding is consistent with multiple studies that show higher rates of mental health issues in the dually diagnosed group when compared to the broader population.

Scales for Measuring Core Symptoms of ASD

The purpose of using assessment scales in the evaluation of ASD is now well established. A substantial number of scales have been developed, and as mentioned there is no one measure that would be considered a "gold standard." Measures have been developed for various purposes; thus the reader needs to be familiar with the general field. For example, while they will not be covered in this chapter because the focus is on ID, multiple scales have been developed to assess Asperger's syndrome, which rarely appears in this lower cognitively functioning group. Also, diagnosis in particular, but assessment in general, should include history, observation, and where possible at least two measures of core symptoms of ASD, a measure of adaptive behavior, an I.Q. scale, a measure of challenging behaviors, a measure of comorbid psychopathology and when appropriate a functional assessment.

There has been considerable growth and refinement in measures of ASD over the years (Matson, Nebel-Schwalm, & Matson, 2007). One major reason for this continued evolution is that biomarkers, chemical assays, and other physiological methods have borne little fruit with respect to the assessment of ASD to date (Matson, 2007).

Early Scale Development

Schopler, Reichler, DeVellis, and Daly (1980) describe the state of assessment when they opened an autism in clinic in 1966. They were using the criteria laid out by Kanner (1943) in his classic paper. Also, they used the Rimland

Checklist, first published in 1964 and revised in 1971 (Rimland, 1964, 1971). This scale has largely fallen out of favor, and Schopler et al. (1980) noted that the Kanner criteria were not particularly workable, leading to the use of the Creak (1964) criteria. The latter criteria also proved to be problematic. As a result, the authors created the Childhood Autism Rating Scale (CARS). This scale, which is still in wide use today, broadened and redefined existing criteria. Major components of the scale included items which tapped impaired social development with respect to objects, people, and events. Also, criteria for disturbances in cognition and language as well as onset before 30 months were included.

Many studies have since built on this early research. Perry, Condillac, Freeman, Dunn-Geier, and Belair (2005), for example, evaluated 224 children between the ages of 2 and 6 years on the CARS. The authors found a strong relationship between CARS scores and DSM-IV diagnoses of autism. Similarly, Magyar and Pandolfi (2007) found that the ASD constructs identified with the CARS were consistent with the autism constructs reported in the DSM-IV.

The psychometrics of the CARS has also been compared to other measures developed to detect the core symptoms of ASD. In one such study, 65 children 18 months to 11 years of age were tested using the CARS and the Autism Behavior Checklist (ABC) (Rellini, Tortolani, Trillo, Carbone, & Montecchi, 2004). Validity was tested by matching test scores to DSM-IV criteria. Eighty-three percent of the people tested had a diagnosis of autism, 8% had an Asperger's syndrome diagnosis, 6% were diagnosed with Pervasive Developmental Disorder-Not Otherwise Specified (PDD-NOS), while 1½% had ADHD and 1½% had a language delay. The CARS proved to be better at identifying ASD symptoms when compared to the ABC. Finally, CARS scores for 143 children with I.Q.'s ranging from 30 to 138 were assessed. Eighty-nine kids had autism, 10 had PDD-NOS, and 44 had a range of other childhood disorders (Mayes et al., 2014). The diagnostic cutoff for the CARS correlated 84% with a DSM-5 diagnosis of ASD. However, going to one fewer symptom on social communication and interaction increased agreement to 94%. A considerable number of other scales have also been designed to measure core symptoms of autism. Some of these scales will now be reviewed.

Diagnostic Interview for Social and Communication Disorders (DISCO)

The DISCO is a scale used to interview parents and caregivers (Leekam, Libby, Wing, Gould, & Taylor, 2002). The scale can be used to help establish a diagnosis of autism via items in developmental history and core symptoms. The sample consisted of parent interviews of 36 children with autism, 17 with learning disability, and 14 with language disorder. The authors concluded that the DISCO was a reliable and valid instrument for diagnosing autism. Long-term outcomes on the DISCO have also been studied with 105 children diagnosed with autism (Billstedt, Carina Gillberg, & Gillberg, 2007).

Behavior Observation Scale for Autism (BOS)

Information was collected on 36 children with autism, 30 with ID, and 23 typically developing kids (Freeman, Ritvo, Guthrie, Schroth, & Ball, 1978). Diagnoses were made by two board-certified child psychiatrists. This scale is somewhat unique in that the 67 items in the scale were rated through a one way mirror while the child was in a playroom setting. The authors concluded that the BOS was easy to learn and a reliable instrument.

Ritvo-Freeman Real Life Rating Scale

The Ritvo-Freeman Real Life Rating Scale is a modified version of the BOS (Freeman, Ritvo, Yokota, & Ritvo, 1986). To simplify the BOS, this new measure deleted the client observation aspect of the scale. Efforts were also made to

simplify the definitions of the 47 behaviors included in the scale. These items were divided into five subscales, sensory motor, social relationship to people, affectual responses, sensory responses, and language. The authors report good reliability. In practice, their measure received much more attention than the BOS although it is not used a great deal presently.

Autism Behavior Checklist (ABC)

The ABC is a 57-item questionnaire that is designed to be completed by parents or teachers. Krug, Arick, and Almond (1980) generated items using nine sets of criteria including those of Kanner, Rimland and Creak. In a group of 62 participants, good reliability and validity of the scale were noted. Conversely, Volkmar and colleagues (1988) assessed 157 individuals: 94 with a diagnosis of autism and 63 without this diagnosis. They concluded that the scale had value but primarily as a screener. Subsequently, Miranda-Linné and Melin (2002) proposed a 5-factor analytic solution with 383 people diagnosed with autism. The sample ranged in age from 5 to 22 years. Their factors did not correspond to Krug et al. (1980) or that of Wadden, Bryson, and Rodger (1991). Thus, the overall value of the scale was further weakened.

Eaves and Williams (2006) tested 198 individuals with a mean age of 101 months on reliability and construct validity. As with Volkmar and colleagues (1988), they concluded that the scale was adequate but only for a screener. In a later study, with 107 participants, Eaves, Campbell, and Chambers (2000) reported good sensitivity and specificity of scores for making an autism diagnosis. These results are inconsistent with Rellini et al. (2004) who reported poor sensitivity for the ABC.

Nordin and Gillberg (1996) have also studied the ABC. In their paper, children with autism and ID were assessed. These researchers point to the fact that problem behaviors detected with the scale include not just core symptoms of autism but other problems typical of persons with ID.

Social Responsiveness Scale (SRS)

The SRS is a 65-item scale designed to measure core symptoms of autism in children and adolescents 4–18 years of age (Constantino & Gruber, 2005). Items are scored on a three-point Likert scale based on behavior over the last 6 months. The authors suggest that the measure can be used both as a screener and as an aid in diagnosing ASD.

A number of studies have been published on the psychometrics of this test and on its clinical utility. For example, Hus, Bishop, Gotham, Huerta, and Lord (2013) studied 2368 people with ASD and 1913 siblings without ASD. They concluded that for the ASD group, higher SRS scores were correlated with being older, having more challenging behaviors, having more language impairments, and having greater cognitive deficits. For the controls, higher SRS scores were associated with being male, being younger, and having poorer adaptive, expressive, and social behaviors.

Aldridge, Gibbs, Schmidhofer, and Williams (2012) point out that the SRS is a frequently used tool to aid in identifying ASD. And in this paper where 48 children were evaluated, high parent and teacher SRS scores were highly related to a diagnosis of ASD. In another study aimed at addressing the psychometrics of the SRS, the relationship between SRS and Autism Diagnostic Interview-Revised (ADI-R) scores was compared to DSM-IV diagnoses with 61 children (Constantino et al., 2003). The SRS scores correlated highly with the other autism measures and DSM-IV criteria. The authors concluded that the SRS is a valid measure of autism traits. Similarly, good reliability and validity of the SRS were noted in a German sample. In this study, 20 adults with ASD and 62 people with other mental disorders were assessed (Bölte, 2012).

Another study using a German sample was reported by Bölte, Poustka, and Constantino (2008). This study had a much larger sample than the Bölte (2012) study. They tested 838 typically developing children and 527 children with clinical conditions. The authors noted

that 160 received an ASD diagnosis. The convergent validity of the ADI-R and Autism Diagnostic Observation Schedule (ADOS) and the SRS proved to be high. Also, SRS raw scores for other mental health disorders were higher than what was reported with US samples. Conversely, in a sample of 500 5–8-year-olds from the UK, the SRS scores were very similar to US samples (Wigham et al., 2012). Additionally, the SRS proved to be useful in the evaluation of ASD symptoms in 21 children with tuberous sclerosis complex (Granader et al., 2010).

More recently, a revised version of the SRS, the SRS-2 has been published. Frazier, Ratliff, Zhang, Law, and Constantino (2014) studied 9635 individuals across a broad range of ages, severity of ASD symptoms, and who serve as the informant. A confirmatory factor analysis was run and resulted in two factors: social communication and repetitive/restricted behaviors. The authors point out that this factor solution conforms to DSM-5, ASD criteria.

Autism Diagnostic Interview-Revised (ADI-R)

The ADI-R is the updated version of a semi-structured interview for care providers for children and adults suspected of having ASD (Lord, Rutter, & Le Couteur, 1994). The scale is linked to DSM-IV and ICD-10 autism spectrum criteria. Additionally, this scale has been shortened, modified, and reorganized from the original scale. This measure has also been correlated with other well-established scales designed to measure ASD such as the CARS (Pilowsky, Yirmiya, Shulman, & Dover, 1998; Saemundsen, Magnússon, Smári, & Sigurdardóttir, 2003). The ADI-R was not specifically developed for persons with dual diagnosis. This scale is mentioned here because of its broad overall use in the diagnosis of ASD, especially in the context of university clinics and research protocols.

Autism Diagnostic Observation Schedule-Generic

Developed by the same group of researchers who developed the ADI-R, the scale was an evolutionary advance over the ADOS (Lord et al., 1989) and the Pre-Linguistic Autism Diagnostic Observation Schedule (PL-ADOS) (DiLavore, Lord, & Rutter, 1995). They note that the creation of the ADOS and the ADI (Le Couteur et al., 1989) led to the decision to develop the ADOS-G (Lord et al., 2000). The authors rightly note that most diagnoses of ASD are now done at an early age. Thus, the revisions were designed to extend the age for assessment downward. Components were added to the scale to address nonverbal children while also addressing the abilities and functioning of toddlers and infants. This scale, while not specific just to a dual diagnosis population, does address many of the concerns these individuals display.

Autism Spectrum Disorder-Diagnostic for Children (ASD-DC)

The ASD-DC is a measure of core symptoms of ASD in children, embedded in a test battery that also addresses comorbid psychopathology and problem behaviors. This scale is informant-based and involves interviewing a caregiver on the 40-item test. The scale, which is normed on 3–16-year-olds, has good reliability (Matson, Gonzalez, Wilkins, & Rivet, 2008). Good validity of the scale has been established as well (Matson, Gonzales, & Wilkins, 2009).

In one validity study, the ASD-DC was compared to the CARS with 3–14-year-old children. Total and subscale scores for both measures were highly correlated (Matson, Mahan, Hess, Fodstad, & Neal, 2010).

Autism Spectrum Disorders-Diagnostic Scale for Intellectually Disabled Adults (ASD-DA)

As the name implies, this scale was designed specifically for a dually diagnosed population. Additionally, unlike most scales that address core symptoms of ASD, this measure focuses on adults. The focus of the scale is to differentiate persons with ID only, compared to persons with ID and ASD. The scale consists of 31 items rated yes or no for the presence of symptoms addressing limited interests, repetitive movements of the limbs, changes in routine, and the ability to identify nonverbal social cues in others. In one study, 232 people with profound to mild ID and who had ID only or ID plus ASD were studied. Cutoff scores for ASD were established (Matson, Boisjoli, Gonzalez, Smith, & Wilkins, 2007). Also, sensitivity and specificity were found to be acceptable. Test-retest and inter-rater reliability were found to be good with a sample of 92 people (Matson & Boisjoli, 2008; Matson, Wilkins, & Gonzalez, 2007).

Validity of the scale was also reported. The ASD-DA was a good fit with DSM-IV-TR and ICD-10 criteria (Matson, Wilkins, Boisjoli, & Smith, 2008). In this later study, 156 adults with ID and with autism or PDD-NOS were compared to 151 people with ID only. The ASD-DA correlated weakly with the general psychopathology measure of the Diagnostic Assessment for the Severely Handicapped II (DASH-II).

Diagnostic Assessment for the Severely Handicapped II (DASH-II)

Another scale designed specifically for the measurement of psychopathology among persons with ID is the DASH-II. Autism is among the disorders that are assessed. Matson, Smiroldo, and Hastings (1998) did validate this particular subscale. Using the DASH-II, they found that the autism subscale was just as accurate as the CARS for classifying persons with autism and for classifying controls without autism. These results have been replicated by others (Hill & Furniss, 2006). The authors assessed 82 individuals with severe ID and challenging behaviors.

The DASH-II has also been used to measure impulse control, organic problems, anxiety, mood disorders, mania, schizophrenia, stereotypies, self-injurious behavior, elimination disorders, autism, eating disorders, and sleep and sexual problems. Only six items are specific to core symptoms of autism. Therefore, when this subscale is high, another measure specific to ASD should also be used to obtain a better overview of which core symptoms are present and what behaviors should be targeted for treatment.

Behavioral Summarized Evaluation (BSE)

The BSE is a 20-item checklist designed to evaluate autistic traits (Barthelemy et al., 1990). These authors found that the scale had good reliability and validity. Barthelemy et al. (1990) suggest that the test can be useful as a measure of treatment effectiveness in clinical trials.

Parent Interview for Autism (PIA)

A structured interview administered to primarily caregivers, the PIA was evaluated with 165 children under the ages of 6 years (Stone & Hogan, 1993). The initial paper reported good internal consistency and test-retest reliability. This scale also was useful in differentiating a group with ID versus children with autism.

Gilliam Autism Rating Scale (GARS)

The GARS was initially normed on 1092 people from the USA and Canada who ranged in age from 3 to 22 (Gilliam, 1995). There are 56 items over 4 subscales including social interaction, communication, stereotyped behavior, and developmental disturbances on a 4-point scale, In one major replication study however, the GARS consistently missed a number of persons with autism

in a sample of 119 children with DSM-IV diagnoses of autism (South et al., 2002).

Early Detection

Identifying ASD as early as possible is one of the primary goals in assessment. This goal can be complicated by autistic regression (around 20–24 months), having milder symptoms, having high I.Q., or evincing comorbidities, particularly ID (Vostanis, Smith, Chung, & Corbett, 1994). At this time, it is hard to determine what measure is best since few direct comparisons between scales have been made (Norris & Lecavalier, 2010). As a result only indirect comparisons are available, most notably based on the number and quality of studies published and the degree to which scales have been replicated by research teams other than the developers.

Baby and Infant Screen for Children with aUtIsm Traits (BISCUIT)

Easily the most heavily researched of the early childhood diagnostic screening instruments is the BISCUIT. The initial study evaluated the reliability of the measure. The scale was individually administered to caregivers of 276 children 17–37 months of age (Matson et al., 2009). Two major ethnic groups with a few exceptions were assessed: White (53%) and African American (40%). The BISCUIT consists of three parts: I, a 71-item measure of core symptoms; II an 84-item scale to assess comorbidities such as conduct disorder, ADHD, Tics, obsessive-compulsive disorder, phobias, and eating difficulties; and, III, a 20-item assessment of challenging behaviors including aggressive, disruptive, self-injurious, and stereotyped behaviors. The reliability for all of these scales proved to be good.

The BISCUIT-Part I has also been factor analyzed. Matson, Boisjoli, Hess, and Wilkins (2010) evaluated 1287 children between 17 and 37 months of age who were "at risk" for developmental disabilities. The test was administered to a parent of each of the children. A three-factor solution emerged, and the scale was able to differentiate at risk children with and without autism. Similarly, the scale proved to have very good sensitivity and specificity (Matson et al., 2009). This set of psychometric properties was established with an at-risk population of 1007 children. Additionally, the BISCUIT-Part I has been validated, showing strong convergent validity with the Modified Checklist for Autism in Toddlers (M-CHAT) and the personal social domain of the Battelle Developmental Inventory, Second Addition (BDI-2) (Matson, Wilkins, & Fodstad, 2011).

The BISCUIT-Part I has also been used to assess a wide range of differences in diagnostic patterns. For example, in a study of 7464 toddlers from 17 to 37 months of age, no disparities between children of European and African decent on core symptoms of autism were found (Williams, Matson, Beighley, & Konst, 2015).

Checklist for Autism in Toddlers (CHAT) and the Modified Checklist for Autism in Toddlers (M-CHAT)

Initially developed as the CHAT, the scale was designed for the early detection of ASD (Baron-Cohen et al., 2000). The scale consisted of nine questions in section A, answered by a parent. The remaining five questions in section B are completed by a healthcare provider. The authors screened 16,235, 18-month-olds from a general population of children. There were 369 kids labeled as moderate risk for autism, with 38 children labeled as high risk. The authors concluded that the scale was a good screener. However, Mawle and Griffiths (2006) reached the opposite conclusion. They note that "the CHAT demonstrates a level of sensitivity unlikely to be useful for population screening purposes." Soon after the development of the CHAT (less than a decade), a modified version of the scale was published, the M-CHAT. Mawle and Griffiths (2006) report that the revised scale may be more sensitive and thus more appropriate than the CHAT as a screener.

Robins, Fein, Barton, and Green (2001) report on the M-CHAT. They used the 23-item, yes/no scale to screen 1293 children at their 18 or 24 month pediatric checkups. Their screen identified 58 children, 39 of whom were later diagnosed with ASD. Cutoff scores were created, and good reliability was established. Six items proved to be the best for identifying ASD. Kleinman et al. (2008) also describe the M-CHAT as a promising ASD screener. In their study, 3793 children between the ages of 16–30 months of age were screened. Retest was conducted for 1416 of these children at 42–54 months of age.

The M-CHAT has also established psychometrics across a number of countries. Wong et al. (2004) tested 212 children who were 18–24 months of age. This Chinese sample was set up in two stages. If the child scored positive on two or more of the initial seven items, then the entire 23-item scale was administered. These authors called for more international studies on the scale. The M-CHAT was also administered to a cohort of children 16–23 months of age and a second cohort of 24–30 months old. The M-CHAT was equally useful in detecting ASD for both groups (Pandey et al., 2008).

The newest version of this test is the Modified Checklist for Autism in Toddlers-Revised with Follow-Up (M-CHAT-R/F). In this study 16,071 18–24-month-olds were screened (Robins et al., 2014). Those children, who were deemed as at-risk for ASD, were administered the M-CHAT--R/F a second time. The authors conclude that this new version is better at detecting ASD than the M-CHAT. The M-CHAT and M-CHAT-R/F were also compared in a general sample of 18,989 young children. Fifty-four percent of the children appeared to be at risk. Of this group, 98% evinced sufficient developmental delays to warrant early intervention (Chlebowski, Robins, Barton, & Fein, 2013).

Autism Spectrum Rating Scales (ASRS)

This scale is broken into three subscales using factor analysis: social/communication, unusual behaviors, and self-regulation. The authors report good reliability (Goldstein & Naglieri, 2012). One particularly novel aspect of the scale is that it has scoring for persons with minimal or no speech. To date the ASRS has not been extensively researched.

Developmental Behavior Checklist (DBC)

Developed by Einfeld and Tonge (1991, 1995), this scale aims to aid in the screening of young people with autism. The measure was filled out by lay informants on 664 children and adolescents with ID and emotional disturbance. Six subscales were established, and good internal reliability, inter-rater reliability, and test-retest reliability were established.

Brereton, Tonge, Mackinnon, and Einfeld (2002) made an effort to extend the DBC, a 96-item scale, to determine if a subset of items in the scale could be used to screen for autism. They evaluated 381 children and adolescents between 4 and 18 years of age who had received a DSM-IV diagnosis of autism from an experienced clinician. These authors established a 29-item subgroup from the DBC that was useful in screening at-risk children including those with ID to determine if autism was present.

Additional modifications of the DBC have been reported elsewhere in the literature. Dekker, Nunn, Einfeld, Tonge, and Koot (2002), for example, obtained parent and teacher report data on 1536 Dutch and Australian participants ranging in age from 3 to 22. The people in their sample ranged from mild to profound ID. Five subscales emerged via factor analysis: disruptive/antisocial, self-absorbed, communication, disturbance, and anxiety and social relating. Using this same sample, Dekker, Nunn, and Koot (2002) reported good reliability. A 24-item short form of the DBC has also been developed (Taffe et al., 2007). These authors note that this shorter version has low bias and high precision in cross validation.

Screening Tool for Autism in 2-Year-Olds (STAT)

Initial psychometrics of this scale, the STAT, were reported by Stone, Coonrod, Turner, and Pozdol (2004). Study one was designed to identify cutoff scores in two groups of 2-year-olds; children with autism and children with nonspectrum problems. These authors report high sensitivity and specificity validated against the ADOS-G. These authors also report good levels of inter-rater agreement and rest-retest reliability. Strong psychometrics is also reported with a younger sample, 12–23 months of age (Stone, McMahon, & Henderson, 2008).

The STAT has also been translated into other languages. For example, Chiang et al. (2013) tested a Taiwanese sample. Study one had 15 children with autism and 15 with developmental delay (DD) or language impairment (LI). In study two, 77 young kids with autism, Pervasive Developmental Disorder-Not Otherwise Specified, or DD/LI were evaluated. These authors found excellent sensitivity and specificity, and they established cutoff scores. Chiang et al. (2013) concluded that the Taiwanese-STAT (T-STAT) was a useful screener for 2–3-year-olds in their country.

Checklist for Autism in Young Children

This scale consists of 30 items and was administered to 143 children (Mayes & Calhoun, 1999). The scale compared favorably to DSM-IV diagnosis of autism. The development of this scale and the other early detection measures are very important to ensure that young children have access to comprehensive services (Vostanis et al., 1994).

Assessing Treatment Effectiveness

Pervasive Developmental Disorders Behavior Inventory (PDDBI)

Another and very novel approach to the assessment of autism was the development of a scale to assess treatment effectiveness (Cohen, Schmidt-Lackner, Romanczyk, & Sudhalter, 2003). The Pervasive Developmental Disorders Behavior Inventory (PDDBI) is filled out by teachers or parents and measures both adaptive and maladaptive behaviors. Topics addressed include adaptive, language, and social skills. Specific items that differentiate children on the autism spectrum from the general population were included. To establish the scale's psychometrics, 311 parents of children and adolescents aged 1–17 years of age were tested. For 270 cases, a teacher was also assessed. Typical items included empathy behaviors, offered help when others are sick, and displayed social play behaviors. Good reliability and validity were reported.

Comorbid Disorders

Within the area of dual diagnosis, many individuals have multiple problems. Herring et al. (2006) note, for example, that challenging behaviors and mental health disorders occur at high rates in persons with ID, often beginning early in life. These problems are considered to be particularly acute among persons with ID and autism. One of the more common mental health issues in this latter group is symptoms of depression (Perry, Marston, Hinder, Munden, & Roy, 2001). Bakken et al. (2010) in a Norwegian sample studied a group with ID only and a group with autism and ID. Over 50% of the ID plus autism group evinced mental health disorders, while just under 20% of the ID group had these concerns. Melville et al. (2008) using a similar methodology found no differences in rates of mental health disorders between these groups.

Social skills have also been evaluated in relationship to persons with ID only and ID and autism or PDD-NOS (Matson, Dempsey, & Rivet, 2009). For the ASD group, there were particularly high scores on impulsivity and mania.

Some disorders may have different symptom presentation. Anxiety is one such problem. In a sample diagnosed with autism and ID, many symptoms were similar. However, signs of psychological arousal may be more difficult to identify (Hagopian & Jennett, 2008; Helverschou & Martinsen, 2011). Also, for persons with autism

and/or ID, mediation, sleep problems, and anxiety were associated with increased rates of challenging behaviors (Rzepecka, McKenzie, McClure, & Murphy, 2011).

ADHD and autism have also been evaluated in the context of ID (Mayes, Calhoun, Mayes, & Molitoris, 2012). These authors studied 1005 children and concluded that ADHD symptoms were frequently reported in children with autism. Similarly, level of intelligence for persons with autism did not differ based on ADHD symptoms of attention, impulsivity, and hyperactivity. This finding is interesting and is underscored by the fact that persons with autism 4–18 years of age have greater rates of emotional and behavioral problems than persons with ID (Brereton, Tonge, & Einfeld, 2006). This finding is significant when taken in the light of the fact that persons with ID have much higher rates of psychopathology than the general population. These data are supported by the works of Bradley, Summers, Wood, and Bryson (2004) who found high rates of psychopathology in persons with autism. Specific types of psychopathology were also more common among persons with autism. Disorders were particularly prevalent for anxiety, mood, sleep, organic syndromes, and stereotypies/tics. In a related study with 112 10–14-year-olds with autism, 70% had at least one comorbid disorder, while 41% of the sample evinced 2 or more comorbid mental health issues. Similarly, La Malfa et al. (2007) also found that pervasive developmental disorders were a risk factor for other forms of psychopathology.

Tsakanikos et al. (2006) addressed the issue of comorbid mental health disorders among adults with autism and ID ($n = 147$) and 605 adults with ID only. These findings differed from what was noted previously in that these authors found no differences in rates of comorbid psychopathology in the two groups.

Measures of Comorbid Psychopathology

Several tests have been developed to evaluate emotional disorders that accompany ID.

Autism Spectrum Disorders-Comorbidity for Adults (ASD-CA)

In one study designed to establish cutoff scores for the ASD-CA, 313 participants who lived in two developmental centers served as the sample (LoVullo & Matson, 2009). The persons who were tested were divided into three groups: ID, ID plus ASD, and ID plus ASD plus another emotional disorder. In addition, to establish cutoff scores, the frequency of various disorders was reported. The most frequent disorder was pica followed by bipolar, mood disorder, major depression, posttraumatic stress disorder, psychotic disorder, tics, Alzheimer condition, anxiety, ADHD, and rumination.

Psychopathology in Autism Checklist (PAC)

This scale was developed to assess various mental health disorders in persons with autism and ID. The population studied was from Norway. Their sample consisted of 32 participants with mild or moderate ID and 30 persons with severe ID (Bakken et al., 2010). Over half of the autism and ID group evinced another disorder, while the ID only group had about a 20% rate of additional forms of psychopathology. About 60% of persons with comorbid psychopathology experienced at least two more disorders. Disorders measured with the PAC included psychosis, depression, anxiety disorders, obsessive-compulsive disorder, and general adjustment problems.

Child Behavior Checklist (CBCL)

The CBCL is a widely recognized measure of psychopathology in the general population of children. It is a parent report scale for children and adolescents 4–18 years of age. Internalizing disorders such as anxiety, depression, thought problems, and attention issues are measured. Externalizing problems include destructive behavior, social problems, aggression, and delinquent behavior. The scale has also been used to

identify children with autism and related problems. A Brazilian sample of 101 children age 4–11 years was studied (Duarte, Bordin, De Oliveira, & Bird, 2003). In this sample, there were 36 participants with autism and related conditions, 31 people with psychiatric disorders, and 34 typically developing children. These authors concluded that the Autism/Bizarre and Thought Problems subscales differentiated children with autism from other mental health disorders or children with no mental health problems. These data were confirmed in a study by Biederman et al. (2010). They studied 65 children with I.Q.s above 70 using the CBCL and 85 children without ASD and I.Q.s over 70. They found that these two groups could be differentiated using the withdrawn, social, and thought problems subscales. Dekker, Koot, Ende, and Verhulst (2002) also looked at differences on the CBCL and levels of ID. Participants were 6–18 years of age. Two groups of special education students (n = 1041) were divided into a 60–80 I.Q. group and a 30–60 I.Q. group and then compared to 1855 typically developing children. Challenging behaviors were more prevalent in the ID groups. Persons in the 60–80 I.Q. range were more likely to display social problems, attention difficulties, and aggression. Participants in the 30–60 I.Q. groups on the other hand more frequently exhibited social problems, attention difficulties, withdrawal, and thought problems.

Developmental Behavior Checklist (DBC)

Another measure used to assess emotional disorders among persons with ID is the DBC (Dekker, Nunn, Einfeld, et al., 2002). These authors evaluated parents and teachers of 1536 Dutch and Australian participants, 3–22 years of age. The sample consisted of individuals across the spectrum of mild to profound ID. Dekker et al. concluded that the DBC was useful for assessing emotional problems and challenging behaviors. Psychometric characteristics with respect to good validity of the DBC have also been reported (Einfeld & Tonge, 1995). Einfeld and Tonge (1995) also studied the DBC. They reported good reliability and validity with a group of children who evinced emotional disorders, behaviors problems, and ID.

Autism Spectrum Disorders-Behavior Problem-Adult Version (ASD-BP)

The ASD-BP is a 19-item scale that has been item and factor analyzed. The four factors generated were aggression/destruction, stereotypy, self-injurious behavior, and disruptive behaviors. Also, the scale which was designed for persons with autism and ID has good reliability and validity. Trend analysis has also shown that persons with autism, ID, and epilepsy had more challenging behaviors than persons with ID only (Smith & Matson, 2010). Matson and Rivet (2008) also using this scale found that the severity of core autism symptoms among persons with autism and ID resulted in more challenging behaviors. These findings are supported by other studies (Matson & Nebel-Schwalm, 2007; Totsika, Hastings, Emerson, Lancaster, & Berridge, 2011). This latter finding was replicated using the Disability Assessment Schedule with 562 participants (McCarthy et al., 2010).

Adaptive Behavior

The measurement of adaptive behavior is particularly pertinent for persons with ID or individuals who are suspected of having ID. A number of studies have been conducted with a dually diagnosed sample, particularly persons with autism and ID. Autism is a particularly popular topic of study at this writing, and as noted earlier in the chapter, a considerable level of comorbidity between the two conditions exists.

In one study, persons with autism or PDD-NOS and some persons with and some without ID were studied using the Vineland Adaptive Behavior Scales. There were 34 people with an I.Q. over 70 and 33 people with an I.Q. below 70. The general finding was that lower I.Q. negatively impacted communication.

Paul et al. (2004) looked at the differences in adaptive behavior in persons with mild or moderate ID who were placed in an autism group or a group with PDD-NOS. They found few differences in adaptive behavior. However, where disparities were evident, they tended to involve expressive communication, with greater problems in the autism group. Specific deficits in communication were also noted in a sample of 684 children with autism (Carter et al., 1998). Not surprisingly, greater deficits were noted in children who were nonverbal versus children who had some verbal skills. Finally, in another study using the Vineland Adaptive Behavior Scales, children with autism displayed more deficits in adaptive and social behaviors than a developmentally disabled group without autism (Volkmar et al., 1987).

Conclusions

The area of dual diagnosis has expanded markedly, particularly in the area of assessment. A number of normed scales with established reliability and validity are available to measure a range of emotional disorders, challenging behaviors, and adaptive skills in this population. Instruments that cover children, adolescents, and adults have been developed, and the measures cover persons from mild to profound ID. In the process of evaluation of persons with possible mental health concerns, it is highly recommended that one or more of the "best fit" measures of autism be included in a comprehensive evaluation also involving history and observation.

References

Aldridge, F. J., Gibbs, V. M., Schmidhofer, K., & Williams, M. (2012). Investigating the clinical usefulness of the social responsiveness scale (SRS) in a tertiary level, autism spectrum disorder specific assessment clinic. *Journal of Autism and Developmental Disorders, 42*, 294–300.

Bakken, T. L., Helverschou, S. B., Eilertsen, D. E., Heggelund, T., Myrbakk, E., & Martinsen, H. (2010). Psychiatric disorders in adolescents and adults with autism and intellectual disability: A representative study in one county in Norway. *Research in Developmental Disabilities, 31*, 1669–1677.

Baron-Cohen, S., Wheelwright, S., Cox, A., Baird, G., Charman, T., Swettenham, J., … Doehring, P. (2000). Early identification of autism by the checklist for autism in toddlers (CHAT). *Journal of the Royal Society of Medicine, 93*, 521–525.

Barthelemy, C., Adrien, J. L., Tanguay, P., Garreau, B., Fermanian, J., Roux, S., … Lelord, G. (1990). The behavioral summarized evaluation: Validity and reliability of a scale for the assessment of autistic behaviors. *Journal of Autism and Developmental Disorders, 20*, 189–204.

Biederman, J., Petty, C. R., Fried, R., Wozniak, J., Micco, J. A., … Faraone, S. V. (2010). Child behavior checklist clinical scales discriminate referred youth with autism spectrum disorder: A preliminary study. *Journal of Developmental and Behavioral Pediatrics, 31*, 485–490.

Billstedt, E., Carina Gillberg, I., & Gillberg, C. (2007). Autism in adults: Symptom patterns and early childhood predictors. Use of the DISCO in a community sample followed from childhood. *Journal of Child Psychology and Psychiatry, 48*, 1102–1110.

Bölte, S. (2012). Brief report: The social responsiveness scale for adults (SRS-A): Initial results in a German cohort. *Journal of Autism and Developmental Disorders, 42*, 1998–1999.

Bölte, S., Poustka, F., & Constantino, J. N. (2008). Assessing autistic traits: Cross-cultural validation of the social responsiveness scale (SRS). *Autism Research, 1*, 354–363.

Bradley, E. A., Summers, J. A., Wood, H. L., & Bryson, S. E. (2004). Comparing rates of psychiatric and behavior disorders in adolescents and young adults with severe intellectual disability with and without autism. *Journal of Autism and Developmental Disorders, 34*, 151–161.

Brereton, A. V., Tonge, B. J., & Einfeld, S. L. (2006). Psychopathology in children and adolescents with autism compared to young people with intellectual disability. *Journal of Autism and Developmental Disorders, 36*, 863–870.

Brereton, A. V., Tonge, B. J., Mackinnon, A. J., & Einfeld, S. L. (2002). Screening young people for autism with the developmental behavior checklist. *Journal of the American Academy of Child and Adolescent Psychiatry, 41*, 1369–1375.

Carter, A. S., Volkmar, F. R., Sparrow, S. S., Wang, J. J., Lord, C., Dawson, G., … Schopler, E. (1998). The Vineland adaptive behavior scales: Supplementary norms for individuals with autism. *Journal of Autism and Developmental Disorders, 28*, 287–302.

Chiang, C. H., Wu, C. C., Hou, Y. M., Chu, C. L., Liu, J. H., & Soong, W. T. (2013). Development of T-STAT for early autism screening. *Journal of Autism and Developmental Disorders, 43*, 1028–1037.

Chlebowski, C., Robins, D. L., Barton, M. L., & Fein, D. (2013). Large-scale use of the modified checklist for autism in low-risk toddlers. *Pediatrics, 131*, e1121–e1127.

Cohen, I. L., Schmidt-Lackner, S., Romanczyk, R., & Sudhalter, V. (2003). The PDD behavior inventory: A rating scale for assessing response to intervention in children with pervasive developmental disorder. *Journal of Autism and Developmental Disorders, 33*, 31–45.

Constantino, J. N., Davis, S. A., Todd, R. D., Schindler, M. K., Gross, M. M., Brophy, S. L., … Reich, W. (2003). Validation of a brief quantitative measure of autistic traits: Comparison of the social responsiveness scale with the autism diagnostic interview-revised. *Journal of Autism and Developmental Disorders, 33*, 427–433.

Constantino, J. N., & Gruber, C. P. (2005). The social responsiveness scale: Western Psychological Services.

Creak, E. M. (1964). Schizophrenic syndrome in childhood: Further progress report of a working party (April, 1961). *Developmental Medicine and Child Neurology, 4*, 530–535.

Dekker, M. C., Koot, H. M., Ende, J. V. D., & Verhulst, F. C. (2002). Emotional and behavioral problems in children and adolescents with and without intellectual disability. *Journal of Child Psychology and Psychiatry, 43*, 1087–1098.

Dekker, M. C., Nunn, R., & Koot, H. M. (2002). Psychometric properties of the revised developmental behaviour checklist scales in Dutch children with intellectual disability. *Journal of Intellectual Disability Research, 46*, 61–75.

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*, 601–610.

DiLavore, P. C., Lord, C., & Rutter, M. (1995). The prelinguistic autism diagnostic observation schedule. *Journal of Autism and Developmental Disorders, 25*, 355–379.

Duarte, C. S., Bordin, I. A., De Oliveira, A., & Bird, H. (2003). The CBCL and the identification of children with autism and related conditions in Brazil: Pilot findings. *Journal of Autism and Developmental Disorders, 33*, 703–707.

Eaves, R. C., Campbell, H. A., & Chambers, D. (2000). Criterion-related and construct validity of the pervasive developmental disorders rating scale and the autism behavior checklist. *Psychology in the Schools, 37*, 311–321.

Eaves, R. C., & Williams, T. O. (2006). The reliability and construct validity of ratings for the autism behavior checklist. *Psychology in the Schools, 43*, 129–142.

Einfeld, S. L., & Tonge, B. J. (1991). Psychometric and clinical assessment of psychopathology in developmentally disabled children. *Australia and New Zealand Journal of Developmental Disabilities, 17*, 147–154.

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*, 81–104.

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

Falkmer, T., Anderson, K., Falkmer, M., & Horlin, C. (2013). Diagnostic procedures in autism spectrum disorders: A systematic literature review. *European Child and Adolescent Psychiatry, 22*, 329–340.

Frazier, T. W., Ratliff, K. R., Gruber, C., Zhang, Y., Law, P. A., & Constantino, J. N. (2014). Confirmatory factor analytic structure and measurement invariance of quantitative autistic traits measured by the social responsiveness scale-2. *Autism, 18*, 31–44.

Freeman, B. J., Ritvo, E. R., Yokota, A., & Ritvo, A. (1986). A scale for rating symptoms of patients with the syndrome of autism in real life settings. *Journal of the American Academy of Child Psychiatry, 25*(1), 130–136.

Freeman, B. J., Ritvo, E. R., Guthrie, D., Schroth, P., & Ball, J. (1978). The behavior observation scale for autism: Initial methodology, data analysis, and preliminary findings on 89 children. *Journal of the American Academy of Child Psychiatry, 17*, 576–588.

Gilliam, J. E. (1995). *Gilliam autism rating scale*. Austin, TX: Pro-Ed.

Goldstein, S., & Naglieri, J. A. (2012). *Autism spectrum rating scales (ASRS)*. Toronto, ON: Multi-Health Systems.

Granader, Y. E., Bender, H. A., Zemon, V., Rathi, S., Nass, R., & MacAllister, W. S. (2010). The clinical utility of the social responsiveness scale and social communication questionnaire in tuberous sclerosis complex. *Epilepsy and Behavior, 18*, 262–266.

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.

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.

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.

Hill, J., & Furniss, F. (2006). Patterns of emotional and behavioural disturbance associated with autistic traits in young people with severe intellectual disabilities and challenging behaviours. *Research in developmental disabilities, 27*(5), 517–528.

Hus, V., Bishop, S., Gotham, K., Huerta, M., & Lord, C. (2013). Factors influencing scores on the social responsiveness scale. *Journal of Child Psychology and Psychiatry, 54*, 216–224.

Johnson, C. P., Myers, S. M., & The Council on Children with Disabilities. (2007). Identification and evaluation of children with autism spectrum disorders. *The American Academy of Pediatrics, 120*, 126–134.

Kanner, L. (1943). Autistic disturbances of affective contact. *Nervous Child: Journal of Psychopathology, Psychotherapy, Mental Hygiene, and Guidance of the Child, 2*, 217–250.

Kleinman, J. M., Robins, D. L., Ventola, P. E., Pandey, J., Boorstein, H. C., … Fein, D. (2008). The modified checklist for autism in toddlers: A follow-up study investigating the early detection of autism spectrum disorders. *Journal of Autism and Developmental Disorders, 38*, 827–839.

Krug, D. A., Arick, J., & Almond, P. (1980). Behavior checklist for identifying severely handicapped individuals with high levels of autistic behavior. *Journal of Child Psychology and Psychiatry, 21*, 221–229.

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*, 262–267.

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*, 218–228.

Le Couteur, A., Rutter, M., Lord, C., Rios, P., Robertson, S., Holdgrafer, M., & McLennan, J. (1989). Autism diagnostic interview: A semistructured interview for parents and caregivers of autistic persons. *Journal of Autism and Developmental Disorders, 19*, 363–387.

Leekam, S. R., Libby, S. J., Wing, L., Gould, J., & Taylor, C. (2002). The diagnostic interview for social and communication disorders: Algorithms for ICD-10 childhood autism and Wing and Gould autistic spectrum disorder. *Journal of Child Psychology and Psychiatry, 43*, 327–342.

Lord, C., Risi, S., Lambrecht, L., Cook, E. H., Jr., Leventhal, B. L., … Rutter, M. (2000). The autism diagnostic observation schedule-generic: A standard measure of social and communication deficits associated with the spectrum of autism. *Journal of Autism and Developmental Disorders, 30*, 205–223.

Lord, C., Rutter, M., Goode, S., Heemsbergen, J., Jordan, H., Mawhood, L., & Schopler, E. (1989). Autism diagnostic observation schedule: A standardized observation of communicative and social behavior. *Journal of Autism and Developmental Disorders, 19*, 185–212.

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 of Autism and Developmental Disorders, 24*, 659–685.

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.

Magyar, C. I., & Pandolfi, V. (2007). Factor structure evaluation of the childhood autism rating scale. *Journal of Autism and Developmental Disorders, 37*, 1787–1794.

Matson, J. L. (2007). Current status of differential diagnosis for children with autism spectrum disorders. *Research in Developmental Disabilities, 28*, 109–118.

Matson, J. L., & Boisjoli, J. A. (2008). Autism spectrum disorders in adults with intellectual disability and comorbid psychopathology: Scale development and reliability of the ASD-CA. *Research in Autism Spectrum Disorders, 2*, 276–287.

Matson, J. L., Boisjoli, J. A., Gonzalez, 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*, 330–338.

Matson, J. L., Boisjoli, J. A., Hess, J. A., & Wilkins, J. (2010). Factor structure and diagnostic fidelity of the Baby and Infant Screen for Children with aUtIsm Traits–Part 1 (BISCUIT–Part 1). *Developmental Neurorehabilitation, 13*, 72–79.

Matson, J. L., Dempsey, T., LoVullo, S. V., & Wilkins, J. (2008). The effects of intellectual functioning on the range of core symptoms of autism spectrum disorders. *Research in Developmental Disabilities, 29*, 341–350.

Matson, J. L., Dempsey, T., & Rivet, T. T. (2009). The interrelationships of psychopathology symptoms on social skills in adults with autism or PDD-NOS and intellectual disability. *Journal of Developmental and Physical Disabilities, 21*, 39–55.

Matson, J. L., Gonzalez, M. L., Wilkins, J., & Rivet, T. T. (2008). Reliability of the autism spectrum disorder-diagnostic for children (ASD-DC). *Research in Autism Spectrum Disorders, 2*, 533–545.

Matson, J. L., Mahan, S., Hess, J. A., Fodstad, J. C., & Neal, D. (2010). Convergent validity of the autism spectrum disorder-diagnostic for children (ASD-DC) and childhood autism rating scales (CARS). *Research in Autism Spectrum Disorders, 4*, 633–638.

Matson, J. L., & Nebel-Schwalm, M. (2007). Assessing challenging behaviors in children with autism spectrum disorders: A review. *Research in Developmental Disabilities, 28*, 567–579.

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*, 38–54.

Matson, J. L., & Rivet, T. T. (2008). The effects of severity of autism and PDD-NOS symptoms on challenging behaviors in adults with intellectual disabilities. *Journal of Developmental and Physical Disabilities, 20*, 41–51.

Matson, J. L., & Shoemaker, M. (2009). Intellectual disability and its relationship to autism spectrum disorders. *Research in Developmental Disabilities, 30*, 1107–1114.

Matson, J. L., Smiroldo, B. B., & Hastings, T. L. (1998). Validity of the autism/pervasive developmental disorder subscale of the diagnostic assessment for the

severely handicapped-II. *Journal of Autism and Developmental Disorders, 28*, 77–81.

Matson, J. L., Wilkins, J., & Ancona, M. (2008). Autism in adults with severe intellectual disability: An empirical study of symptom presentation. *Journal of Intellectual and Developmental Disability, 33*, 36–42.

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.

Matson, J. L., Wilkins, J., & Fodstad, J. C. (2011). The validity of the Baby and Infant Screen for Children with aUtIsm Traits: Part 1 (BISCUIT: Part 1). *Journal of Autism and Developmental Disorders, 41*, 1139–1146.

Matson, J. L., Wilkins, J., & Gonzalez, M. (2007). Reliability and factor structure of the autism spectrum disorders—diagnosis scale for intellectually disabled adults (ASD—DA). *Journal of Developmental and Physical Disabilities, 19*, 565–577.

Matson, J. L., Wilkins, J., Sevin, J. A., Knight, C., Boisjoli, J. A., & Sharp, B. (2009). Reliability and item content of the Baby and Infant Screen for Children with aUtIsm Traits (BISCUIT): Parts 1–3. *Research in Autism Spectrum Disorders, 3*, 336–344.

Matson, J. L., Wilkins, J., Sharp, B., Knight, C., Sevin, J. A., & Boisjoli, J. A. (2009). Sensitivity and specificity of the Baby and Infant Screen for Children with aUtIsm Traits (BISCUIT): Validity and cutoff scores for autism and PDD-NOS in toddlers. *Research in Autism Spectrum Disorders, 3*, 924–930.

Matson, J. L., Gonzalez, M., & Wilkins, J. (2009). Validity study of the autism spectrum disorders-diagnostic for children (ASD-DC). *Research in Autism Spectrum Disorders, 3*(1), 196–206.

Mawle, E., & Griffiths, P. (2006). Screening for autism in pre-school children in primary care: Systematic review of English language tools. *International Journal of Nursing Studies, 43*, 623–636.

Mayes, S. D., & Calhoun, S. L. (1999). Early classification and detection. *Infants and Young Children, 12*, 90–97.

Mayes, S. D., Calhoun, S. L., Mayes, R. D., & Molitoris, S. (2012). Autism and ADHD: Overlapping and discriminating symptoms. *Research in Autism Spectrum Disorders, 6*, 277–285.

Mayes, S. D., Calhoun, S. L., Murray, M. J., Pearl, A., Black, A., & Tierney, C. D. (2014). Final DSM-5 under-identifies mild autism spectrum disorder: Agreement between the DSM-5, CARS, CASD, and clinical diagnoses. *Research in Autism Spectrum Disorders, 8*, 68–73.

McCarthy, 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*, 362–366.

Melville, C. A., Cooper, S. A., Morrison, J., Smiley, E., Allan, L., … 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.

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*, 1541–1568.

Miranda-Linné, F. M., & Melin, L. (2002). A factor analytic study of the autism behavior checklist. *Journal of Autism and Developmental Disorders, 32*, 181–188.

Nordin, V., & Gillberg, C. (1996). Autism spectrum disorders in children with physical or mental disability or both. II: Screening aspects. *Developmental Medicine & Child Neurology, 38*(4), 314–324.

Norris, M., & Lecavalier, L. (2010). Screening accuracy of level 2 autism spectrum disorder rating scales: A review of selected instruments. *The International Journal of Research and Practice, 14*, 263–284.

Pandey, J., Verbalis, A., Robins, D. L., Boorstein, H., Klin, A., … Fein, D. (2008). Screening for autism in older and younger toddlers with the modified checklist for autism in toddlers. *Journal of Autism and Developmental Disorders, 31*, 131–144.

Paul, R., Miles, S., Cicchetti, D., Sparrow, S., Klin, A., … Booker, S. (2004). Adaptive behavior in autism and pervasive developmental disorder-not otherwise specified: Microanalysis of scores on the Vineland adaptive behavior scales. *Journal of Autism and Developmental Disorders, 34*, 223–228.

Perry, A., Condillac, R. A., Freeman, N. L., Dunn-Geier, J., & Belair, J. (2005). Multi-site study of the childhood autism rating scale (CARS) in five clinical groups of young children. *Journal of Autism and Developmental Disorders, 35*, 625–634.

Perry, D. W., Marston, G. M., Hinder, S. A. J., Munden, A. C., & Roy, A. (2001). The phenomenology of depressive illness in people with a learning disability and autism. *The International Journal of Research and Practice, 5*, 265–275.

Pilowsky, T., Yirmiya, N., Shulman, C., & Dover, R. (1998). The autism diagnostic interview-revised and the childhood autism rating scale: Differences between diagnostic systems and comparison between genders. *Journal of Autism and Developmental Disorders, 28*, 143–151.

Prior, M. R. (1979). Cognitive abilities and disabilities in infantile autism: A review. *Journal of Abnormal Child Psychology, 7*, 357–380.

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*, 703–708.

Rimland, B. (1964). *Infantile autism: The syndrome and its implication for a neural theory of behaviour (2nd printing)*. New York, NY: Appleton-Century-Crofts.

Rimland, B. (1971). The differentiation of childhood psychoses: An analysis of checklists for 2,218 psychotic children. *Journal of Autism and Childhood Schizophrenia, 1*, 161–174.

Robins, D. L., Casagrande, K., Barton, M., Chen, C. M. A., Dumont-Mathieu, T., & Fein, D. (2014). Validation of the modified checklist for autism in toddlers, revised with follow-up (M-CHAT-R/F). *Pediatrics, 133*, 37.

Robins, D. L., Fein, D., Barton, M. L., & Green, J. A. (2001). The modified checklist for autism in toddlers: An initial study investigating the early detection of autism and pervasive developmental disorders. *Journal of Autism and Developmental Disorders, 31*, 131–144.

Rzepecka, H., McKenzie, K., McClure, I., & Murphy, S. (2011). Sleep, anxiety and challenging behaviour in children with intellectual disability and/or autism spectrum disorder. *Research in Developmental Disabilities, 32*, 2758–2766.

Saemundsen, E., Magnússon, P., Smári, J., & Sigurdardóttir, S. (2003). Autism diagnostic interview-revised and the childhood autism rating scale: Convergence and discrepancy in diagnosing autism. *Journal of Autism and Developmental Disorders, 33*, 319–328.

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*, 91–103.

Smith, K. R., & Matson, J. L. (2010). Behavior problems: Differences among intellectually disabled adults with co-morbid autism spectrum disorders and epilepsy. *Research in Developmental Disabilities, 31*, 1062–1069.

South, M., Williams, B. J., McMahon, W. M., Owley, T., Filipek, P. A., … Ozonoff, S. (2002). Utility of the Gilliam autism rating scale in research and clinical populations. *Journal of Autism and Developmental Disorders, 32*, 593–599.

Stone, W. L., Coonrod, E. E., Turner, L. M., & Pozdol, S. L. (2004). Psychometric properties of the STAT for early autism screening. *Journal of Autism and Developmental Disorders, 34*, 691–701.

Stone, W. L., & Hogan, K. L. (1993). A structured parent interview for identifying young children with autism. *Journal of Autism and Developmental Disorders, 23*, 639–652.

Stone, W. L., McMahon, C. R., & Henderson, L. M. (2008). Use of the screening tool for autism in two-year-olds (STAT) for children under 24 months: An exploratory study. *The International Journal of Research and Practice, 12*, 557–573.

Szatmari, P., Merette, C., Bryson, S. E., Thivierge, J., Roy, M. A., … Maziade, M. (2002). Quantifying dimensions in autism: A factor-analytic study. *Journal of the American Academy of Child and Adolescent Psychiatry, 41*, 467–474.

Taffe, J. R., Gray, K. M., Einfeld, S. L., Dekker, M. C., Koot, H. M., … Tonge, B. J. (2007). Short form of the developmental behaviour checklist. *American Journal on Mental Retardation, 112*, 31–39.

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*, 91–99.

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*, 1123–1129.

Volkmar, F. R., Cicchetti, D. V., Dykens, E., Sparrow, S. S., Leckman, J. F., & Cohen, D. J. (1988). An evaluation of the autism behavior checklist. *Journal of Autism and Developmental Disorders, 18*, 81–97.

Volkmar, F. R., Sparrow, S. S., Goudreau, D., Cicchetti, D. V., Paul, R., & Cohen, D. J. (1987). Social deficits in autism: An operational approach using the Vineland adaptive behavior scales. *Journal of the American Academy of Child and Adolescent Psychiatry, 26*, 156–161.

Vostanis, P., Smith, B., Chung, M. C., & Corbett, J. (1994). Early detection of childhood autism: A review of screening instruments and rating scales. *Child: Care, Health and Development, 20*, 165–177.

Wadden, N. P., Bryson, S. E., & Rodger, R. S. (1991). A closer look at the autism behavior checklist: Discriminant validity and factor structure. *Journal of Autism and Developmental Disorders, 21*, 529–541.

Wigham, S., McConachie, H., Tandos, J., Le Couteur, A. S., & Gateshead Millennium Study Core Team. (2012). The reliability and validity of the social responsiveness scale in a UK general child population. *Research in Developmental Disabilities, 33*, 944–950.

Williams, L. W., Matson, J. L., Beighley, J. S., & Konst, M. (2015). Ethnic disparities in early autism assessment: A large scale screening study of infants and toddlers. *Journal of Developmental and Physical Disabilities, 27*, 141–148.

Wong, V., Hui, L. H. S., Lee, W. C., Leung, L. S. J., Ho, P. K. P., … Chung, B. (2004). A modified screening tool for autism (checklist for autism in toddlers [CHAT-23]) for Chinese children. *Pediatrics, 114*, e166–e176.

17 Assessment and Diagnosis of Attention-Deficit/Hyperactivity Disorder in Individuals with Intellectual Disability

Maya Matheis

Attention-deficit/hyperactivity disorder (ADHD) is one of the most common co-occurring psychiatric disorders among individuals with intellectual disability (ID; Clark & Bélanger, 2018). Research has also shown that individuals with dual diagnoses of ID and ADHD have greater impairments in adaptive behavior, suggesting that ADHD may be another source of disability (Carmeli, Klein, & Sohn, 2007). Despite the increased prevalence of ADHD among individuals with ID (Baker, Neece, Fenning, & Blacher, 2010; Neece, Baker, Blacher, & Crnic, 2011; Neece, Baker, Crnic, & Blacher, 2013), symptoms of ADHD in this population are often subject to diagnostic overshadowing, in which they are overlooked or attributed to cognitive deficits without further assessment or treatment (Evans & Trollor, 2016; Jopp & Keys, 2001; Mason & Scior, 2004). The assessment of ADHD in individuals with ID can present significant challenges, especially as there is a lack of research on ADHD in this population.

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Attention-Deficit/Hyperactivity Disorder

ADHD is a neurodevelopmental disorder that emerges in early childhood and is characterized by symptoms of inattention, impulsivity, and/or hyperactivity that cause functional impairment across settings (American Psychiatric Association, 2013; Nigg & Barkley, 2014). A large body of empirical evidence has validated the diagnosis of ADHD among children, adolescents, and adults with average intellectual functioning (Roberts, Milich, & Barkley, 2014). It is the most common neurodevelopmental disorder, estimated to affect 9.4% of children aged 2–17 and 4.4% of adults in the United States (Danielson et al., 2018; Kessler et al., 2006). The following section describes core symptoms of ADHD in the general population, outlines diagnostic criteria, and also discusses the application of ADHD diagnostic criteria to individuals with ID.

ADHD Symptomology

Core symptoms ADHD is conceptualized as having two primary symptom domains, inattention, and hyperactivity/impulsivity, which are highly correlated but distinct factors (American Psychiatric Association, 2013; Nigg & Barkley, 2014; Roberts et al., 2014). Difficulties with sustaining attention, thought to be reflective of

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problems with executive function, often result in disorganization, problems in planning, inability to remember and follow through with instructions or rules, and reduced resistance to distractions (Roberts et al., 2014). Common parent and teacher concerns for children who struggle with inattention include difficulty listening, failure to finish tasks, and having difficulty with concentration (Nigg & Barkley, 2014). While these types of concerns may be present with a number of different learning disabilities or psychiatric conditions, research has found that children with ADHD exhibit significantly more off-task behavior, more difficulty with shifting tasks, and slower return to an activity after interruption than children with other conditions (Nigg & Barkley, 2014).

Symptoms in the domain of hyperactivity/impulsivity include those related to physical overactivity as well as those related to impulsivity and disinhibition (Roberts et al., 2014). While hyperactivity and impulsivity are separate and distinct concepts, research has demonstrated that these symptoms are correlated strongly enough in individuals with ADHD to represent a single symptom factor (Nigg & Barkley, 2014). Children with ADHD who have difficulties with hyperactivity demonstrate an excessive level of physical activity, which may be characterized by fidgetiness or difficulty remaining seated when necessary, talking excessively, as well as more general activity (e.g., running, climbing). Difficulties with impulsivity often results in difficulty in waiting one's turn, responding quickly to situations without waiting for instructions or thinking about consequences, and generally difficulty regulating behavior (Nigg & Barkley, 2014).

Environmental context Research has shown that ADHD symptoms are not consistent across settings. Symptoms of ADHD are most likely to emerge in settings that require an individual to regulate their behavior, resist impulses, restrict their movement, and sustain attention to a task or activity (Roberts et al., 2014). The novelty of a task has also been shown to be related to the level of behavioral difficulties in children with ADHD, with behavior worsening as familiarity with a task increases (Beike & Zentall, 2012). Differences in levels of ADHD symptoms and disruptive behavior have also been seen between different caregivers, across times of day (e.g., worse behavior when fatigued or toward the end of the school day), and across levels of supervision (Roberts et al., 2014). Lower rates of symptoms and better behavior have also been found when children with ADHD are engaged in highly reinforcing activities (Barkley, Copeland, & Sivage, 1980).

Developmental progression Onset of ADHD symptoms typically occurs around ages 3–4, with symptoms related to hyperactivity/impulsivity often emerging first (Nigg & Barkley, 2014; Owens, Cardoos, & Hinshaw, 2014). Symptoms related to inattention typically emerge around ages 5–8, within the first few grades of elementary school, and are likely to co-occur with symptoms of hyperactivity/impulsivity through middle and late childhood (Nigg & Barkley, 2014; Owens et al., 2014). The severity of ADHD symptoms has been found to wane in adolescence and adulthood, with difficulties related to hyperactivity/impulsivity in particular becoming less pronounced, although many adults continue to have significant difficulties with inattention and/or hyperactivity/impulsivity throughout their lifetime (Barkley, Murphy, & Fischer, 2010; Surman & Goodman, 2017).

Gender differences Similar to other neurodevelopmental disorders, ADHD is more prevalent among males, with an estimated male: female ratio of 2:1 in children (Polanczyk, de Lima, Horta, Biederman, & Rohde, 2007). Research studies comparing ADHD symptoms between the genders have found that girls with ADHD are more likely to present with internalizing symptoms, while boys with ADHD are more likely to have externalizing symptoms such as aggressive behavior (Nigg & Barkley, 2014; Owens et al., 2014). Higher rates of disruptive behavior in boys may results in them being referred for ADHD assessment more frequently compared to

girls (Nigg & Barkley, 2014). Boys and girls with ADHD have been found to have similar difficulties with executive functioning, similar rates of academic and social impairments, and higher levels of difficulties in these areas compared to children without ADHD (Nigg & Barkley, 2014). Among adults, ADHD symptoms have been found to be similar in men and women (Owens et al., 2014).

Associated impairments Children and adolescents with ADHD are at greater risk for a number of functional problems, including academic difficulties and academic underachievement, motor impairments, language ability, and difficulties with emotional regulation and self-esteem (Nigg & Barkley, 2014; Weyandt & Gudmundsdottir, 2014). Social difficulties, such as social skill deficits, troublemaking and keeping friends, and rejection by peers, are also common for children and adolescents with ADHD (Nigg & Barkley, 2014; Owens et al., 2014). ADHD in childhood has also been found to increase the risk oppositional defiant disorder (ODD) or conduct disorder (CD), either co-occurring in childhood or later in adolescence (Owens et al., 2014). Adolescents with ADHD are at greater risk for driving problems (e.g., reckless driving, driving citations, accidents), nicotine use, and risky sexual behavior compared to adolescents without ADHD (Owens et al., 2014). Adults with ADHD have been found to have more difficulties in occupational functioning, to be at greater risk for mood and anxiety disorders, and to be a greater risk for substance use disorders compared to adults without ADHD (Adler, Spencer, Stein, & Newcorn, 2008; Owens et al., 2014).

Diagnostic Criteria for ADHD

DSM-5 Criteria for a diagnosis of ADHD are specified in the fifth edition of the *Diagnostic and Statistical Manual of Mental Disorders* (*DSM-5;* American Psychiatric Association, 2013). These criteria are based on decades of research studies examining ADHD symptomology and the validity of an ADHD diagnosis, as well as expert opinion (Roberts et al., 2014). Two core symptom areas of inattention and hyperactivity/impulsivity are described within Criteria A and B, with nine symptoms in each criterion (American Psychiatric Association, 2013). Criterion A includes inattentive symptoms: making careless mistakes; having difficulty sustaining attention during tasks or play activities; not listening to when spoken to directly; not following through on tasks; difficulty organizing tasks and activities; avoiding tasks that require sustained attention; often losing things; being easily distracted; and being forgetful regarding daily activities. Criterion B, focused on symptoms related to hyperactivity and impulsivity, includes often fidgeting with hands or feet; leaving one's seat in situations that require remaining in place; running or climbing when such activities are inappropriate; having difficulty being quiet during play or other activities; restlessness; talking excessively; blurting out answers; having difficulty waiting one's turn; and interrupting or intruding on others. These symptoms must be inconsistent with the individual's developmental level and negatively impact daily activities (American Psychiatric Association, 2013). For children and adolescents under the age of 17, at least six symptoms from within one category must occur persistently; for youth and adults 17 years and older, five symptoms from at least one category must be present.

Criterion D specifies that ADHD symptoms must interfere with social, academic, or occupational functioning (American Psychiatric Association, 2013). Additional criteria specify that symptoms must have an onset before 12 years of age (Criterion B), be present in two or more settings (Criterion C), and cannot be better explained by other disorder or occur exclusively in the context of a psychotic disorder (Criterion E). The *DSM-5* describes three presentations of ADHD: combined presentation, in which criteria for both the inattention and hyperactivity/impulsivity categories are met; predominantly inattentive presentation, in which criteria for only the inattention category are met; and predominantly hyperactive/impulsive presentation, in which

criteria for only the hyperactivity/impulsivity category are met. Specifiers can be added to indicate the severity of symptoms or functional impairment (i.e., mild, moderate, severe), as well as if the condition is in partial remission (i.e., when full diagnostic criteria is not currently met, but was in the past, with symptoms still resulting in impairment).

Historical changes in DSM criteria "Hyperkinetic reaction of childhood" first appeared in the second edition of the *Diagnostic and Statistical Manual of Mental Disorders* (*DSM-II)* and was succinctly described: "characterized by overactivity, restlessness, distractibility, and short attention span, especially in young children; the behavior usually diminishes by adolescence" (American Psychiatric Association, 1968, p. 50). With the third edition *(DSM-III)*, this disorder was renamed "attention deficit disorder (ADD) (with and without hyperactivity)" and was reconceptualized to be characterized primarily by difficulties with inattention and impulsivity, with symptoms of hyperactivity as a secondary, and nonessential, feature (American Psychiatric Association, 1980). The *DSM-III* included three categories of symptoms for inattention, impulsivity, and hyperactivity. The term attention-deficit/hyperactivity disorder (ADHD) was introduced in the revised third edition *(DSM-III-R),* reflecting the reintegration of hyperactivity as a core feature of the disorder (American Psychiatric Association, 1987). Symptoms of inattention, hyperactivity, and impulsivity were combined into a single list of symptoms. In the fourth edition *(DSM-IV)*, the three subtypes of ADHD were introduced (i.e., predominantly inattentive type, predominantly hyperactive-impulsive type, and combined type), which represented a return to conceptualizing the possibility of a form of the disorder with solely inattentive symptoms (American Psychiatric Association, 1994; Lange, Reichl, Lange, Tucha, & Tucha, 2010). The *DSM-IV* criteria included two categories of symptoms (i.e., inattention, hyperactivity/impulsivity) and also described examples of ADHD symptoms in adulthood. ADHD diagnostic criteria remained unchanged with the release of the text revision of the fourth edition *(DSM-IV-TR;* American Psychiatric Association, 2000*).*

Changes to ADHD criteria between the *DSM-IV-TR* and *DSM-5* were not as drastic as those between prior *DSM* editions, with the core structure of the criteria remaining stable. Minor changes included a reduction in the minimum required number of symptoms for older adolescents and adults, adjusting the required onset of symptoms to before age 7 to before age 12 and adding modifiers for severity and partial remission (Epstein & Loren, 2013). One of the most notable changes, particularly in the context of individuals with ID, was the removal of pervasive developmental disorders (PDD) as exclusionary diagnoses from Criterion E. In the *DSM-IV-TR*, PDD referred to a category of conditions including autistic disorder, Asperger's disorder, and PDD-not otherwise specified, which were consolidated into the diagnosis of autism spectrum disorder (ASD) within the *DSM-5*, reflecting advancements in our understanding of autism as a single disorder with heterogenous presentations (Mahjouri & Lord, 2012). The exclusion of PDD meant that individuals with a diagnosis of autism could not receive a co-occurring diagnosis of ADHD prior to the *DSM-5*. Given that approximately 30–60% of individuals with ASD are estimated to have comorbid ID (Baio et al., 2018; Matson & Shoemaker, 2009), this exclusionary criteria prevented many individuals with ID from receiving a diagnosis of ADHD prior to the release of the DSM-5 in 2013, as well as likely excluding them from participating in research on the co-occurrence of ADHD with ID.

ICD-11 The most recent revision of the World Health Organization's *International Classification of Diseases* (*ICD-11*; World Health Organization, 2018) includes diagnostic criteria for ADHD, which was known as "hyperkinetic disorder" (HKD) in previous versions. Diagnostic criteria for ADHD in the *ICD-11* is very similar to that of the *DSM-5*, albeit with a few notable differences. The *ICD-11* describes symptom domains of inattention and hyperactivity-impulsivity, without

specifying a number of discrete symptoms of each domain that must be met for diagnostic threshold. Further, ADHD criteria in the *ICD-11* do not have a specific age requirement for onset of symptoms, instead requiring onset within the general periods of early to mid-childhood (World Health Organization, 2018).

Like in the *DSM-5*, symptoms must be present in more than one setting, significantly impairing and be inconsistent with an individual's developmental level, with the *ICD-11* specifying that symptoms must be "outside the limits of normal variation expected for age and level of intellectual functioning" (World Health Organization, 2018). In addition to ADHD presentation subtypes of combined presentation (ADHD-C), predominantly inattentive presentation (ADHD-PI), and predominantly hyperactive-impulsive presentation (ADHD-PHI), the *ICD-11* also includes subtypes for other specified presentation (ADHD-Y) and presentation unspecified (ADHD-Z).

Application of Diagnostic Criteria to Individuals with ID

Application of the *DSM-5* and *ICD-11* diagnostic criteria for ADHD to individuals with ID may be challenging and may require consideration of equivalents in symptom presentation as well as symptoms that cannot be identified. The *Diagnostic Manual-Intellectual Disability 2 (DM-ID-2;* Fletcher & Cooper, 2017) was created to address the need for adaptations of mental health and psychiatric disorder criteria for children, adolescents, and adults with ID. It provides recommendations for applying *DSM-5* ADHD criteria, with specific examples of how to reframe individual criterion in consideration of the developmental level of an individual. For example, it stresses the need to consider symptoms within the context of developmentally appropriate activities, rather than tasks that may be expected of the individual based on chronological age but that may be developmentally inappropriate (e.g., schoolwork, chores).

Both *DSM-5* and *ICD-11* criteria for ADHD require symptoms to be present in at least two different settings (American Psychiatric Association, 2013; World Health Organization, 2018). Unlike for typically developing children, for whom ADHD symptoms in the school setting are common and are often a primary referral concern, difficulties in the school environment may be difficult to assess with children with ID in relation to ADHD. Children with ID may experience difficulties in school related to disparities in their cognitive functioning, and the expectations of their environments, or conversely, may be receiving substantial supports and accommodations in their academic settings, which may make it difficult to discern if ADHD symptoms are causing significant impairment in that setting. For these reasons, the *DM-ID-2* highlights the fact that diagnostic criteria does not require ADHD symptoms to be present or impairing in the school setting and that a variety of alternative settings (e.g., playground, supermarket, restaurant) can and should be considered when applying this criteria to individuals with ID (Fletcher & Cooper, 2017). Additionally, the *DM-ID-2* stresses the need to consider expressive language ability when applying ADHD criteria to individuals with ID, as *DSM-5* specified symptoms of speaking excessively, and blurting out answers may not be relevant to individuals with limited verbal abilities.

Diagnostic Validity Among Individuals with ID

Until recently, ADHD was not considered to co-occur with ID, as symptoms of inattention, hyperactivity, and impulsivity in this population were assumed to be inherently related to an individual's cognitive deficits (Antshel, Phillips, Gordon, Barkley, & Faraone, 2006; Deutsch, Dube, & McIlvane, 2008). This view dominated clinical practice and research for many decades, as many asserted that low cognitive functioning was characterized by deficits in attention (Neece et al., 2011; Tonge et al., 1996). The "Defect Approach," as this perspective was named, was based on a

body of literature that seemingly demonstrated that individuals with cognitive deficits consistently had difficulties related to attention (Burack, Evans, Klaiman, & Iarocci, 2001). Critics of the "Defect Approach" argued that there was not enough evidence to support this stance and that the supporting research base contained a number of methodological problems, most notably the common practice of comparing the performance of individuals with ID to typically developing individuals matched by chronological age (Burack et al., 2001; Iarocci & Burack, 1998).

The "Developmental Model" was a general approach introduced by Zigler (1969), who maintained that developmental principles applied to people universally, regardless of cognitive ability. Within this lens, individuals with ID are thought to follow a similar developmental trajectory as typically developing individuals, just at a slower rate. Proponents of this viewpoint argued the need for research to compare individuals with ID to others based on mental age (MA), not chronological age, and maintained that individuals with ID showed variation in a number of different functioning domains, including inattention and hyperactivity/impulsivity (Burack et al., 2001). The Developmental Model was reflected in diagnostic criteria for ADHD in the *DSM-III-R, DSM-IV,* and *DSM-IV-TR,* which specified that a diagnosis of ADHD could be made if the related symptoms exceeded those compatible with an individual's MA (American Psychiatric Association, 1987, 1994, 2000). However, application of ADHD criteria to this population was still controversial, as, until recently, there was a limited amount of research to help guide our understanding of when symptom elevation could be considered inconsistent with an individual's level of developmental functioning.

The diagnostic validity of ADHD in children with ID was examined by Antshel et al. (2006) using the validation model developed by Robins and Guze (1970). Within this model, data related to clinical correlates, family history, laboratory studies, treatment response, lifetime course, and outcomes are evaluated to determine if there is a consistent pattern. In their review of the literature, Antshel et al. (2006) noted that while there was limited data related to familial risk, course/outcomes, or genetic/neurobiological mechanisms, there was substantial evidence indicating that established pharmacological and behavioral treatment approaches for ADHD are also efficacious for children with ID and ADHD. Based on this data, they concluded that there was preliminary evidence to support the validity of an ADHD diagnosis in children with ID.

In the past two decades, a growing body of research has provided support for ADHD as a valid diagnosis for individuals with ID. In a sample of children with moderate to borderline intellectual functioning, significant differences in attentional and activity observation measures were found between children with and without elevated levels of ADHD symptoms, even after controlling for cognitive functioning (Handen, McAuliffe, Janosky, Feldman, & Breaux, 1998). This finding demonstrated that individuals with ID present with varying levels of attentional deficits and that these variations do not correlate with intellectual functioning. Additional research has found that children with ID have a similar ADHD symptom presentation compared to typically developing children, both in regard to the number of symptoms and the frequency with which specific symptoms are present (Ahuja, Martin, Langley, & Thapar, 2013; Baker et al., 2010; Neece et al., 2011). A longitudinal examination of ADHD symptoms found a similar developmental trajectory of inattentive and hyperactive/impulsive symptoms between children with and without ID (Neece et al., 2011). Children with ID have also been demonstrated to be at greater risk for ADHD, independent of their chronological or MA, compared to typically developing children (Hastings, Beck, Daley, & Hill, 2005).

Prevalence Estimates

ADHD has been estimated to be three times more prevalent among children and adolescents with ID compared to the general population (Baker et al., 2010; Neece et al., 2011, 2013). A study that assessed for ADHD in 5-year-old children with both ID and typical development through

clinical interviews with caregivers, standardized measures, and clinical observation found that 12.1% of typically developing 5-year-olds met criteria for ADHD, compared to 38.9% of 5-year-olds with ID (Baker et al., 2010). A similar study with 13-year-olds found that only 12% of typically developing participants met criteria for ADHD, compared to 40.5% of those with ID (Neece et al., 2013). Among a sample of adults with ID with an average age of 37.6 years, 19.6% of participants were found to meet ADHD diagnostic criteria based on clinical observation (La Malfa, Lassi, Bertelli, Pallanti, & Albertini, 2008). Further research is needed to estimate the prevalence of ADHD among individuals with ID using population-level data.

Etiology

Research has shown that ADHD has a heterogenous etiology, with both neurological and genetic factors at play, and with various developmental pathways that lead to similar symptom presentation (Nigg, 2012; Nigg & Barkley, 2014). Family and twin studies have shown familial associations of ADHD, with 50–95% of the variation in ADHD traits accounted for by genetic contribution (Barkley, 2014a). Prenatal risk factors associated with ADHD include prenatal exposure to nicotine or alcohol, low birth weight, and maternal phenylalanine levels (Barkley, 2014a). Brain imaging studies have found differences in brain functioning between individuals with and without ADHD, with differences in areas such as the prefrontal cortex, the basal ganglia, and the cerebellum (Barkley, 2014a; Nigg, 2012).

Very little research to date has examined the etiology of ADHD co-occurring with ID. Using population-level data from Sweden, a research study found a significant familial association between ADHD and ID, with 91% of the correlation attributable to genetic factors (Faraone, Ghirardi, Kuja-Halkola, Lichtenstein, & Larsson, 2017). This finding suggests that genetics play an important role in the co-occurrence of ADHD and ID. Results also demonstrated that the comorbidity between ADHD and ID decreased as the severity of an individual's cognitive impairments increased, with a nonsignificant relationship between ADHD symptoms and profound ID, as well as a nonsignificant familial association between ADHD and profound ID. These results may suggest that more severe presentations of ID have different etiologies compared to less severe presentations.

ID is a heterogenous disorder that has been documented to be related to hundreds of different organic disorders (Matson et al., 2019). There may be differences in the etiology and phenotype of ADHD related to specific developmental disorders. Emerging research has examined ADHD symptomology and functioning related to Fragile X syndrome (Cornish, Cole, Longhi, Karmiloff-Smith, & Scerif, 2013), Down syndrome (Breckenridge, Braddick, Anker, Woodhouse, & Atkinson, 2013), and Williams syndrome (Breckenridge et al., 2013). A larger focus within the literature has been the examination of ADHD co-occurring with ASD. High rates of ADHD have been found among individuals diagnosed with ASD, with estimates of comorbidity as high as 59% (Antshel, Zhang-James, & Faraone, 2013; Jang et al., 2013), and findings suggest that comorbid ID/ADHD is associated with increased ADHD symptoms (Hastings et al., 2005). Research has suggested that there are common genetic and neurobiological features underlying ASD and ADHD (Antshel et al., 2013; Visser, Rommelse, Greven, & Buitelaar, 2016).

Clinical Presentation in ID

Unfortunately, little research has been done to date on the clinical presentation of ADHD in individuals with ID. Children and adolescents with ID are often excluded from clinical and treatment studies, and only a handful of studies have directly examined ADHD symptomatology in this population. One such study found that the core symptoms of ADHD tend to have earlier onset for children with ID compared to typically developing children, with 70.6% of children with ID receiving an ADHD diagnosis by age 5, compared to 63.1% of the typically developing group

(Neece et al., 2011). The same study found that ADHD symptoms were more stable over time among children with ID compared to typically developing children (Neece et al., 2011). This finding was echoed by results from a study examining ADHD symptoms in adults with ID, which found that adults with ID and ADHD had more severe ADHD symptoms compared to typically developing adults, with less abatement between childhood and adulthood (Xenitidis, Paliokosta, Rose, Maltezos, & Bramham, 2010). In a large sample of children with ID, Hastings et al. (2005) found little evidence of differences in ADHD symptom presentation or severity between males and females, in contrast to a body of literature documenting gender differences in ADHD symptomatology among typically developing children. Taken together, these findings suggest that for individuals with ID, ADHD symptoms are likely to emerge earlier, be more stable over time, and be more similar between males and females when compared to the general population.

Assessment of ADHD

An assessment for ADHD is a complex process that involves data collection from multiple sources, clinical skill and judgement, and knowledge of a wide range of mental health and developmental disorders to assist with differential diagnosis. This process may be a targeted assessment to determine the presence or absence of ADHD but also may be part of a comprehensive assessment aimed at identifying comorbid psychiatric conditions to guide treatment decision-making. Clinicians preparing for an assessment with a child, adolescent, or adult with ID, or who are suspected of having ID, are strongly encouraged to ensure that they have an adequate understanding of the functional impact and developmental considerations that are associated with cognitive impairments. For more information on the assessment of ID and its impact on the assessment of comorbid disorders, please reference Chap. 15 of this volume. The following section will focus on the assessment of symptoms related to ADHD within this population.

Best practice guidelines for the assessment of ADHD within the general population recommend a multimodal approach (Barkley, 2014c; Nigg & Barkley, 2014; Pelham, Fabiano, & Massetti, 2005; Subcommittee on Attention-Deficit/Hyperactivity Disorder et al., 2011). This typically includes an evaluation of current ADHD symptoms, ADHD symptoms during the developmental period, presence of symptoms across a variety of settings, and the impact of symptoms on functioning based on a combination of clinical interview, direct observation, and rating measures. Generally, this same process can be used with individuals with ID, with a few modifications and considerations.

Important Considerations

The assessment of ADHD in people with ID poses several clinical challenges that should be carefully considered throughout the assessment process. Firstly, ADHD may not be a primary referral concern for individuals with ID. Instead, it is common for individuals with ADHD to have referral concerns related to challenging behaviors, such as physical aggression, noncompliance, or other disruptive behaviors (Perera, 2018). As symptoms of ADHD may be perceived by others as challenging behavior, clinicians performing assessments with individuals with ID should be prepared to assess for the presence of ADHD symptoms even when concerns related to inattention and/or hyperactivity are not directly articulated. Secondly, clinicians should be conscious of potential diagnostic overshadowing of ADHD symptoms and make efforts to avoid attributing any difficulties with attention, hyperactivity, or impulsivity to an individual's cognitive impairments without first evaluating whether the severity of these difficulties is incompatible with an individual's level of developmental functioning. Thirdly, clinicians should be cautious when using and interpreting standardized measures for ADHD within their assessment process, as the vast majority of such measures have been normed with typically developing children and adults.

Assessment Methods

Review of history Every effort should be made to gain access to documentation of a patient's previous medical, educational, psychological, and other service records. Requests for such information can be made prior to the start of an evaluation to aid with assessment planning but can also be requested during the course of an assessment. Review of records can provide valuable information about the patient's previous diagnoses, treatment history, medical conditions, and historical symptoms. Historical records provide an important data source for the evaluation, as direct review of the information bypasses the potential memory errors and other biases in collateral reports of historical information.

Parent/caregiver interview A clinical interview with a parent or caregiver is a key component of an assessment for ADHD for children and adolescents (Barkley, 2014c; Quinlan, 2009). For adults with ID, a parent/caregiver interview is also recommended, as adults with ID may have restrictions in their ability to self-report on symptoms related to ADHD, whether due to limited verbal skills or limited insight into their behavior. Parents and caregivers can provide important information about a patient's symptoms, as well as the environmental contexts associated with behavior problems. While their reports are subjective and based on their personal perspective, parents and caregivers provide important ecologically valid information about a patient's functioning. The reliability of parent/caregiver report can be increased when the clinician poses highly specific questions about symptoms associated with ADHD (Barkley, 2014c).

A parent/caregiver interview should include collection of information related to a patient's general demographics, living situation, and current service provision. Asking parents and caregivers about their concerns early in the interview process can help frame subsequent conversations about specific symptoms and guide assessment planning. Review of major developmental domains, such as motor, language, academic, and social functioning, is important in informing understanding of the patient's developmental history and current functioning level. If records related to medical or previous assessment history were unavailable prior to the parent/caregiver interview, a patient's diagnostic and treatment history should also be obtained with the parent/caregiver. It is also helpful to gather information about a patient's positive characteristics and attributes, which can be valuable in the treatment planning process.

Information related to symptoms of ADHD and other psychiatric disorders should be elicited through targeted questions about specific symptoms. When challenging behaviors are present, a behavior analysis approach is recommended, in which the parent/caregiver is asked to provide information about the topography and frequency of the behavior, as well as common settings, antecedents, and responses from others (O'Neill, Albin, Storey, Horner, & Sprague, 2014). ADHD symptoms should be reviewed and discussed within the context of a variety of contexts, such as daily routines (e.g., meal times, bathing), leisure/play activities, academic tasks or chores, highly preferred activities, low-demand activities (e.g., watching television), in public places, in school/vocational settings, in the car, etc. For assessments with adolescents and adults, clinicians should also be intentional about collecting information about the onset of ADHD and progression of symptoms through childhood and other developmental periods.

Additional collateral interviews Interviews with teachers, support staff, or other services providers can also be conducted. These additional collateral interviews may be helpful in establishing the number of settings in which ADHD symptoms occur and are interfering. For assessments in which a parent/caregiver clinical interview has been conducted, these collateral interviews can be considered secondary sources of information and can be targeted toward collecting information about the patient's functioning in certain contexts. To save time and resources, these secondary collateral interviews

are often conducted via telephone. Interviews with teachers should collect information about the patient's academic performance, the types of difficulties the patient is experiencing, the types of support and accommodations the patient is receiving, and the nature of expectations for the patient's academic performance and behavior in the academic setting. This information is helpful in determining if symptoms of inattention and hyperactivity/impulsivity in the school setting are related to incongruity between expectations and the patient's abilities.

If a parent/caregiver is not available to participate in the assessment process (such as when assessing adults with ID), a comprehensive clinical interview should be conducted with a service provider who knows the patient well and who can speak to his or her functioning both in the present and recent past, gathering information as previously described in relation to parent/caregiver interviews.

Direct observation A portion of the assessment should be spent directly interacting with the patient. General observations should be noted in regard to his/her appearance, behavior, demeanor, social skills, and developmental characteristics. While individuals with ID may not be able to give reliable reports on their own symptoms due to limited verbal abilities or self-awareness, efforts should be made when possible to collect basic information about the patient's living environment, school/vocational setting, social network, emotional state, and any problems they might be experiencing. While not always possible due to time and resource constraints, school observations are very helpful, as they allow for direct observation of ADHD symptoms and off-task behavior in the school setting, as well as of the support and response the patient is receiving from others in that setting.

Assessment of cognitive and academic functioning Having an estimate of the patient's level of cognitive functioning is critical in determining if ADHD symptoms are inconsistent with his/her developmental level. With this in mind, intelligence testing should be included as part of an assessment of ADHD if there are concerns about an individual's cognitive functioning, or if the individual has a previous diagnosis of global developmental delay (GDD) or ID. If cognitive assessment has been completed with the patient recently, prior test results may be used to inform the assessment in lieu of administering a standardized test of intelligence. However, if testing was completed more than 5 years prior, a re-evaluation of cognitive functioning is advised. Standardized intelligence tests should be selected based on consideration of the patient's abilities (e.g., verbal abilities, fine motor skills) in relation to test administration and response methods. Academic achievement testing may also be considered if behavioral concerns are related specifically to the school setting. When administering a standardized test, valuable observational data can be collected in relation to the patient's compliance, disruptive behavior, attention span, and impulse control. However, the absence of ADHD symptoms during test administration should not be used to preclude the possibility that such symptoms occur in other settings with less novel tasks or less individualized support and prompting.

Assessment Tools

Many standardized ADHD assessment tools have been developed and found to have sound psychometric properties when used with typically developing children and adults (Barkley, 2014b, 2014c; Quinlan, 2009). However, when conducting an ADHD assessment with an individual with ID, it is important to be aware of the limitations of these measures for this population. Standardized ADHD measures have been normed with samples of average cognitive functioning, meaning that standardized scores from these measures, when used with an individual with ID, represent a comparison between that individual and an "average" typically developing individual of the same chronological age. This also means that the psychometric properties found in studies

with typically developing samples are not meaningful when these measures are used with individuals with ID (Miller, Fee, & Jones, 2004; Miller, Fee, & Netterville, 2004). The following section reviews commonly used ADHD assessment tools, as well as a brief discussion of their use with individuals with ID. Measures that have been developed specifically for individuals with ID and other developmental delays are also described.

Structured caregiver interviews A number of structured and semi-structured interviews have been developed to guide the clinical interview with a parent/caregiver to inform assessment of a child or adolescent. These interviews typically cover multiple symptom domains related to many different diagnoses. Such measures can be useful, as they provide a framework for systematically gathering information are increased interrater reliability but typically require a lengthy administration time (Leffler, Riebel, & Hughes, 2015). Commonly used structured interviews include the Diagnostic Interview Schedule for Children (DISC-IV; Shaffer, Fisher, Lucas, Dulcan, & Schwab-Stone, 2000) and the Schedule for Affective Disorders and Schizophrenia for School-Aged Children Present and Lifetime Version (K-SADS-PL; Kaufman et al., 1997). Structured and semi-structured interviews may be useful when conducting a comprehensive assessment with a child with ID to guide the parent/caregiver interview, but clinicians should be prepared to rephrase and omit questions as needed to align the interview with the developmental level of the child.

Direct observational measures Standardized measures for direct observation of behavior related to ADHD have been developed for both children and adults. For children aged 2–18, the Achenbach System of Empirically Based Assessment (ASEBA) has developed the Test Observation Form (TOF), designed for use during administration of standardized tests, and the Classroom Observation Form, designed for school observations (McConaughy, Antshel, Gordon, & Eiraldi, 2010). Observers rate the child on 125 items based on direct observation. The measure provides subscale scores related to a number of factors, including attention problems, and also a *DSM*-based ADHD scale. Use of the TOF and Classroom Observation Form may be helpful in the assessment of children with ID, as the results would provide information related to symptom endorsement. The Conners' Adult ADHD Rating Scales Observer Report form (CAARS-O; C. Conners, Erhardt, & Sparrow, 1998) has been used in research with adults with ID (La Malfa et al., 2008). The CAARS-O is designed for use with individuals 18 years and older and is available in long, short, and screening forms and provides standardized scores related to inattention/memory problems, hyperactivity/restlessness, impulsivity/emotional lability, and problems with self-concept.

Performance-based measures A number of performance-based measures are commonly used with the general population to assess for deficits in executive functioning thought to be associated with ADHD. These include computerized continuous performance tests (CPT), tests of memory such as digit span or list learning, as well as the Stroop Color-Word Interference Test (Quinlan, 2009). However, the general utility of such tests in the assessment of ADHD has been disputed (Barkley, 2014b). These tests are very unlikely to be useful in the assessment of ADHD with individuals with ID, as not only have they not been validated with this population, but they also often rely on complex verbal instructions, which would make it difficult to conclude if deficits revealed through such tests were related to attention specifically.

Self-report rating scales Self-report rating scales are commonly used in the assessment of ADHD amongst typically developing adolescents and adults (Barkley, 2014b, 2014c; Quinlan, 2009). However, self-rating scales are unlikely to be useful when assessing individual with ID, as

they require good insight into one's behavior, as well as a certain level of reading comprehension.

Parent/teacher rating scales Parent and teacher rating scales are one of the most frequently used tools in the assessment of ADHD in typically developing children and adolescents (Barkley, 2014c; Pelham et al., 2005; Quinlan, 2009). While the validity of parent/teacher rating scales with the ID population is still unknown, research studies with children with ID frequently use some of the most popular measures. Commonly used are broadband rating scales, which assess across multiple symptom areas. The Achenbach System of Empirically Based Assessment (ASEBA) includes the Child Behavior Checklist (CBCL) as well as the Teacher Report Forms (TRF), with forms for ages 1.5–18 (Achenbach & Rescorla, 2000, 2001). The Behavior Assessment System for Children, Third Edition (BASC-3) is another commonly used system, which includes parent and teacher forms for ages 2–21 years (Reynolds & Kamphaus, 2015). Rating scales specific to ADHD have also been used frequently in research with children with ID: the Conners' Third Edition system includes the Conners' 3 Parent (Conners 3-P) and Conners' 3 Teacher (Conners-3-T) forms for ages 6–18 (Conners, 2008), and the National Institute for Children's Health Quality (NICHQ) Vanderbilt Assessment Scales is available for ages 6–12 (Wolraich et al., 2003). While these rating scales may provide useful information, results should be interpreted with caution. Research examining the use of the Conners' rating scales with children with ID has found that the parent rating scale was able to distinguish between children with and without co-occurring ADHD, while the teacher rating scale did not (Deb, Dhaliwal, & Roy, 2008).

Measures for individuals with ID A couple of rating scales have been developed specifically for use with developmental delays. The Aberrant Behavior Checklist – Second Edition (ABC-2) is a widely used symptom checklist used to assess for a number of problem behaviors for children and adults with developmental disabilities (Aman & Singh, 2017). It includes 58 items that are rated and resolved into five subscales: irritability, social withdrawal, stereotypic behavior, hyperactivity/noncompliance, and inappropriate speech. Previous versions of the ABC have been found to have sound psychometric properties with individuals with ASD and ID (Kaat, Lecavalier, & Aman, 2014; Rojahn, Aman, Matson, & Mayville, 2003; Schmidt, Huete, Fodstad, Chin, & Kurtz, 2013). The ABC-2 includes forms for individuals who live in community and residential settings and is designed for ages 5 to adult.

The Scale of Attention in Intellectual Disability – Teacher version (T-SAID) is a teacher rating measure that was developed specifically to assess for ADHD symptoms in children and adolescents with ID (Freeman, Gray, Taffe, & Cornish, 2015, 2016). The T-SAID includes 44 items that compose two scales: hyperactivity and impulsivity. This measure purposefully avoided items that assume literacy and numeracy skills, which are commonly referenced in other teacher rating scales for ADHD and may not be appropriate for students with ID. Preliminary research on the psychometric properties of the T-SAID is promising; however, its efficacy as a screening tool has not yet been fully evaluated (Freeman et al., 2016). At the time of publishing, the T-SAID was not yet distributed for general use.

Integration of Data

Integration of data is an ongoing part of the assessment process. Data from each source (e.g., clinical interviews, direct observation, rating scales, standardized assessment) should be weighed and compared. Diagnostic criteria for ADHD should be carefully reviewed in relation to symptom endorsement, age of onset of symptoms, number of settings in which symptoms are present, and the associated degree of impairment

in social, academic, or occupational settings. Careful consideration should be given to whether the number and severity of ADHD symptoms exceed that which would be expected based on the patient's level of developmental and cognitive functioning.

Utility of an ADHD Diagnosis

Assessment is generally considered the first step in the treatment process. For an individual with ID, a dual diagnosis of ADHD may prove instrumental in addressing behavioral concerns and decreasing functional impairments by guiding intervention. Parents and caregivers of individuals with ID, as well as teachers and support staff working with these individuals, may find psychoeducation about ADHD to be helpful in understanding an individual's behaviors and needs and helpful for behavior management. Behavioral and pharmacological treatments for ADHD have been found to be efficacious for individuals with ID and co-occurring ID (Antshel et al., 2006; Evans & Trollor, 2016). Treatment planning should be based on assessment findings related to an individual's unique needs and strengths. For more information about treating ADHD symptoms in this population, please see Chap. 34 of this volume, which addresses this topic in detail.

Future Directions

There is currently a limited amount of information about ADHD co-occurring with ID. Further research is required to examine the prevalence of ADHD among children, adolescents, and adults with ID using population-level data. Research is sorely needed to establish baseline rates of ADHD symptoms among individuals with ID. This work will be valuable in norming existing measures for individuals with ID as well as developing measures specifically designed for this population. Studies are also needed to identify risk factors for ADHD in children with developmental delays. Additionally, more research is needed to understand the clinical presentation of ADHD in this population across the life span and the etiology of comorbid ID/ADHD.

Conclusions

ADHD is a common comorbid condition for individuals with ID, with prevalence rates three times higher compared to the general population. While symptoms of inattention and hyperactivity among individuals with ID have previously been assumed to be inherently related to their cognitive deficits, a growing body of research has supported the validity of a co-occurring ADHD diagnosis. Assessment for ADHD in children, adolescents, and adults with ID should incorporate information from multiple sources, such as direct observation, clinical interviews, and rating scales. Particular attention should be paid through the assessment process to whether symptoms are inconsistent with an individual's level of developmental functioning. A co-occurring diagnosis of ADHD can provide valuable guidance in treatment planning.

References

Achenbach, T. M., & Rescorla, L. A. (2000). *Manual for the Achenbach system of empirically based assessment preschool forms profiles*. Burlington, VT: ASEBA.

Achenbach, T. M., & Rescorla, L. A. (2001). *Manual for the Achenbach system of empirically based assessment school-age forms profiles*. Burlington, VT: ASEBA.

Adler, L. A., Spencer, T. J., Stein, M. A., & Newcorn, J. H. (2008). Best practices in adult ADHD: Epidemiology, impairments, and differential diagnosis. *CNS Spectrums, 13*(S12), 18–18. https://doi.org/10.1017/S1092852900003217

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.e1. https://doi.org/10.1016/j.jpeds.2013.02.043

Aman, M. G., & Singh, N. N. (2017). *Aberrant behavior checklist* (2nd ed.). Wood Dale, IL: Stoelting.

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

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

American Psychiatric Association. (1987). *Diagnostic and statistical manual of mental disorders* (3rd ed.-Revised). Washington, D.C.: American Psychiatric Association.

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

American Psychiatric Association. (2000). *Diagnostic and statistical manual of mental disorders* (4th ed.-Text Revision). Washington, D.C.: American Psychiatric Association.

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

Antshel, K. M., Phillips, M. H., Gordon, M., Barkley, R., & Faraone, S. V. (2006). Is ADHD a valid disorder in children with intellectual delays? *Clinical Psychology Review, 26*(5), 555–572. https://doi.org/10.1016/j.cpr.2006.03.002

Antshel, K. M., Zhang-James, Y., & Faraone, S. V. (2013). The comorbidity of ADHD and autism spectrum disorder. *Expert Review of Neurotherapeutics, 13*(10), 1117–1128. https://doi.org/10.1586/14737175.2013.840417

Baio, J., Wiggins, L., Christensen, D. L., Maenner, M. J., Daniels, J., Warren, Z., … Dowling, N. F. (2018). Prevalence of autism spectrum disorder among children aged 8 years – Autism and Developmental Disabilities Monitoring Network, 11 Sites, United States, 2014. *MMWR Surveillance Summaries, 67*(6), 1–23.

Baker, B. L., Neece, C. L., Fenning, R. M., & Blacher, J. (2010). Mental disorders in five-year-old children with or without developmental delay: Focus on ADHD. *Journal of Clinical Child & Adolescent Psychology, 39*(4), 492–505. https://doi.org/10.1080/15374416.2010.486321

Barkley, R. A. (2014a). Etiologies of ADHD. In R. A. Barkley (Ed.), *Attention-deficit hyperactivity disorder, Fourth edition: A handbook for diagnosis and treatment* (pp. 356–390). New York, NY: Guilford Publications.

Barkley, R. A. (2014b). Psychological assessment of adults with ADHD. In R. A. Barkley (Ed.), *Attention-deficit hyperactivity disorder, Fourth edition: A handbook for diagnosis and treatment* (pp. 475–500). New York, NY: Guilford Publications.

Barkley, R. A. (2014c). Psychological assessment of children with ADHD. In R. A. Barkley (Ed.), *Attention-deficit hyperactivity disorder, Fourth edition: A handbook for diagnosis and treatment* (pp. 455–474). New York, NY: Guilford Publications.

Barkley, R. A., Copeland, A. P., & Sivage, C. (1980). A self-control classroom for hyperactive children. *Journal of Autism and Developmental Disorders, 10*(1), 75–89. https://doi.org/10.1007/BF02408435

Barkley, R. A., Murphy, K. R., & Fischer, M. (2010). *ADHD in adults: What the science says*. New York, NY: Guilford Press.

Beike, S. M., & Zentall, S. S. (2012). "The snake raised its head": Content novelty alters the reading performance of students at risk for reading disabilities and ADHD. *Journal of Educational Psychology, 104*(3), 529–540. https://doi.org/10.1037/a0027216

Breckenridge, K., Braddick, O., Anker, S., Woodhouse, M., & Atkinson, J. (2013). Attention in Williams syndrome and Down's syndrome: Performance on the new early childhood attention battery. *British Journal of Developmental Psychology, 31*(2), 257–269. https://doi.org/10.1111/bjdp.12003

Burack, J. A., Evans, D. W., Klaiman, C., & Iarocci, G. (2001). The mysterious myth of attention deficits and other defect stories: Contemporary issues in the developmental approach to mental retardation. In *International review of research in mental retardation* (Vol. 24, pp. 299–320). https://doi.org/10.1016/S0074-7750(01)80012-4

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.

Clark, B., & Bélanger, S. A. (2018). ADHD in children and youth: Part 3—Assessment and treatment with comorbid ASD, ID, or prematurity. *Paediatrics & Child Health, 23*(7), 485–490. https://doi.org/10.1093/pch/pxy111

Conners, C., Erhardt, D., & Sparrow, E. (1998). *The Conners Adult ADHD Rating Scale (CAARS)*. Toronto, Canada: Multi-Health Systems.

Conners, C. K. (2008). *Conners comprehensive behavior rating scales manual*. North Tonawanda, NY: Multi-Health Systems.

Cornish, K., Cole, V., Longhi, E., Karmiloff-Smith, A., & Scerif, G. (2013). Mapping developmental trajectories of attention and working memory in Fragile X syndrome: Developmental freeze or developmental change? *Development and Psychopathology, 25*(2), 365–376. https://doi.org/10.1017/S0954579412001113

Danielson, M. L., Bitsko, R. H., Ghandour, R. M., Holbrook, J. R., Kogan, M. D., & Blumberg, S. J. (2018). Prevalence of parent-reported ADHD diagnosis and associated treatment among U.S. children and adolescents, 2016. *Journal of Clinical Child & Adolescent Psychology, 47*(2), 199–212. https://doi.org/10.1080/15374416.2017.1417860

Deb, S., Dhaliwal, A.-J., & Roy, M. (2008). The usefulness of Conners' Rating Scales-Revised in screening for Attention Deficit Hyperactivity Disorder in children with intellectual disabilities and borderline intelligence. *Journal of Intellectual Disability Research, 52*(11), 950–965. https://doi.org/10.1111/j.1365-2788.2007.01035.x

Deutsch, C. K., Dube, W. V., & McIlvane, W. J. (2008). Attention deficits, attention-deficit hyperactivity disorder, and intellectual disabilities. *Developmental Disabilities Research Reviews, 14*(4), 285–292. https://doi.org/10.1002/ddrr.42

Epstein, J. N., & Loren, R. E. A. (2013). Changes in the definition of ADHD in DSM-5: Subtle but important. *Neuropsychiatry, 3*(5), 455–458. https://doi.org/10.2217/npy.13.59

Evans, E., & Trollor, J. (2016). Attention-deficit/hyperactivity disorder (ADHD). In C. Hemmings & N. Bouras (Eds.), *Psychiatric and behavioral disorders in intellectual and developmental disabilities* (3rd ed., pp. 129–138). Cambridge, UK: Cambridge University Press. https://doi.org/10.1017/CBO9781107588714.013

Faraone, S. V., Ghirardi, L., Kuja-Halkola, R., Lichtenstein, P., & Larsson, H. (2017). The familial co-aggregation of attention-deficit/hyperactivity disorder and intellectual disability: A register-based family study. *Journal of the American Academy of Child & Adolescent Psychiatry, 56*(2), 167–174.e1. https://doi.org/10.1016/j.jaac.2016.11.011

Fletcher, R. J., & Cooper, S.-A. (2017). *Diagnostic manual-Intellectual disability 2 (DM-ID): A textbook of diagnosis of mental disorders in persons with intellectual disability*. Kingston, NY: NADD Press.

Freeman, N. C., Gray, K. M., Taffe, J. R., & Cornish, K. M. (2015). Development of a new attention rating scale for children with intellectual disability: The Scale of Attention in Intellectual Disability (SAID). *American Journal on Intellectual and Developmental Disabilities, 120*(2), 91–109. https://doi.org/10.1352/1944-7558-120.2.91

Freeman, N. C., Gray, K. M., Taffe, J. R., & Cornish, K. M. (2016). A cross-syndrome evaluation of a new attention rating scale: The Scale of Attention in Intellectual Disability. *Research in Developmental Disabilities, 57*, 18–28. https://doi.org/10.1016/j.ridd.2016.06.005

Handen, B. L., McAuliffe, S., Janosky, J., Feldman, H., & Breaux, A. M. (1998). A playroom observation procedure to assess children with mental retardation and ADHD. *Journal of Abnormal Child Psychology, 26*(4), 269–277. https://doi.org/10.1023/A:1022654417460

Hastings, R. P., Beck, A., Daley, D., & Hill, C. (2005). Symptoms of ADHD and their correlates in children with intellectual disabilities. *Research in Developmental Disabilities, 26*(5), 456–468. https://doi.org/10.1016/j.ridd.2004.10.003

Iarocci, G., & Burack, J. A. (1998). Understanding the development of attention in persons with mental retardation: Challenging the myths. In *Handbook of mental retardation and development* (pp. 349–381). New York, NY: Cambridge University Press.

Jang, J., Matson, J. L., Williams, L. W., Tureck, K., Goldin, R. L., & Cervantes, P. E. (2013). Rates of comorbid symptoms in children with ASD, ADHD, and comorbid ASD and ADHD. *Research in Developmental Disabilities, 34*(8), 2369–2378. https://doi.org/10.1016/j.ridd.2013.04.021

Jopp, D. A., & Keys, C. B. (2001). Diagnostic overshadowing reviewed and reconsidered. *American Journal of Mental Retardation, 106*(5), 416–433. https://doi.org/10.1352/0895-8017(2001)106<0416:DORAR>2.0.CO;2

Kaat, A. J., Lecavalier, L., & Aman, M. G. (2014). Validity of the Aberrant Behavior Checklist in children with autism spectrum disorder. *Journal of Autism and Developmental Disorders, 44*(5), 1103–1116. https://doi.org/10.1007/s10803-013-1970-0

Kaufman, J., Birmaher, B., Brent, D., Rao, U., Flynn, C., Moreci, P., … Ryan, N. (1997). Schedule for Affective Disorders and Schizophrenia for School-Age Children-Present and Lifetime Version (K-SADS-PL): Initial reliability and validity data. *Journal of the American Academy of Child & Adolescent Psychiatry, 36*(7), 980–988. https://doi.org/10.1097/00004583-199707000-00021

Kessler, R. C., Adler, L., Barkley, R., Biederman, J., Conners, C. K., Demler, O., … Zaslavsky, A. M. (2006). The prevalence and correlates of adult ADHD in the United States: Results from the National Comorbidity Survey Replication. *American Journal of Psychiatry, 163*(4), 716–723. https://doi.org/10.1176/ajp.2006.163.4.716

La Malfa, G., Lassi, S., Bertelli, M., Pallanti, S., & Albertini, G. (2008). Detecting attention-deficit/hyperactivity disorder (ADHD) in adults with intellectual disability: The use of Conners' Adult ADHD Rating Scales (CAARS). *Research in Developmental Disabilities, 29*(2), 158–164. https://doi.org/10.1016/j.ridd.2007.02.002

Lange, K. W., Reichl, S., Lange, K. M., Tucha, L., & Tucha, O. (2010). The history of attention deficit hyperactivity disorder. *Attention Deficit and Hyperactivity Disorders, 2*(4), 241–255. https://doi.org/10.1007/s12402-010-0045-8

Leffler, J. M., Riebel, J., & Hughes, H. M. (2015). A review of child and adolescent diagnostic interviews for clinical practitioners. *Assessment, 22*(6), 690–703. https://doi.org/10.1177/1073191114561253

Mahjouri, S., & Lord, C. E. (2012). What the DSM-5 portends for research, diagnosis, and treatment of Autism Spectrum Disorders. *Current Psychiatry Reports, 14*(6), 739–747. https://doi.org/10.1007/s11920-012-0327-2

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. https://doi.org/10.1111/j.1360-2322.2004.00184.x

Matson, J. L., Matheis, M., Estabillo, J. A., Issarraras, A., Peters, W. J., & Jiang, X. (2019). Intellectual disability. In M. J. Prinstein, E. A. Youngstrom, E. J. Mash, & R. A. Barkley (Eds.), *Treatment of disorders in childhood and adolescence* (4th ed., pp. 416–447). New York, NY: Guilford Publications.

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

McConaughy, S. H., Antshel, K. M., Gordon, M., & Eiraldi, R. B. (2010). Observational Assessment of

ADHD with the ASEBA forms. *The ADHD Report, 18*(6), 3–8. https://doi.org/10.1521/adhd.2010.18.6.3

Miller, M. L., Fee, V. E., & Jones, C. J. (2004). Psychometric properties of ADHD rating scales among children with mental retardation II: Validity. *Research in Developmental Disabilities, 25*(5), 477–492. https://doi.org/10.1016/j.ridd.2003.11.002

Miller, M. L., Fee, V. E., & Netterville, A. K. (2004). Psychometric properties of ADHD rating scales among children with mental retardation I: Reliability. *Research in Developmental Disabilities, 25*(5), 459–476. https://doi.org/10.1016/j.ridd.2003.11.003

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. https://doi.org/10.1111/j.1365-2788.2011.01416.x

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. https://doi.org/10.1007/s10802-012-9698-4

Nigg, J. T. (2012). Future directions in ADHD etiology research. *Journal of Clinical Child & Adolescent Psychology, 41*(4), 524–533. https://doi.org/10.1080/15374416.2012.686870

Nigg, J. T., & Barkley, R. A. (2014). Attention-deficit/hyperactivity disorder. In E. J. Mash & R. A. Barkley (Eds.), *Child psychopathology* (3rd ed., pp. 75–144). New York, NY: Guilford Publications.

O'Neill, R. E., Albin, R. W., Storey, K., Horner, R. H., & Sprague, J. R. (2014). *Functional assessment and program development for problem behavior: A practical handbook* (3rd ed.). Stamford, CT: Cengage Learning.

Owens, E. B., Cardoos, S. L., & Hinshaw, S. P. (2014). Developmental progression and gender difference among individuals with ADHD. In R. A. Barkley (Ed.), *Attention-deficit hyperactivity disorder, Fourth edition: A handbook for diagnosis and treatment* (pp. 223–255). New York, NY: Guilford Publications.

Pelham, W. E., Fabiano, G. A., & Massetti, G. M. (2005). Evidence-based assessment of attention deficit hyperactivity disorder in children and adolescents. *Journal of Clinical Child & Adolescent Psychology, 34*(3), 449–476. https://doi.org/10.1207/s15374424jccp3403_5

Perera, B. (2018). Attention deficit hyperactivity disorder in people with intellectual disability. *Irish Journal of Psychological Medicine, 35*(3), 213–219. https://doi.org/10.1017/ipm.2018.7

Polanczyk, G., de Lima, M. S., Horta, B. L., Biederman, J., & Rohde, L. A. (2007). The worldwide prevalence of ADHD: A systematic review and metaregression analysis. *American Journal of Psychiatry, 164*(6), 942–948. https://doi.org/10.1176/ajp.2007.164.6.942

Quinlan, D. M. (2009). Assessment of ADHD and comorbidities. In T. E. Brown (Ed.), *ADHD comorbidities: Handbook for ADHD complications in children and adults* (pp. 317–338). Arlington, VA: American Psychiatric Publishing.

Reynolds, C. R., & Kamphaus, R. W. (2015). *Behavior assessment system for children* (3rd ed.). San Antonio, TX: Pearson.

Roberts, W., Milich, R., & Barkley, R. A. (2014). Primary symptoms, diagnostic criteria, subtyping, and prevalence of ADHD. In R. A. Barkley (Ed.), *Attention-deficit hyperactivity disorder, Fourth edition: A handbook for diagnosis and treatment* (pp. 51–80). New York, NY: Guilford Publications.

Robins, E., & Guze, S. B. (1970). Establishment of diagnostic validity in psychiatric illness: Its application to schizophrenia. *American Journal of Psychiatry, 126*(7), 983–987. https://doi.org/10.1176/ajp.126.7.983

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*(5), 391–404. https://doi.org/10.1016/S0891-4222(03)00055-6

Schmidt, J. D., Huete, J. M., Fodstad, J. C., Chin, M. D., & Kurtz, P. F. (2013). An evaluation of the Aberrant Behavior Checklist for children under age 5. *Research in Developmental Disabilities, 34*(4), 1190–1197. https://doi.org/10.1016/j.ridd.2013.01.002

Shaffer, D., Fisher, P., Lucas, C. P., Dulcan, M. K., & Schwab-Stone, M. E. (2000). NIMH Diagnostic Interview Schedule for Children Version IV (NIMH DISC-IV): Description, differences from previous versions, and reliability of some common diagnoses. *Journal of the American Academy of Child and Adolescent Psychiatry, 39*(1), 28–38. https://doi.org/10.1097/00004583-200001000-00014

Subcommittee on Attention-Deficit/Hyperactivity Disorder, Steering Committee on Quality Improvement and Management, Wolraich, M., Brown, L., Brown, R. T., DuPaul, G., … Visser, S. (2011). ADHD: Clinical practice guideline for the diagnosis, evaluation, and treatment of attention-deficit/hyperactivity disorder in children and adolescents. *Pediatrics, 128*(5), 1007–1022. https://doi.org/10.1542/peds.2011-2654

Surman, C. B. H., & Goodman, D. W. (2017). Is ADHD a valid diagnosis in older adults? *ADHD Attention Deficit and Hyperactivity Disorders, 9*(3), 161–168. https://doi.org/10.1007/s12402-017-0217-x

Tonge, B. J., Einfeld, S. L., Krupinski, J., Mackenzie, A., McLaughlin, M., Florio, T., & Nunn, R. J. (1996). The use of factor analysis for ascertaining patterns of psychopathology in children with intellectual disability. *Journal of Intellectual Disability Research, 40*(3), 198–207. https://doi.org/10.1111/j.1365-2788.1996.tb00623.x

Visser, J. C., Rommelse, N. N. J., Greven, C. U., & Buitelaar, J. K. (2016). Autism spectrum disorder and attention-deficit/hyperactivity disorder in early childhood: A review of unique and shared characteristics and developmental antecedents. *Neuroscience*

& Biobehavioral Reviews, 65, 229–263. https://doi.org/10.1016/j.neubiorev.2016.03.019

Weyandt, L. L., & Gudmundsdottir, B. G. (2014). Developmental and neuropsychological deficits in children with ADHD. In R. A. Barkley (Ed.), *Attention-deficit hyperactivity disorder, Fourth edition: A handbook for diagnosis and treatment* (pp. 116–139). New York, NY: Guilford Publications.

Wolraich, M. L., Lambert, W., Doffing, M. A., Bickman, L., Simmons, T., & Worley, K. (2003). Psychometric properties of the Vanderbilt ADHD diagnostic parent rating scale in a referred population. *Journal of Pediatric Psychology, 28*(8), 559–567. https://doi.org/10.1093/jpepsy/jsg046

World Health Organization. (2018). *International statistical classification of diseases and related health problems* (11th Revision). Retrieved from https://icd.who.int/browse11/l-m/en

Xenitidis, K., Paliokosta, E., Rose, E., Maltezos, S., & Bramham, J. (2010). ADHD symptom presentation and trajectory in adults with borderline and mild intellectual disability. *Journal of Intellectual Disability Research, 54*(7), 668–677. https://doi.org/10.1111/j.1365-2788.2010.01270.x

Zigler, E. (1969). Developmental versus difference theories of mental retardation and the problem of motivation. *American Journal of Mental Deficiency, 73*(4), 536–556.

Substance Abuse in Dual Diagnosis

18

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Definition of Substance Abuse

What is substance abuse and what is not? This question has gained increasing attention over the last several years. Diagnostic manuals and screening tools remain equivocal in defining what constitutes substance abuse and what level substance abuse quantifies as a problem in modern society. With terms such as "tolerance," "dependence," "addiction," "overdosing," and "substance abuse problem" used interchangeably to highlight substance abuse problem (Bozzelli, 2008), what constitutes substance abuse problem needs to be conceptualized; thus, this chapter comes to two conclusions.

First, the American Psychiatric Association formulated the fifth edition of the *Diagnostic and Statistical Manual of Mental Disorders*(DSM-5) to better characterize behavioral symptoms of groups of people seeking clinical help. The terms "substance abuse" and "substance dependence" are no longer used; rather terms are substituted with substance use disorders, "which are defined as mild, moderate, or severe to indicate the level of severity." The number of diagnostic criteria met by an individual determines the level of severity (Substance Abuse and Mental Health Services Administration, 2015). DSM-5, a diagnosis of substance use disorder, is based on evidence of impaired control, social impairment, risky use, and pharmacological criteria.

Second, the tenth revision of the International Statistical Classification of Diseases and Related Health Problems (ICD-10) copyrighted by the World Health Organization defines substance abuse as the use of harmful or hazardous psychoactive substances, including alcohol and illicit drugs. The use of psychoactive substances can lead to dependence syndrome characterized by a cluster of behavioral, cognitive, and physiological phenomena that develop after repeated substance use (World Health Organization, 2018). Dependence syndrome includes a strong desire to take the drug, difficulties in controlling its use, persisting in its use despite harmful consequences, a higher priority given to drug use than to other activities and obligations, increased tolerance, and sometimes a physical withdrawal state (World Health Organization, 2018). To illustrate the point and underscore the terms "substance abuse and substance use disorders," we need to consider the striking consistency in

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both definitions: harmful use of pharmacological and non-pharmacological substances that alters brain function leading to impairment in perception, mood, consciousness, cognition, or behavior.

Common Substance Abused and Their Description

Let us recall that as discussed above, we learned that psychoactive substances are hazardous and lead to dependence. This section summarizes the common substance abused:

- Tobacco and tobacco products: Tobacco products include cigarettes, loose tobacco in a pipe or hookah, cigars, bidis, and kreteks. Chewed tobacco products include chewing tobacco, snuff, dip, and snus; snuff can also be sniffed (National Institute on Drug Abuse (NIDA), 2018b).
- Alcohol known in its chemical form as ethanol or ethyl alcohol is a psychoactive substance found in beer, wine, distilled spirits (hard liquor), and beverages (Collins & Kirouac, 2013). According to the NIDA (2018a), alcohol is the most commonly abuse substance and notably a social propriety, but then individuals who consume excessive amounts of alcohol are at risk of developing serious health problems in addition to issues associated with intoxication behaviors and alcohol withdrawal symptoms.
- Cannabis (other names: marijuana, weed, and pot) is the second most commonly abused substance after alcohol. Cannabis is a depressant drug, which means it slows down messages travelling between your brain and your body. Its effect is fascinating as it varies from one person to another. Cannabis affects everyone differently, and its effects fall under three categories: depressant, opioid, and hallucinogen.
- Opioids are derived from opium that comes from the poppy plant. Opioids are a group of drugs primarily used for pain relief. Opioids include illegal drug heroin, synthetic opioids such as fentanyl (Duragesic), and pain relievers available legally by prescription such as oxycodone (OxyContin, Percocet), hydrocodone (Vicodin, Hycodan), codeine, morphine (MS Contin, Kadian), and hydromorphone (Dilaudid) (Schiller & Mechanic, 2018). Dextromethorphan (DXM), a cough suppressant, and loperamide, an antidiarrheal, are over-the-counter opioids commonly misused by young people. DXM and loperamide cause euphoric and depressant and hallucinogenic effects when taken in large doses (NIDA, 2018a, 2018b).
- Hallucinogens are a diverse group of drugs that alter perception, thoughts, and feelings. Hallucinogens also cause hallucinations or sensations and distortions when viewing images. Hallucinogens, such as MDMA (3,4-methylenedioxymethamphetamine) or ecstasy, chemically synthesized (as with lysergic acid diethylamide (LSD)) or may occur naturally (as with psilocybin mushrooms, peyote) (NIDA, 2016, 2020; Wu, Ringwalt, Mannelli, & Patkar, 2008).
- Inhalants refer to the various dangerous substances that have psychoactive properties when inhaled. These substances include solvents (liquids that become gas at room temperature), aerosol sprays, gases, and nitrites (e.g., prescription medicines for chest pain, leather cleaner).
- Depressants include sedatives, tranquilizers, and hypnotics. Sedatives include barbiturates used to treat acute anxiety, tension, and sleep disorders (e.g., phenobarbital and mephobarbital). Tranquilizers including benzodiazepines such as Xanax and valium are used in treating anxiety, insomnia, seizure, and panic attacks. Depressants slow brain activity, which can cause drowsiness, slurred speech, poor concentration, confusion, dizziness, problems with movement and memory, lowered blood pressure, and slowed breathing, especially when misused (NIDA, 2018a, 2018b).
- Stimulants include methylphenidate (Ritalin®, Concerta®), methamphetamines (Adderall®, Dexedrine®), and cocaine. They have been used to treat attention deficit hyperactivity disorder, narcolepsy, and, sometimes,

depression (Lakhan & Kirchgessner, 2012). According to the Substance Abuse and Mental Health Services Administration (SAMHSA, 2016) stimulants increase alertness, attention, and energy, as well as elevate blood pressure, heart rate, and respiration. Like other prescription medications, stimulants can be diverted for illegal use.

Epidemiology of Substance Abuse

Epidemiological findings of substance abuse have provided enormous data on the patterns of substance abuse in nationally representative samples across global regions (Merikangas & McClair, 2012).

Prevalence and Incidence of Substance Abuse Globally The abuse of substances poses a significant threat to the social, economic, and physical health of families and communities all over the world. An estimated 5.3% of the global population use illegal substances in 2015. An analysis of data obtained from the World Health Organization, United Nations Office on Drugs and Crime, and Institute for Health Metrics and Evaluation reports that in 2015, the estimated prevalence of alcohol, tobacco, and illicit substance abuse among the adult population was 18.4% for heavy episodic alcohol use, 15.2% for daily tobacco smoking, 3.8 for cannabis use, 0.77 for amphetamine use, 0.37 for opioid use, and 0.35% for cocaine use (Peacock et al., 2018). An epidemiological, cross-sectional study was conducted in Lucknow, India, in 2014 with a total of 3437 participants, out of which 82.9% were male and 17.1% were female. Smokeless tobacco was the substance with highly prevalent consumption rate in the population surveyed (Kumar et al., 2015).

In 2015, Europeans and Americans suffered proportionately more, but the mortality rate is highest in low- and middle-income countries with large populations where the quality of data was more limited. The problem of substance abuse also varies among countries as different countries practice different substance abuse laws, enforce different drug penalties for breaking these laws, experience varying availabilities of substances, and possess unique cultural perspectives on specific substances. European regions had the highest prevalence of heavy episodic alcohol use and daily tobacco use. High-income North America region had among the highest prevalence rates of cannabis, opioid, and cocaine dependence. When the attributable disability-adjusted life-years (DALYs), that is, the years of life lost due to premature mortality and the years of life lost due to living with disability, was calculated, the highest was tobacco smoking (170.9 million DALYs), followed by alcohol (85.0 million) and use of illicit drugs (27.8 million) (Peacock et al., 2018). Tobacco smoking has the highest substance-attributable mortality rate (110.7 deaths per 100,000 people), followed by alcohol and illicit drugs (33.0 and 6.9 deaths per 100,000 people, respectively). Substance use is estimated at 2 billion alcohol users, 1.3 billion smokers, and 185 million illicit drug users (Peacock et al., 2018; World Health Organization, 2018).

Prevalence and Incidence of Substance Abuse in the United States The US National Institute on Drug Abuse (NIDA) uses a variety of sources of information to monitor the prevalence and trends regarding drug abuse in the United States. Substance abuse in the United States is currently at its highest point since 2008. Approximately 1 in 3 (30%) of the addicted population are women mostly of childbearing age between 15 and 44 (Wendell, 2013). The United States spends more than $740 billion annually in substance abuse costs related to crime, lost work productivity, and health care. About 10.6% of Americans with substance use disorder seek treatment, and 40–60% of those people relapse within a year. Since 1999, opiate overdose deaths have increased 265% among men and 400% among women. The number of people who abuse substances varies by type. An estimate of 119 million people use marijuana and about 14.5 million use methamphetamine at some point in their life. An increase in the use of marijuana would be

unsurprising given the recent legalization in 30 states and rise in public support. In 2013, 6.5 million Americans aged 12 or older (or 2.5%) had used prescription drugs nonmedically in the past month. Prescription drugs include pain relievers, tranquilizers, stimulants, and sedatives. And 1.3 million Americans (0.5%) had used hallucinogens (a category that includes ecstasy and LSD) in the past month. Substance abuse is increasing among people in their 50s and early 60s. This increase is, in part, due to the aging of the baby boomers, whose rates of illicit drug use have historically been higher than those of previous generations (NIDA, 2018).

The prevalence of any illicit drugs among children from 8th through 12th grade remains stable between 1991 to 2019. However, prevalence spiked to 43.3% in 1997 from 30.4% in 1991 and dropped back to 34.8% in 2019 (Statista, 2020). Drug use is highest among people in their late teens and 20s. In 2013, 22.6% of 18- to 20-year-olds reported using an illicit drug, mostly marijuana in the past month. The data from 72,561 youths interviewed by the National Survey on Drug Use and Health (2011) Please add the reference in list. I will provide it in email because I am unable to add this in list reports that 37% of those aged between 12 and 17 had used alcohol or other drugs at least once in the past year. Nearly 8% met the criteria for a substance use disorder, either the less severe "substance abuse" diagnosis or the more problematic "substance dependence," which is more commonly known as addiction.

The study by Wu, Woody, Yang, Pan, and Blazer (2011) controlled for variables like socioeconomic status because rates of severe drug problems tend to be greater among the poor. Despite this, Native American youth have the worst with 15% having a substance use disorder, compared to 9.2% for people of mixed racial heritage, 9.0% for Whites, 7.7% for Hispanics, 5% for African Americans, and 3.5% for Asians and Pacific Islanders. Analgesic opioids were the second most commonly used illegal drugs, following marijuana, in all racial/ethnic groups; analgesic opioid use was comparatively prevalent among adolescents of Native American (9.7%) and multiple race/ethnicity (8.8%). Among 27,705 past year alcohol or drug users, Native Americans (31.5%), adolescents of multiple race/ethnicity (25.2%), adolescents of White race/ethnicity (22.9%), and Hispanics (21.0%) had the highest rates of substance-related disorders.

The 2002–2014 National Surveys on Drug Use and Health highlights the number of American adults with substance use disorders (SUDs) – which includes substance abuse or dependence data (Lipari & Van Horn, 2017). Approximately 20.2 million adults aged 18 or older had a past year SUD. Of these adults, 16.3 million had an alcohol use disorder, and 6.2 million had an illicit drug use disorder. The percentage of adults with a past year SUD in 2014 was similar to the percentages in 2010–2013 but was lower than the percentages in 2002–2009. In 2014, approximately 3.5 million adults had a past year disorder related to their use of marijuana, and 1.8 million adults had a disorder related to their nonmedical use of prescription opioids (Figs. 18.1 and 18.2).

The past several decades have revealed varied differences in substance abuse among ethnicities as the influence of acculturation, social-structural, family structure, gender, and sociodemographic factors reflects the substance most likely abused. In 2012–2013, the National Epidemiologic Survey on Alcohol and Related Conditions-III conducted a cross-sectional representative survey in the United States. In-person interviews were conducted with 36,309 adults. Prevalence of 12-month and lifetime substance use disorders were 3.9% and 9.9%, respectively. Twelve-month and lifetime substance use disorder was based on amphetamine, cannabis, club drug, cocaine, hallucinogen, heroin, non-heroin opioid, sedative/tranquilizer, and/or solvent/inhalant use disorders. Substance use disorder was generally greater among men, White and Native American individuals, younger and previously or never married adults, those with lower education and income, and those residing in the West (Grant et al., 2016).

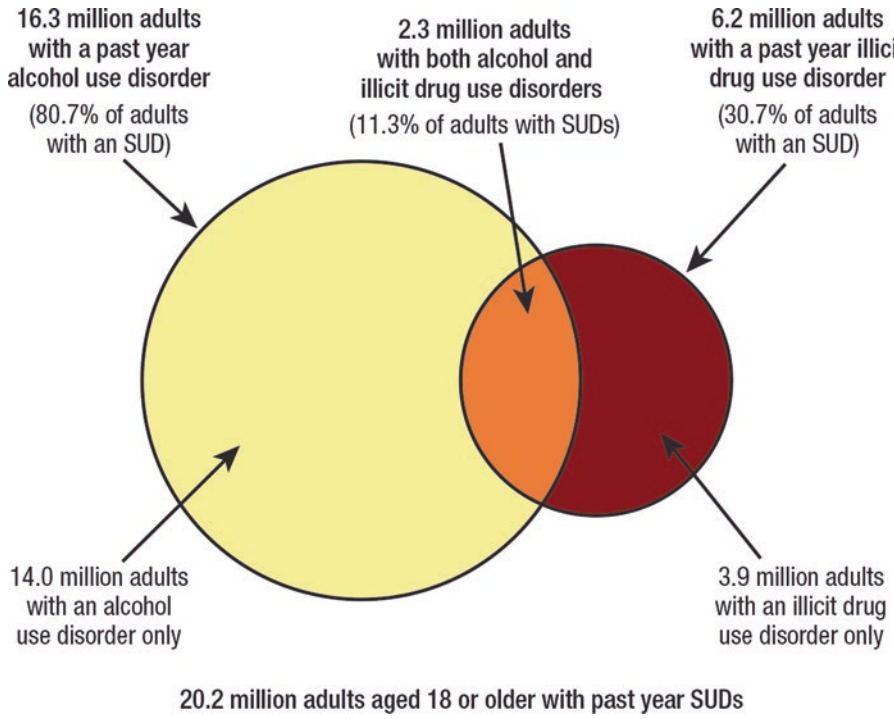

Fig. 18.1 SUDs in the past year among adults aged 18 or older: 2014. (Image Source: SAMHSA, Center for Behavioral Health Statistics and Quality, National Survey on Drug Use and Health (NSDUH), 2014)

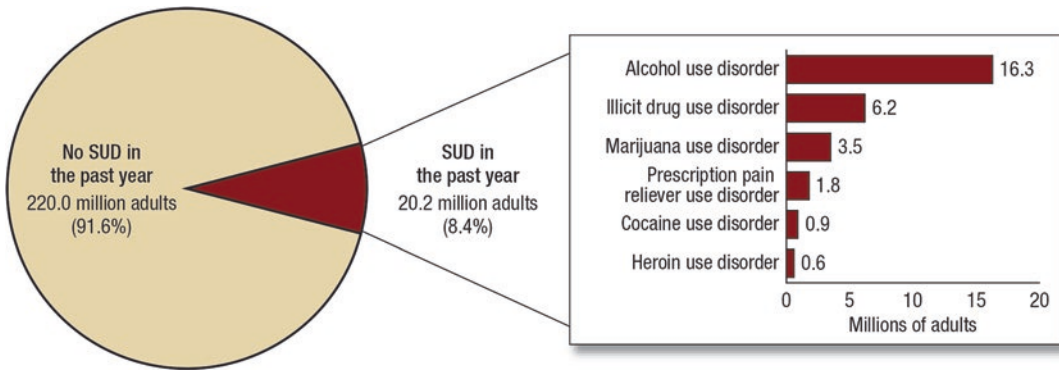

Fig. 18.2 SUD in the past year among adults aged 18 or older, by selected types of substances: 2014. (Image Source: SAMHSA, Center for Behavioral Health Statistics and Quality, National Survey on Drug Use and Health (NSDUH), 2014)

Consequences of Substance Abuse Including Mortality Data Substance abuse often begins as a pleasure activity; the addictive properties of the substances rapidly move from a perceived recreational activity into a continuous need to remain high. This craving is uncontrollable and may interfere with one's daily life often leading to death. The mortality data for substance abuse varies considerably by state, but 2016 saw a national average of 14 substance overdose deaths per 100,000 population. Such overdoses included traditional illicit drugs, but also misused prescription drugs, particularly opioids, abuse which has become a serious problem in recent years. Overdoses with opioid pharmaceuticals led to almost 17,000 deaths in 2011. More than 72,000

Americans died from drug overdoses in 2017, including illicit drugs and prescription opioid, displaying a twofold increase in a decade. From 2002 to 2017, the total number of deaths from substance abuse saw a 3.2-fold increase (NIDA, 2018a, 2018b).

The consequences of substance abuse disorders have both short-term and long-term effects which may vary from person to person. Short-term effects can occur after just one use. This can range from changes in appetite, wakefulness, heart rate, blood pressure, and/or mood to heart attack, stroke, psychosis, overdose, and even death. Longer-term effects can include heart or lung disease, cancer, mental illness, HIV/AIDS, hepatitis, and others. Long-term drug use can also lead to addiction. Drug addiction is a brain disorder. Not everyone who uses drugs will become addicted, but for some, drug use can change how certain brain circuit work. These brain changes interfere with how people experience normal pleasures in life such as food and sex, their ability to control their stress level, their decision-making, their ability to learn and remember, etc. These changes make it much more difficult for someone to stop taking the drug even when it is having negative effects on their life and they want to quit.

Symptoms experienced by each person may depend on age, gender, specific drug or drugs used, how they are taken, how much is taken, the person's health, individual physiology, genetic makeup, and mental health condition prior to substance abuse. And while some side effects are ranging from relatively mild physical effects including nausea and dehydration to work-related consequences such as reduced productivity, the greatest risks of substance abuse are dependence and life-threatening outcomes, particularly as a person's pattern of use progresses.

Substance use can lead to significant impairment in many areas of a person's life, from school to work and interpersonal relationships. This can include affecting a person's nutrition, sleep, decision-making and impulsivity, and risk for trauma, violence, injury, and communicable diseases. Other negative outcomes may be seen in education level, employment, housing, relationships, and criminal justice involvement.

Substance abuse in pregnant women has been linked to various short-term consequences and medical issues in newborns including preterm birth, low birth weight, congenital anomalies (e.g., oral facial clefts), neonatal abstinence syndrome or withdrawal symptoms, and neurobehavioral abnormalities (impaired orientation, autonomic regulation, and abnormalities of muscle tone). After controlling a variety of potentially confounding variables, babies born to substance abuse mothers encounter long-term consequences including poor growth throughout early childhood and adolescence, visions and hearing problems, behavioral issues (hyperactivity, negative and externalizing behaviors, delinquent and criminal behavior, disrupted school experiences, and peer problems), abnormalities in learning and memory, and poor language development in early childhood.

The negative consequences of substance abuse affect emotional and behavioral patterns of the parents, resulting in poor outcomes for the children. Parents with substance use disorders are three times more likely to physically or sexually abuse their child. Children who have experienced abuse are more likely to have the externalizing disorders such as anger, aggression, conduct, and behavioral problems, whereas children who experience neglect are more likely to have internalizing disorders (depression, anxiety, social withdrawal, poor peer relations). Incest has a very high association with parental substance abuse as do all types of sexual abuse. About two-thirds of incest perpetrators report using alcohol directly before the offending incident. Children from substance abuse homes are more than 50% more likely to be arrested as juveniles and 40% more likely to commit a violent crime (Lander, Howsare, & Byrne, 2013). Different substances pose different health risks. The heart, brain, liver, and kidneys are the most commonly affected organs. See Table 18.1 below for the short-term and long-term consequences of substance abuse.

Risk Factors and Determinants of Substance Abuse Substance abuse affects everyone

Table 18.1 Short-term and long-term consequences of substance abuse

	Short term	Long term
Alcohol	Mood swings Impaired judgment Coordination issues Trouble concentrating Memory problems Slurred speech Uncontrolled eye movements Stupor Coma	Cirrhosis Alcoholic hepatitis Liver cancer Pancreatitis Cardiomyopathy (stretching and weakening of heart muscle) Irregular heart rhythm High blood pressure Stroke Mouth and throat cancer Breast cancer Weakened immune system Irritability Suicidal ideation
Hallucinogens	Hallucinations Synesthesia or mixing of senses Intensified perceptions Significant anxiety or depression Increased heart rate Heart palpitations Dilated pupils Blurred vision Excessive sweating Tremors Paranoia Impaired judgment Impaired motor control	Persistent psychosis characterized by paranoia, mood and visual disturbances, and disorganized thought Hallucinogen persisting perception disorder (HPPD) Characterized by hallucinations, intensified colors, and other visual disturbances
Opioids	Euphoria followed by apathy Dysphoria or unease Nausea Vomiting Pinpoint pupils Itching skin Inattention to the environment Slowed thinking and movements Attention problems Memory impairments Drowsiness Slurred speech Coma.	Deterioration of the White matter in the brain Severe constipation and related gastrointestinal conditions (e.g., bowel obstruction, bowel perforation) Sexual dysfunction Irregular menses in women Intravenous consequences (e.g., track lines or puncture marks, peripheral edema, cellulitis, abscesses, tuberculosis, HIV or hepatitis virus contraction, and infection of the heart lining) Intranasal effects (e.g., irritation of nasal lining, perforation of nasal septum, and nasal bleeding)
Sedatives (e.g., barbiturates)	Mood swings Poor judgment Cognitive dysfunction	Physical injury resulting from accidents Assaults or fights Bullous (blistering) skin lesions
	Confusion	Irritability
	Drowsiness	Legal problems
	Sedation	School or work difficulties
	Slurred speech	Slowed pulse
	Trouble with coordination	Decreased respiratory rate
	Unsteady gait	Lower blood pressure
	ncontrolled eye movements	Memory loss
	Stupor	Changes in alertness
	Coma	
Inhalants	Euphoria	Liver damage
	Apathy	Kidney damage

(continued)

Table 18.1 (continued)

	Short term	Long term
	Lethargy	Hearing loss
	Poor judgment	Bone marrow damage
	Dizziness	Loss of coordination
	Nausea or vomiting	Limb spasms
	Hallucinations	Brain damage
	Delusions	Tuberculosis
	Blurred vision	Bronchitis
	Slurred speech	Asthma
	Impaired coordination	Sinus infections
	Muscle weakness	Depression
	Slowed or delayed reflexes	Anxiety
	Slow movement and thought	
	Tremors	
	Stupor	
	Coma	
Depressants	Slow brain function	Chronic fatigue
	Slowed pulse and breathing	Breathing difficulties
	Lowered blood pressure	Sexual problems
	Poor concentration	Sleep problems
	Confusion	
	Fatigue	
	Dizziness	
	Slurred speech	
	Fever	
	Sluggishness	
	Visual disturbances	
	Dilated pupils	
	Disorientation, lack of coordination	
	Depression	
	Difficulty or inability to urinate	
Stimulants	Rapid neurotransmitter release resulting in euphoria, increased energy and libido, reduced fatigue and appetite Behavioral responses, e.g., increased self-confidence and alertness Acute adrenergic effects include dose-responsive tachycardia and elevated blood pressure	Spectrum of psychotic features including paranoia, delusions, tactile hallucinations, or formication, colloquially referred to as "tweaking" Tissue ischemia Cerebrovascular disease and injury, including hemorrhagic and ischemic stroke Excess nervous system stimulation including seizures Cardiac arrhythmias Myocarditis and cardiomyopathy
Cannabis	Physically inactive Changes in mannerisms: talkative, laugh easily, and feel less inhibited Changes in the perception of time space, perspectives, and distances become distorted Reduced concentration and impaired memory and judgment Physiological changes: rapid heart rate, increased appetite, dryness of the mouth and throat, reddening of the eyes and drowsiness, feeling of confusion and anxiety	Depression and extreme suspicion of others Chronic paranoia: nervousness, irritability, and short temper Bronchitis Conjunctivitis Endocrine disorder Asthma Cancer if smoked with tobacco Reduced sex drive

regardless of age, race, gender, ethnicity, or economic status. Some drugs are also highly addictive (e.g., nicotine, heroin) especially when they are used excessively or on a daily basis. However, the vast majority of people who use alcohol and other drugs, including even many who use them heavily or regularly, do not develop the disease of abuse and addiction, some do. This brings up the question of whether or not certain risk factors and determinants are associated with addiction.

Assessing the risk in either adolescents or adults helps practitioners select appropriate interventions. Recent research points to a considerable number of such factors, including several biological, social, environmental, psychological, and genetic factors, are associated with substance abuse. These factors can include gender, race and ethnicity, age, income level, educational attainment, and sexual orientation (CDC, 2011). For easier clarification of this section, these factors have been grouped into two: biological disposition and environmental and psychological factors. Having one or more of these substance abuse risk factors does not mean someone will become addicted, but it does mean the odds are greater. The greater the propensity of risk factors, the greater is the chance that an individual will develop substance abuse disorders.

1. Biological predisposition: Certain biological factors that can increase an individual's likelihood of drug abuse which include:
   (a) Genetics/family history – genetic factors appear to have more of an influence in determining who progresses from substance use to addiction. Certain brain characteristics that can make someone more vulnerable to addictive substances than the average person. An individual who has a close relative with an addiction problem has a higher risk of eventually having one themselves. Although it may be argued that exposure to similar environmental and circumstantial factors among close family members could be the prominent causes.

   Researchers from the Universidad de Granada, Spain, in a study with 200 families revealed that *there is a genetic predisposition to alcohol addiction as lack of endorphin is hereditary (Community Research and Development Information Service,*2007*). Children aged between 6 months and 10 years old were enrolled from these 200 families. Children had lower beta-endorphin levels than other children of the same age and even lower if both parents were alcohol abusers.* Some geneticists also believe that the reason some people smoke cigarettes once in a while and are less likely to be addicted than others is probably linked to the type of genes inherited from parents. Geneticists believe that gene influences the way the receptors on the surface of our brain nerve cells respond to nicotine.

   Other biological predisposition include:
   (b) Mental illness (specifically those with ADHD, depression or anxiety, as well as other mental illnesses)
   (c) How the body metabolizes the substance (those with a higher natural tolerance may ingest more to achieve the drug's effects, thus raising their likelihood of addiction)
   (d) Gender – men are twice as likely to have drug abuse problems than women.
2. Interactions between environmental and psychological factors: Psychological and environmental factors appear to be more influential in determining whether an individual starts to use substances. These include exposure to physical, sexual, or emotional abuse or trauma, substance use, or addiction in the family or among peers, access to an addictive substance, and exposure to popular culture references that encourage substance use, stress, personality traits like high impulsivity or sensation seeking, depression, anxiety, eating disorders, personality, and other psychiatric disorders.

A research report published by the National Institute on Drug Abuse (2003) has shown that major transitions in people's lives starting from childhood are key risk periods for exposure to substance abuse. The first big transition occurs when a child leaves the security they had at home and enters school. Advancements from elementary school to middle school further expose the student to new academic and social situations, such as learning to get along with new peer groups. The first exposure to substance occurs during this early adolescence stage and through high school when they face additional social, emotional, and educational challenges. As one moves to teenage years, one may be exposed to greater availability of drugs, drug abusers, and social activities involving drugs. When young adults leave home for college or work and mingle with people of age groups, their risk for drug and alcohol abuse becomes very high.

Certain environmental factors are also related to psychological influences such as low socioeconomic status, academic failure or lack of academic motivation, alienation from peers or family, lack of strong family strength, childhood trauma, and domestic violence. According to a report by the Hawaii State Department of Health (2018), living in an economically depressed area with high unemployment, inadequate housing, high prevalence of crime, and high prevalence of illegal drug use increases the odds for substance abuse. Sadly, minority status statistically increases substance abuse. This points to racial discrimination, culture devalued in American society, differing generational levels of assimilation, cultural and language barriers to getting adequate health-care and other social services, low educational levels, and low achievement expectations from society (State of Hawaii Health Department, 2018).

A recent study was based on data gathered on over 2853 Hispanic and Black adults who live in high poverty areas. Some of the leading factors that are linked to substance abuse include poverty, homelessness, incarceration, low health literacy, symptoms of depression, and lack of support (Cleland, Lanza, Vasilenko, & Gwadz, 2017). According to Cleland et al. (2017), one-third (36%) of the study's participants met the diagnostic criteria for a substance abuse diagnosis. A multistate surveillance project by Diaz and colleagues found that among injection drug users, 35% of White men, 64% of Black men, and 67% of Puerto Rican men had not completed 12 years of school overall. Another study by Robles, Colon, Matos, Marrero, and Lopez (1990) has corroborated a high prevalence of school dropout status among Puerto Rican injection drug users. In a 2005 longitudinal study by Jaccard and colleagues, approximately 1700 peer dyads in Grades 7–11, it was found that the influence of friends on substance use was limited when controlling for parallel event and selection effects.

Methods and Procedures for Assessment

Preliminary reports in this chapter explain that most substance abuse cases both globally and in the United States are related to trends in alcohol and tobacco use. Although the rates of alcohol and tobacco use are higher, long-term reports imply that there is an increase in the percentage of adults who use and abuse illicit substances. To reduce the significant impact of substance abuse on clinically and functional impairment, such as health problems, disability, and failure to meet substantial responsibilities at work, school, or home, the complexity involved in attempting to assess substance abuse problem should be clearly defined. Given the nature of substance abuse, optimal treatment can only be achieved with proper assessment. The increased use of substance increases tolerance, meaning people who engage in substance abuse require higher doses to make the desired similar effect. Using substances or taking higher doses of substances increases an individual's risk of overdose. In early recovery, relapses are still likely to occur. The issue of substance abuse is almost a stressful experience for a previous user, which explains the reason why treatment and referral options focus on complete, comprehensive assessment to enable the practi-

tioner direct interventions toward avoiding the use of substances or even total abstinence.

Substance abuse assessment is a procedure for gathering information that assists in describing the nature of the problem, impact of this problem in other life areas, understanding client's readiness for change, and developing specific treatment recommendations for addressing the problem. Understanding methods and procedures for assessing substance abuse provides opportunities to determine the scope of the issue and to plan the prevention and treatment of substance abuse effectively. This understanding begins during the assessment process, which helps match the client with appropriate treatment services. To ensure that relevant information is obtained, providers should use standardized methods and procedures for assessment interview protocols, some of which have been studied for their sensitivity, validity, and accuracy in identifying substance abuse problems.

Substance abuse assessments are the first step in identifying substance abuse and may provide the visual evidence necessary to change a person's decision about treatment. Substance abuse is interview screenings that may be self-administered or with the help of a physician or clinician, family member, parent, friend, or coworker. Assessments also involve a series of questions that help in identifying the warning signs of substance abuse. Rather than using one method for screening, assessment examines a client's life in far more detail so that accurate diagnosis, appropriate treatment placement, problem lists, and treatment goals can be made (Center for Substance Abuse Treatment, 2009; SAMHSA, 2009). A comprehensive assessment generally collects information on four basic categories.

(a) Background – family history and social factors (employment, resources, education, housing status, history of trauma and domestic abuse, cultural relevance, vocational history, legal and financial situation) that increase the odds for substance abuse
(b) Substance use – age of first use, the person/persons who initially introduced substances, family history of substance abuse, type of substance abused, the amount consumed, frequency (i.e., daily, monthly, or yearly) or duration of use (i.e., how long abuse may have occurred), and pattern of use
(c) Psychological health – co-occurring psychiatric disorders, present mental illness and previous history of mental health problems, family history of mental illness, history of personal strengths and coping strategies and styles, and readiness to change and co-partner in treatment options
(d) Medical history and physical health – health history (e.g., previous health diagnoses), physical examination, and laboratory tests

Substance Abuse Assessment in Adults Obtaining detailed and entirely reliable information from adult clients can be a daunting task. Clients tend to deny or minimize their substance abuse problems. A practical assessment of substance abuse requires the use of evidence-based methods and procedures. The Minnesota Multiphasic Personality Inventory-2 (MMPI-2) that contains three scales, the MacAndrew Alcoholism Scale-Revised (MAC-R), the Addiction Potential Scale (APS), and the Addiction Acknowledgement Scale (AAS), was developed to identify alcohol and other substance abuse in clients. A study was designed by Rouse, Butcher, and Miller (1999) to measure the effectiveness of these scales at detecting substance abuse problems in a community-based mental health sample of 68 substance abusers and 392 non-abusing psychotherapy clients. The results indicated that all three scales could be useful in providing the practitioner with helpful information about the potential for substance abuse in clients.

Another popular tool for adults is the Screening, Brief Intervention, and Referral to Treatment (SBIRT) developed for primary care settings. About 70% of the US adult population visit primary care providers each year (Osborne & Benner, 2012). SBIRT was developing with the guiding constructs of the five stages of the transtheoretical model depicting client's readi-

ness to change: precontemplation, contemplation, preparation, action, and maintenance (Saitz, 2007). SBIRT incorporates motivational interviewing (Prochaska & DiClemente, 1983) which makes it efficacious in assessing and intervening for clients with substance abuse problems.

The assessment of opiate addiction includes the use of clinical examinations and evaluations. The most common method of opiate assessment is urine toxicology screening tests. However, substance abusers have many ways of falsifying their urine test results including switching the urine specimen with a non-abuser, purchasing falsifiers from the Internet, dilution of urines using water and other chemicals including bleach, drinking excessive amount of water and vinegar, and even making a hole on the specimen collector such that the specimen leaks before it arrives at the laboratory (Craig, 2011) – reasons why urine specimen collections require the conscientious effort of a fully trained technician. Other methods of assessing substance abuse include blood, saliva, sweat, and hair. However, blood is an invasive procedure, each sample requires at least 40 strands of hair, and the use of sweat and saliva can be problematic. Screening tests are used to rule out negative tests, to eliminate the tendency of negative tests from the positive tests, and many laboratories use immunoassays because they are consistent and easy to use (Craig, 2011).

Substance Abuse Assessment in Adolescents Substance abuse among teenagers is increasing at an alarming rate globally. Substance abuse alters a child's healthy development and places the child at a higher risk for mental, physical, and emotional problems including poor school performance, juvenile justice involvement, bullying, and violence. Methods and procedures used for assessing adolescents can be classified into three broad categories: screening instruments for evaluating risks, early intervention, and increasing awareness, midrange instruments for determining risks and measuring severity of problems, and comprehensive assessment instrument for identifying substances and predisposing factors, establishing differential diagnosis, initiating a treatment plan, and/or evaluating teenagers in treatment for substance abuse (Bivin & Riaz, 2017).

Screening instruments commonly used include the 15-item Substance Abuse Screening Instrument (SASI), 40-item Personal Experience Screening Questionnaire (PESQ), CRAFFT Screener, Adolescent Drinking Index (ADI), 30-item Drug and Alcohol Problem (DAP) Quick Screen, 13-item Adolescent Drug Involvement Scale (ADIS), Rutgers Alcohol Problem Index (RAPI), 6-item Cannabis Abuse Screening Test (CAST), Substance Abuse Subtle Screening Inventory (SASSI), and Drug Abuse Screening Test (DAST). Midrange assessments include 19-item Client Substance Index (CSI), 10-domain Drug Use Screening Inventory-Revised (DUSI-R), 139-item Problem Oriented Screening Instrument for Teenagers (POSIT), Teen Addiction Severity Index (T-ASI), and 128-question Assessment of Chemical Health Inventory (ACHI) Adolescent version. Comprehensive assessments include the Adolescent Diagnostic Interview (ADI), 150-item Prevention Intervention Management System (PMES), Personal Experience Inventory (PEI), and 150-item Adolescent Drug Abuse Diagnosis (ADAD).

Substance Abuse Assessment in Pregnancy Prenatal substance abuse is a weighty problem in the United States and in global countries which poses significant health risks in the development of the fetus. No single approach accurately determines substance abuse or the amount used during pregnancy; consequently, two basic methods are combined to produce comprehensive assessment in pregnant women: self-reported, structured interview and biological specimens. Self-report is an inexpensive method for obtaining information on the four basic categories, background, substance use, psychological health, and medical history/physical

health, as it relates to pregnant women. Unfortunately, it is faced with the uncertainties regarding the veracity of the informant and recall bias (Behnke & Smith, 2013).

Several biological specimens can be used to screen for substance abuse or exposure with each specimen having its variation. The three most commonly used during the prenatal and perinatal period are urine, meconium, and hair with none being the gold standard. Other studied methods include cord blood, human milk, amniotic fluid, and umbilical cord (Behnke & Smith, 2013). Immunoassays are the most analytical method used for screening biological specimens as it is designed to screen out a drug-free sample. The threshold for immunoassay is set so high which may be too high to detect low-dose or remote exposure; hence, positive results require confirmation using confirmatory tests (e.g., gas chromatography/mass spectrometry). It is important to note that the presence of a substance does not imply substance abuse.

Substance Abuse Assessment in the Verbally Impaired Practitioners are faced with communication issues when assessing deaf and hard of hearing clients. Unfortunately, several agencies that serve deaf and hard of hearing clients attempt to use standardized assessment tools such as the MAST (Michigan Alcohol Screening Tool) (Guthmann & Sandberg, 1998). The problems that emanate with the use of this kind of assessments are the vocabulary and language level of the tool. Many programs that serve deaf or hard of hearing clients have either created a new tool or modified existing ones. Perhaps more important than the nature of the instrument used is the manner in which the assessment interview is conducted. The practitioner or interviewer takes into account the knowledge base, communication, and cultural factors of the client. The process typically incorporates the four basic categories listed above including the isolation or bullying tendencies such a client may have encountered or are currently facing. Please see Appendix 1 for a sample of substance abuse assessment used by Guthmann and Sandberg (1998) among the disabled population (Table 18.2).

Issues in Differential Diagnosis

As per the diagnostic guidelines given in the latest Diagnostic and Statistical Manual of Mental Disorders, Fifth Edition (DSM-5), the previously used terms, substance abuse and substance dependence, have been replaced by substance use disorders (SUD). The SUD Diagnosis is based upon a pathological set of behaviors which falls into four areas: (1) impaired control, (2) social impairment, (3) risky use, and (4) pharmacological indicators which include tolerance and withdrawal. Depending on the severity of met criteria, substance use disorders are classified into mild, moderate, and severe categories. The DSM-5 groups ten individual disorders in the SUD. According to the Substance Abuse and Mental Health Services Administration (SAMSHA), six disorders are highly prevalent: (1) alcohol use disorder (AUD), (2) tobacco use disorder, (3) cannabis use disorder, (4) hallucinogen use disorder, (5) stimulant use disorder, and (6) opioid use disorder. Other four disorders included in DSM-5 are relatively less common: (7) sedative, hypnotic, or anxiolytic disorder, (8) inhalant-related disorder, (9) caffeine, and (10) other (or unknown) substance-related disorders. Table 18.3 shows the 11 criterion behaviors listed for diagnosis of SUD in DSM-5. The persistent presence of two to three behaviors is classified mild SUD, four to five behaviors moderate SUD, and six or more behaviors severe SUD (Table 18.3).

People suffering from SUD often experience the additional burden of other health problems (Whiteford et al., 2013). Psychological and mental issues are highly comorbid with SUD (Brady & Sinha, 2005; Kessler, 2004; Schuckit, 2006). A distinctive relationship between mental problems and SUD can influence each other (Brady & Sinha, 2007). Sometime one causes the other. People uses alcohol and other drugs in form of self-medication in order to relieve some psychological issues such as depression, anxiety, and

Table 18.2 Evidence-based screening tools and assessments for adults and adolescents

Tool	Substance type		Patient age		How tool is administered	
	Alcohol	Drugs	Adults	Adolescents	Self-administered	Clinician-administered
Screens						
Screening to Brief Intervention (S2BI)	X	X		X	X	X
Brief Screener for Tobacco, Alcohol, and Other Drugs (BSTAD)	X	X		X	X	X
Tobacco, Alcohol, Prescription Medication, and Other Substance Use (TAPS)	X	X	X		X	X
NIDA Drug Use Screening Tool: Quick Screen (NMASSIST)	X	X	X	See APA Adapted NM ASSIST tools	See APA Adapted NM ASSIST tools	X
Alcohol Use Disorders Identification Test-C (AUDIT-C (PDF, 41 KB))	X		X		X	X
Alcohol Use Disorders Identification Test (AUDIT (PDF, 233 KB))	X		X			X
Opioid Risk Tool (PDF, 168 KB)		X	X		X	
CAGE-AID (PDF, 30 KB)	X	X	X			X
CAGE (PDF, 14 KB)	X		X			X
Helping Patients Who Drink Too Much: A Clinician's Guide (NIAAA)	X		X			X
Alcohol Screening and Brief Intervention for Youth: A Practitioner's Guide (NIAAA)	X			X		X
Assessments						
Tobacco, Alcohol, Prescription Medication, and Other Substance Use (TAPS)	X	X	X		X	X
CRAFFT	X	X		X	X	X
Drug Abuse Screen Test (DAST-10)[a] For use of this tool – please contact Dr. Harvey Skinner		X	X		X	X
Drug Abuse Screen Test (DAST-20: Adolescent version)[a] For use of this tool – please contact Dr. Harvey Skinner		X		X	X	X
NIDA Drug Use Screening Tool (NMASSIST)	X	X	X			X
Helping Patients Who Drink Too Much: A Clinician's Guide (NIAAA)	X		X			X
Alcohol Screening and Brief Intervention for Youth: A Practitioner's Guide (NIAAA)	X			X		X

[a]Tools with associated fees

Source: Adapted from National Institute for Drug Abuse (NIDA) (2018a, 2018b). https://www.drugabuse.gov/nidamed-medical-health-professionals/tool-resources-your-practice/screening-assessment-drug-testing-resources/chart-evidence-based-screening-tools

Table 18.3 DSM-5 diagnostic criteria for substance use disorder

Areas	Cognitive, behavioral, and physiological symptoms	
Impaired control	1	Persistent desire to cut down or regulate substance use, often to no avail
	2	The individual may spend a great deal of time obtaining the substance, using, and recovering from its effects
	3	Virtually all of the individuals daily activates revolve around the substance, in some more severe disorders
	4	Craving is manifested by an intense desire or usage for the drug that may occurred at any time but is more likely when in an environment where the drug was previously obtained or used
Social impairment	5	Failure to fulfill major obligation roles at work, school, or home
	6	Continued substance use despite having persistent or recurrent social or interpersonal problems caused or exacerbated by the effects of the substance
	7	Withdrawal from family activities and hobbies in order to use the substance
Risky use	8	Recurrent substance use in situations in which it is physically hazardous
	9	The individual may continue to use despite knowledge of having a persistent or recurrent physical or psychological problem that is likely to have been caused or exacerbated by the substance
Pharmacological indicators (tolerance and withdrawal)	10	Tolerance is signaled by the increased dose of the substance to achieve the desired effect or the reduced effect when usual dose is consumed
	11	Withdrawal is a syndrome that occurs when blood or tissue concentrations of a substance decline in an individual who has maintained prolonged heavy use of the substance

stress (Bolton, Robinson, & Sareen, 2009; Robinson, Sareen, Cox, & Bolton, 2011). Studies have shown that self-medication in the form of alcohol and other drugs rather than improving the symptoms worsen the condition (Drake, Mueser, Brunette, & McHugo, 2004). In addition, many of them develop substance use disorder. On the other side, people suffering from SUD develop psychiatric problems on a higher rate than non-SUD due to a chronic presence of substance in the body that alters their neurophysiology of the brain.

The latest version of the International Statistical Classification of Diseases (ICD-10) is used in more than 110 countries in their healthcare system. It places all substance uses under a large category called mental and behavioral disorders due to psychoactive substance use. Unlike DSM-5, ICD-10 does not put substance use in a separate category; rather, it provides coding for mental and behavioral disorders due to the use of certain substances. Primarily, these categories are mental and behavioral disorders due to the use of alcohol, opioids, cannabinoids, sedative hypnotics, cocaine, stimulates including caffeine, hallucinogens, tobacco, volatile solvents, multiple drugs; and other psychoactive substances.

Diagnosis of any disorder is the first step toward the intervention. If diagnosis is unclear, it does not guide enough to the health-care professionals about the etiology and prognosis of the disorder. ICD-10 criteria give more emphasis on mental and behavioral aspects of the disorder, while DSM-5 also considers social aspects for the diagnosis. It is important to have a uniform diagnostic criteria for substance use disorder so the uniform approach for treatment and rehabilitation can be adopted.

The co-occurrence of psychiatric disorders with SUD is well reported in several studies (Brady & Sinha, 2007; Kessler, 2004). People have been given a dual diagnosis, among those many of them reported their first mental disorder at earlier ages than the SUD (Kessler, 2004). The following are important questions: Why do psychiatric and substance use disorders co-occur so often? Also, is there a common pathology or any neurobiological or genetic mediator that triggers both disorders (Brady & Sinha, 2007)? The understanding of the connection between SUD and mental disorders can be tremendously effective in the treatment and prevention of both disorders. However, such derivations do not reflect in

one of the diagnostic criteria proposed by either DSM-5 or ICD-10.

Psychiatric and substance use disorders exhibit very complex symptoms and occur due to similar biological, psychological, and social factors. Therefore, the diagnosis of co-occurring disorders can be extremely difficult. When incorrectly diagnosed, individuals with co-occurring disorders may receive better treatment in one area and poor to none in the other. The training and expertise of the health-care providers and their orientations can also affect the diagnosis. An integrated team approach can lower the risk of favored diagnosis and prevent misdiagnosis (Willenbring, 2005).

Substance Abuse and Intellectual Disabilities

Substance use in special populations has received increasing attention in the literature as well as in clinical practices (van Duijvenbode et al., 2015; Watson, Franklin, Ingram, & Eilenberg, 1998). People with intellectual disabilities are considered a special population. Intellectual disability is a developmental disability. As per the latest criteria laid out in the DSM-5, people with intellectual disabilities have deficits in intellectual functioning and impairment in adaptive behaviors. Intellectual functioning covers reasoning, problem-solving, planning, abstract thinking, judgment, and academic and experiential learning. Adaptive behaviors cover communication, social skills, personal independence at home or in community settings, and school or work functioning. On the basis of intelligence quotient (IQ), intellectual disabilities can be classified into four major categories: profound (IQ below 20–25), severe (20–25 to 35–40), moderate (35–40 to 50–55), and mild (IQ 50–55 to 70). Borderline (IQ 71–84) is also considered a minor form of intellectual disability.

The prevalence of intellectual disabilities around the world is documented to be about 1%. Most prevalence studies are conducted in developed countries. The research from developing countries indicates a prevalence rate of intellectual disabilities two times higher in low- and middle-income countries than high-income countries (Maulik, Mascarenhas, Mathers, Dua, & Saxena, 2011). Despite all medical and public health interventions, the prevalence of intellectual disabilities does not seem to be declining in the world (Bourke, de Klerk, Smith, & Leonard, 2016). In the United States, the rate of intellectual disabilities has been stable for the past few years, while other developmental disabilities such as autism and attention deficit and hyperactive disorders (ADHD) have increased (Boyle et al., 2011). In a large sample study of 26,986 people with autism spectrum disorder (ASD), researchers have estimated a doubled risk of substance use-related problems (Butwicka et al., 2017). Looking at an increasing trend in the rate, we assume that the intellectual disability populations would increase in the future. Thus, it is highly important to address an issue of substance use in this population.

The poor adaptive skills of people with intellectual disabilities in areas of social and practical skills make them more vulnerable for substance use (Sharma & Lakhan, 2017). People with intellectual disabilities also suffer from several psychological, social, and biological factors that impact their functioning, decision-making, and communication abilities in their day-to-day lives (Imrie, 2004). In relation to the nonintellectually disabled population, people with intellectual disabilities experience a greater amount of disease burdens. The psychiatric comorbidities are significantly higher in this population (Lakhan, 2013).

Mental disorders are considered to be one of the primary risk factors of substance use in all populations (Swendsen et al., 2010). People with intellectual disabilities experience a higher risk of substance use due to their coexisting mental disorders (Brunette, Mueser, & Drake, 2004; Chapman & Wu, 2012). In the presence of mental disorders, people with intellectual disabilities not only are limited with a greater risk of susceptibility for substance use but also suffer from an excess burden of negative consequences of substance use and a dual impact of two conditions on their lives (Chaplin, Gilvarry, & Tsakanikos,

2011; Chapman & Wu, 2012). A recent study conducted in Belgium demonstrates that the ID population has a higher prevalence of substance use than reported previously. In this study, 45.5% of people with mild-to-moderate ID living independently in communities were found using cannabis, tobacco, and alcohol (Swerts et al., 2017). Adolescents (aged 11–15) with ID in a self-reported study showed increased rates of smoking and decreased rates of alcohol use (Emerson & Turnbull, 2005).

In the United States alone, it has been estimated that seven to eight million people with intellectual disabilities experience a disproportionately higher burden of substance use (Chapman & Wu, 2012; Lin et al., 2016). However, in comparison to the non-ID population, the prevalence of alcohol and illicit drug use is relatively low, but its abuse is much higher (Chapman & Wu, 2012; VanDerNagel, Kiewik, Buitelaar, & DeJong, 2011; Williams, Kouimtsidis, & Baldacchino, 2018). Once people with moderate to borderline ID begin drinking, their amount of consumption increases rapidly, increasing their risk of intoxication and subsequent behaviors (Reis, Wetzel, & Häßler, 2017). A low prevalence of alcohol use in ID populations may be due to a poor identification of people with alcohol use disorders (Williams et al., 2018). Substance use and related problems with ID have been understudied (Chapman & Wu, 2012; McGillicuddy, 2006). Limited research on the prevalence of substance use disorders among people with ID limits researchers to understand the magnitude of the problem in this population (Chapman & Wu, 2012; Sharma & Lakhan, 2017). Scientific data from US-based studies reports the occurrence of substance use mainly in people with borderline, mild, and moderate ID. Studies from European nations have reported use of all forms of substances in all categories of ID in milder forms than the higher functioning ID populations (To, Neirynck, Vanderplasschen, Vanheule, & Vandevelde, 2014). Tobacco and alcohol are the two main substances that were found to be extensively used. Evidence of marijuana, cocaine, and amphetamines use was also reported in ID populations in the United States (Chapman & Wu, 2012).

Substance Use in ID Population in the United States Based on a few available studies, researchers have found that different forms of substances are used with different living settings (Chapman & Wu, 2012). A research conducted by Dinitto and Krishef from 1983 to 1984 included 274 adults with mild ID living in family, group home, or community settings and found that the alcohol use was highest. About 52% had consumed alcohol, while 47% were drinking monthly, 33% weekly, and 7% daily (Chapman & Wu, 2012; DiNitto & Krishef, 1984). A study by Edgerton (1986) conducted on 48 people with some form of ID in assisted living settings revealed a light-to-moderate use of alcohol and marijuana (Edgerton, 1986). Burtner, Wakham, McNeal, and Garvey (1995) have studied tobacco use in 749 people with different levels of ID living in state-run facilities. Their sample covered 56 mild, 66 moderate, 86 severe, and 541 profound ID. 22.5% moderate-to-mild people with ID had used tobacco (Burtner et al., 1995). Gress and Boss (1996) studied the use of tobacco, alcohol, marijuana, cocaine, and amphetamines in a population of 4114 children from 9th through 12th grade with the diagnosis of developmental handicaps. In 1 month, 26.9% used tobacco, 35.5% alcohol, 13.8% marijuana, 1.5% cocaine, and 2.5% amphetamine (Gress & Boss, 1996). Another study by Pack, Wallander, and Browne (1998) that surveyed 194 African American children with mild ID from 13 to 16 years of age placed in special education classes found that alcohol was used by 48% in the past year and 39% in the past month and marijuana by 13% in the past year and 10% in the past month (Pack et al., 1998). McGillicuddy and Blane (1999) studied 122 adults with the mean age of 27 with moderate-to-mild ID living in community settings. The alcohol use was 39%, tobacco 20.5%, drugs 4%, and misuse of any substance 18% (McGillicuddy & Blane, 1999).

Substance Use in ID Population in International Settings Recently, several studies have been conducted in European countries which attempted to find out the occurrence of different substance uses and their determinants and related outcome in the ID population. The risk-related association between degree of ID and type of substance use has already been established. Currently, focus has shifted in understating substance use in ID populations with their personality factors. Researchers have found that lower levels of anxiety sensitivity, higher levels of negative thinking, and impulsivity and sensation-seeking showed more severe alcohol use and higher levels of negative thinking and sensation-seeking associate with more severe drug use (Poelen, Schijven, Otten, & Didden, 2017).

A study was conducted in Spain among people with ID who were receiving psychiatric services. The sample covered 52.3% mild, 3.4% moderate, 3.4% severe, and 40.9% unspecified degrees of ID. Over 33% of people with ID included in the sample met SUD criteria. Cannabis was used by 25%, alcohol by 22.7%, and cocaine by 13.6%. Cannabis and cocaine were found to be highest among people with mild ID (Salavert et al., 2018).

In a relatively recent study conducted in Southeast London, researchers found that about 15% of people with mild ID had a history of substance use. Eight percent were currently using substances. In this population, alcohol was found to be one of the top most used substances with 80%, followed by cannabis at 28% and cocaine at 12% (Chaplin et al., 2011). In the United Kingdom, 1% of the ID population between 16 and 83 years of age had alcohol- and drug-related disorders (Cooper, Smiley, Morrison, Williamson, & Allen, 2007). In Australia, 25% of people with ID living in community settings smoked (Tracy & Hoskin, 1997). In a study on ID people aged 16–66 years in the Netherlands, 34.8% were using alcohol and cannabis, 32.6% exclusively alcohol, 8% exclusively cannabis, 15.1% stimulants, and 1.2% opiates (VanDerNagel et al., 2011). A study in Northern Ireland showed substance use relationships with the degree of ID. People with borderline and mild ID were found to be the highest users of different types of substances. Specifically among people with borderline ID, 38.1% were found using both alcohol and cannabis, 23.8% stimulants, 19% exclusively alcohol, and 9.5% exclusively cannabis. Among people with mild ID, alcohol use was found to be highest at 45.5%. About 34.1% used both alcohol and cannabis, while 6.8% exclusively used cannabis and stimulants at the same rate (6.8%). About 2.3% of people with mild ID were reported to be using opiates (VanDerNagel et al., 2011). In Greece, 29% of adolescents with mild ID aged 12–16 years living in the community settings used tobacco. This was relatively 8.8% higher than their age-matched non-ID peers (20.2%) living in the same settings (Kalyva, 2007). The prevalence of substance-related and addictive disorders among adults with ID was found to be 6.3% in a cohort study of Ontario's population in Canada. The prevalence was noted to be 2.8% higher compared to 3.5% of people without ID (Lin et al., 2016; Sharma & Lakhan, 2017).

Challenges Related with Substance Use Disorders in People with ID A great interest is seen in studying the various aspects of substance use in ID populations. Without having a proper understanding of the number of people with ID suffering from substance use disorders in this population, what are the risks for their use, and how do certain substances affect their lives, it would be difficult to have appropriate prevention and intervention plans to apply. Some of the challenges faced in this crucial area of research are mentioned here.

Literature indicates that the prevalence of substance use is the highest and an increasing trend among people with borderline and mild ID. Are substances such as alcohol, cannabis, and tobacco the most common in this group of ID? Do they have some form of protective effect for them that

may be leading to an increased use? Mild and borderline ID are two separate categories. Both groups exhibit higher functioning, their defining criteria vary, and they live in different community settings. Co-occurring mental health issues are highly prevalent in both categories. Putting them together in one category as a mild and borderline ID as done in many previous studies does not provide the most accurate prevalence rates of substance use (Didden, 2017; Didden, VanDerNagel & Van Duijvenbode, 2016; Van Duijvenbode et al., 2015). Valid instruments for the measurement of substance use in this population are lacking. Reliable, valid, and standardized screening tools that can detect substance use in people with ID are needed (Sharma & Lakhan, 2017). Compared to non-ID people, the identification and screening of substance use disorders in people with ID is highly challenging. Often, it goes unidentified because of psychiatric comorbidity, lower cognitive functioning, and maladaptive behaviors (Sharma & Lakhan, 2017; To, Vanheule, Vanderplasschen, Audenaert, & Vandevelde, 2015; VanDerNagel et al., 2011). Because of a lack of screening tools used commonly in regular practice in clinical settings, clinicians use their clinical judgment in identifying the presence of substance use disorders which may limit many service providers in identifying people with ID affected with substance use (McLaughlin, Taggart, Quinn, & Milligan, 2007; Sharma & Lakhan, 2017). Stigma and denial associated with the substance use in people with ID and their caregivers is also a factor that affects identification of substance use in certain people with ID. In recent years, deinstitutionalization has been promoted. This presents a greater opportunity for people with ID to be exposed to substance use. Differences in socioeconomic factors, polices related to alcohol and drugs, care for ID populations, and treatment facilities in different countries have an impact on prevalence rates and their interventions (Didden, 2017; Didden, VanDerNagel, & Van Duijvenbode, 2016; Van Duijvenbode et al., 2015). Substance use in people with ID is not viewed the same way as it is in non-ID populations. This approach significantly impacts prevention and also priorities associated with the disorder in this population (van Duijvenbode et al., 2015). Health promotion models for the prevention of substance use disorders based on non-ID population need to be tailored for the ID population to serve them better (Sharma & Lakhan, 2017). Treatment guidelines for substance use are also unclear for this population. In the ID populations, substance use either is considered an outcome of behavioral problems (Dekker & Koot, 2003), mental illness (Merikangas & McClair, 2012), and brain disease (Koob, 2013) or is a chronic medical illness like diabetes, hypertension, and asthma (McLellan, Lewis, O'Brien, & Kleber, 2000). About 14–30% of people with ID without a diagnosed psychiatric disorder receive psychotropic medicines for the management of their behavioral problems (Deb et al., 2009). People with ID acclimate to the unwarranted psychotropic substance. It is important that substance use in people with ID, whether it is self-choice or prescribed, needs to be minimized. An integrated approach of treatment and prevention should be applied (Sharma & Lakhan, 2017).

Conclusions

Substance use in the forms of self-medication, recreational, and prescription has increased in most populations around the world. The burden of substance use disorders is increasing in most societies. People with ID face the worst of this scourge. They are similarly experiencing a significant burden of substance use disorders in their lives. Greater attention is being paid on the substances and their related issues. There has been an issue in defining substance abuse in ways that allow it to be measured. The recent DSM-5 has proposed the most comprehensive criteria for substance abuse and named it "substance use disorders." The DSM-5 combines four areas, *impaired control*, *social impairment*, *risky use*, and *pharmacological implications*, to diagnose

and assess the severity of SUD. Mental problems are highly common in people with SUD. The relationship between SUD and mental problems is two-pronged. Many people with SUD develop mental issues, and some develop SUD because of mental issues. The ICD-10 revised criteria emphasized more on developing mental issues from SUD. It categorized mental and behavioral disorders on the basis of substances. Psychiatric comorbidity creates challenges in diagnosis. In the past decade, several reliable, valid, and standardized instruments have been developed. The SAMSHA and NIDA have taken up the issue of substance use disorders very seriously. They have been instrumental in creating awareness on this issue. A lot of resource material has been prepared. Both organizations are continuing with their effort in addressing the issue of SUD at various levels including prevention, treatment, and policy intervention. People with intellectual disabilities often experience slightly greater risks of SUD. The psychiatric comorbidity is even higher in this population, which presents complex challenges in diagnosis and treatment of SUD and other preexisting medical conditions. Tobacco, alcohol, cigarette smoking, and cocaine were found to be the most common substances used by people with moderate, mild, and borderline ID. The use of prescription drugs in the ID population is also very high. The exact prevalence estimates of all types of substance use and prescription drugs in all categories of ID are not well known. Therefore, the magnitude of the problem cannot be estimated. Specific challenges such as a lack of reliable and valid screening instruments and diagnostic criteria and a lack of trained professionals, stigma, and evidence-based intervention and rehabilitation plans limit effective intervention in this population. More importantly, like SAMSHA and NIDA for non-ID populations, there is no agency that exclusively addresses SUD in ID populations.

Appendix 1

Substance Abuse Assessment -Tim
Minnesota Chemical Dependency Program for
Deaf and Hard of Hearing Individuals

Client Name: _____ Tim _____ Date: 4-1-98_____
Assessor: _Ann Jones_____
Referred by: _____ School Counselor _____ Agency: _____ School for the Deaf _____ Phone: 555-3333
Reason for Referral: _____problems in school _____

Background Information

Date of Birth: 3-28-81 _____ Age: _____ 17 _____ Gender: Male
Marital Status: _____ Single _____ Living Arrangement: _____ Lives w/ mother _____
School Status: _____ Junior in H.S. _____ Employment Status: _____ student_____
Communication Preference: _____Sign language_____
Family Incidence of Hearing Loss? YES / NO If yes, identify members: _____ Family is hearing _____
Family Incidence of alcohol/drug problems? YES If yes, identify members: _____ Unknown _____
_____
Other background information: _____Parents divorced when Tim was 8 years old_____
_____

Treatment History

Admissions for Detox: Place __None reported_____ Dates _____
Place _____ Dates_____
Admissions for Treatment:
Place _____ None reported_____ Inpatient/Outpatient Dates _____
Place _____ Inpatient/Outpatient Dates _____
Place _____ Inpatient/Outpatient Dates _____
Longest period of sobriety after treatment: _____ NA _____
Most recent period of sobriety: _____ NA _____

Problems Related to Chemical Use

Physical Problems

_____ x _____ Hangovers _____ Tolerance _____ Withdrawal
_____ x _____ Blackouts _____ Accidents/Injuries _____ x _____ Passing out
_____ x _____ Fights _____ Injecting drugs _____ Medicating pain
Comments:
Increased frequency of illnesses.

Financial Problems

_____ Unpaid Bills _____ X _____ Borrowing money _____ Outstanding loans
_____ Legal fines _____ Stealing _____ Dealing
_____ Lifestyle change _____ Insufficient income _____ Pawning items
Comments:
Seems to have a lot of money at times....unsure of the source.

Family Problems

_____ x _____ Arguments/fights _____ Abuse _____ Broken promises
_____ x _____ Absence from home _____ Loss of trust _____ X _____ Concerns about use
_____ Use by other members _____ Hiding drugs in home _____ Custody issues
Comments:

Legal Problems

_____ Arrests _____ Near arrests _____ DWI/DUI
_____ Gang Involvement _____ Court Appearances _____ Parole
_____ Restraining order _____ Domestic violence _____ Probation
Comments:
Tim has recently been picked up for curfew violations.

Job/School Problems

_____ X _____ Poor performance _____ X _____ Lateness _____ X _____ Absences

_____ Problems with supervisor _____ Fired/Suspended _____ Disciplined
_____ X _____ Problems with peers _____Using at work/school
Comments:
Pattern of absences/lateness. Declining performance.
Social Problems
_____ x _____ Loss of friends _____ x _____ Change of friends _____ Friends use
_____ Socialization around use _____ Negative reputation _____ Gambling
_____ x _____ Friends older/younger
Comments:
Emotional Problems
_____ Use to feel normal _____ Mood swings _____ Self harm
_____ Suicidal thoughts/behavior _____ Anger problems _____ Depression _____ Use to medicate emotional pain
Comments:
Chemical Use Information
_____ Unplanned use _____ Binge Use _____ Hidden use
_____ Using more than planned _____ Solo Use _____ Daily use
_____ Attempts to control use _____ Relapse _____ Preoccupation
_____ Protecting Supply _____ Poly drug use
Comments:
Identify chemicals used. For each chemical, identity age of first use and present pattern of use.
_____ x _____ Alcohol _____ x _____ Marijuana _____ Cocaine
_____ Crack _____ Inhalants _____ Sedatives
_____ Hallucinogens _____ Amphetamines _____ Opiates
_____ Others: _____ Others: _____ Others:
Use information: _____ _
Diagnostic features: Please check all that apply.
_____ TOLERANCE need for increase amounts of substance to achieve intoxication or markedly diminished effect with continued use of the same amount.
_____ WITHDRAWAL characteristic syndrome or same or closely related substance taken to relieve or avoid withdrawal symptoms.
_____ SUBSTANCE taken in larger amounts or over longer period than intended.
_____ PERSISTENT desire or unsuccessful efforts to cut down or control use.
_____ TIME spent in activities necessary to obtain substance or recover from its use.
_____ x _____ SOCIAL, OCCUPATIONAL, RECREATIONAL activities given up or reduced because of use.
_____ CONTINUED use despite knowledge of physical or psychological problems caused or exacerbated by the use.
Interview Findings and Comments:

References

Behnke, M., & Smith, V. C. (2013). Prenatal substance abuse: Short and long term effects on the exposed fetus. *Pediatrics, 131*(3), e1009–e1024.

Bivin, J. B., & Riaz, K. M. (2017). Assessment of substance abuse among teenagers: Review of instruments commonly used in healthcare and research. *Asian Journal of Nursing Education and Research, 7*(2), 248–254.

Bolton, J. M., Robinson, J., & Sareen, J. (2009). Self-medication of mood disorders with alcohol and drugs in the National Epidemiologic Survey on Alcohol and Related Conditions. *Journal of Affective Disorders, 115*(3), 367–375.

Bourke, J., de Klerk, N., Smith, T., & Leonard, H. (2016). Population-based prevalence of intellectual disability and autism spectrum disorders in Western Australia: A comparison with previous estimates. *Medicine, 95*(21).

Boyle, C. A., Boulet, S., Schieve, L. A., Cohen, R. A., Blumberg, S. J., Yeargin-Allsopp, M., ... Kogan, M. D. (2011). Trends in the prevalence of developmental disabilities in US children, 1997–2008. *Pediatrics*, peds-2010.

Bozzelli, E. K. (2008). *Subjective definitions of substance abuse problems: Does age matter?* (Doctoral dissertation) Miami University. Available at: https://etd.ohiolink.edu/pg_10?0::NO:10:P10_ACCESSION_NUM:miami1220005252

Brady, K. T., & Sinha, R. (2005). Co-occurring mental and substance use disorders: The neurobiological effects of chronic stress. *American Journal of Psychiatry, 162*(8), 1483–1493.

Brady, K. T., & Sinha, R. (2007). Co-occurring mental and substance use disorders: The neurobiological effects of chronic stress. *Focus, 5*(2), 229–239.

Brunette, M. F., Mueser, K. T., & Drake, R. E. (2004). A review of research on residential programs for people with severe mental illness and co-occurring substance use disorders. *Drug and Alcohol Review, 23*(4), 471–481.

Burtner, A., Wakham, M., McNeal, D., & Garvey, T. (1995). Tobacco and the institutionally mentally retarded: Usage choices and the ethical considerations. *Special Care in Dentistry, 15*, 56–60.

Butwicka, A., Långström, N., Larsson, H., Lundström, S., Serlachius, E., Almqvist, C., … Lichtenstein, P. (2017). Increased risk for substance use-related problems in autism spectrum disorders: A population-based cohort study. *Journal of Autism and Developmental Disorders, 47*(1), 80–89.

Center for Substance Abuse Treatment. (2009). Rockville (MD). Retrieved from https://www.ncbi.nlm.nih.gov/books/NBK83253/

Centers for Disease Control and Prevention. (2011). CDC health disparities and inequalities report: United States, 2011. *Morbidity and Mortality Weekly Report, 60* (supplement). Retrieved from http://www.cdc.gov/mmwr/pdf/other/su6001.pdf

Chaplin, E., Gilvarry, C., & Tsakanikos, E. (2011). Recreational substance use patterns and co-morbid psychopathology in adults with intellectual disability. *Research in Developmental Disabilities, 32*(6), 2981–2986. https://doi.org/10.1016/j.ridd.2011.05.002

Chapman, S. L. C., & Wu, L. T. (2012). Substance abuse among individuals with intellectual disabilities. *Research in Developmental Disabilities, 33*(4), 1147–1156. https://doi.org/10.1016/j.ridd.2012.02.009

Cleland, C. M., Lanza, S. T., Vasilenko, S. A., & Gwadz, M. (2017). Syndemic risk classes and substance use problems among adults in high-risk urban areas: A latent analysis. *Frontiers in Public Health, 5*. https://doi.org/10.3389/fpubh.2017.00237

Collins, S. E., & Kirouac, M. (2013). Alcohol consumption. *Encyclopedia of behavioral medicine, 2013*, 61–65. https://doi.org/10.1007/978-1-4419-1005-9_626

Community Research and Development Information Service [CORDIS]. (2007). *Alcohol abuse is in the genes*. Retrieved from https://cordis.europa.eu/news/rcn/114231_en.html

Cooper, S. A., Smiley, E., Morrison, J., Williamson, A., & Allen, L. (2007). Mental ill–health in adults with intellectual disabilities: Prevalence and associated factors. *British Journal of Psychiatry, 190*, 27–35.

Craig, R. J. (2011). Current controversies in the assessment and treatment of heroin addiction. In K. A. Murati & A. G. Fisher (Eds.), *Substance abuse, assessment and addiction* (pp. 97–116). New York: Nova Science Publishers, Inc.

Deb, S., Kwok, H., Bertelli, M., Salvador-Carulla, L., Bradley, E., Torr, ... Barnhill & 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.

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. https://doi.org/10.1097/01.CHI.0000046892.27264.1A

Didden, H. C. M. (2017). Substance use and abuse in individuals with mild intellectual disability or borderline intellectual functioning: An introduction to the special section. *Research in Developmental Disabilities, 63*, 95–98. https://doi.org/10.1016/j.ridd.2017.02.001

Didden, R., VanDerNagel, J., & van Duijvenbode, N. (2016). Substance use disorders. In Handbook of evidence-based practices in intellectual and developmental disabilities (pp. 957–965). Springer, Cham.

DiNitto, D. M., & Krishef, C. H. (1984). Drinking patterns of mentally retarded persons. *Alcohol Health and Research World, 8*(2), 40–42.

Drake, R. E., Mueser, K. T., Brunette, M. F., & McHugo, G. J. (2004). A review of treatments for people with severe mental illnesses and co-occurring substance use disorders. *Psychiatric Rehabilitation Journal, 27*(4), 360.

Edgerton, R. (1986). Alcohol and drug use by mentally retarded adults. *American Journal of Mental Deficiency, 90*, 602–609.

Emerson, E., & Turnbull, L. (2005). Self-reported smoking and alcohol use among adolescents with intellectual disabilities. *Journal of Intellectual Disabilities, 9*(1), 58–69.

Grant, B. F., Saha, T. D., Ruan, W. J., Goldstein, R. B., Chou, S. P., Jung, J., … Hasin, D. S. (2016). Epidemiology of DSM-5 drug use disorder results from the national epidemiologic survey on alcohol and related conditions–III. *JAMA Psychiatry, 73*(1), 39–47. https://doi.org/10.1001/jamapsychiatry.2015.2132

Gress, J., & Boss, M. (1996). Substance abuse differences among students receiving special education school services. *Child Psychiatry and Human Development, 26*, 235–246.

Guthmann, D., & Sandberg, K. (1998). Assessing substance abuse problems in deaf and hard of hearing individuals. *American Annals of the Deaf, 143*(1), 14–21.

Imrie, R. (2004). Demystifying disability: A review of the International Classification of Functioning, Disability and Health. *Sociology of Health & Illness, 26*(3), 287–305.

Kalyva, E. (2007). Prevalence and influences on self-reported smoking among adolescents with mild learning disabilities, attention deficit hyperactivity disorder, and their typically developing peers. *Journal of Intellectual Disability, 11*, 267–279.

Kessler, R. C. (2004). The epidemiology of dual diagnosis. *Biological Psychiatry, 56*(10), 730–737.

Koob, G. F. (2013). Addiction is a reward deficit and stress surfeit disorder. *Frontiers in Psychiatry, 4*(72), 1–18. https://doi.org/10.3389/fpsyt.2013.00072

Kumar, S., Mehrotra, D., Mishra, S., Goel, M. M., Kuman, S., Mathur, P., … Pandey, C. M. (2015). Epidemiology of substance abuse in the population of Lucknow. *Journal of Oral Biology and Craniofacial Research, 5*, 128–133. https://doi.org/10.1016/j.jobcr.2015.08.010

Lakhan, R. (2013). The coexistence of psychiatric disorders and intellectual disability in children aged 3–18 years in the Barwani District, India. *ISRN psychiatry*, *2013*.

Lakhan, S. E., & Kirchgessner, A. (2012). Prescription stimulants in individuals with and without attention deficit hyperactivity disorder: Misuse, cognitive impact, and adverse effects. *Brain and Behavior, 2*(5), 661–677. https://doi.org/10.1002/brb3.78

Lander, L., Howsare, J., & Byrne, M. (2013). The impact of substance use disorders on families and children: From theory to practice. *Social Work in Public Health, 28*(0), 194–205. https://doi.org/10.1080/19371918.2013.759005

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*(9), e011638. https://doi.org/10.1136/bmjopen-2016-011638

Lipari, R. N., & Van Horn, S. L. (2017). *Trends in substance use disorders among adults aged 18 or older.* Retrieved from https://www.samhsa.gov/data/sites/default/files/report_2790/ShortReport-2790.html

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.

McGillicuddy, N. (2006). A review of substance use research among those with mental retardation. *Mental Retardation and Developmental Disability Research Reviews, 12*, 41–47.

McGillicuddy, N., & Blane, H. (1999). Substance use in individuals with mental retardation. *Addictive Behaviors, 24*(6), 869–878.

McLaughlin, D. F., Taggart, L., Quinn, B., & Milligan, V. (2007). The experiences of professionals who care for people with intellectual disability who have substance-related problems. *Journal of Substance Use, 12*(2), 133–143. https://doi.org/10.1080/14659890701237041

McLellan, A. T., Lewis, D. C., O'Brien, C. P., & Kleber, H. D. (2000). Drug dependence, a chronic medical illness: Implications for treatment, insurance, and outcomes evaluation. *JAMA, 284*(13), 1689–1695. https://doi.org/10.1001/jama.284.13.1689

Merikangas, K. R., & McClair, V. L. (2012). Epidemiology of substance use disorders. *Human Genetics, 131*(6), 779–789. https://doi.org/10.1007/s00439-012-1168-0

National Institute on Drug Abuse. (2003). *Preventing drug use among children and adolescents (in brief).* Retrieved from https://www.drugabuse.gov/publications/preventing-drug-use-among-children-adolescents-in-brief

National Survey on Drug Use and Health. (2011). Results from the 2011 National Survey on Drug Use and Health: Mental Health Findings. Retrieved from https://www.samhsa.gov/data/sites/default/files/2011MHFDT/2k11MHFR/Web/NSDUHmhfr2011.htm

National Institute on Drug Abuse. (2016). *Hallucinogens.* Retrieved from https://www.drugabuse.gov/publications/drugfacts/hallucinogens

National Institute on Drug Abuse. (2018a). *Media guide.* Retrieved from https://www.drugabuse.gov/publications/media-guide

National Institute on Drug Abuse. (2018b). *Cigarettes and other tobacco products.* Retrieved from https://www.drugabuse.gov/publications/drugfacts/cigarettes-other-tobacco-products

National Institute on Drug Use [NIDA]. (2018). *Alcohol.* Retrieved from https://www.drugabuse.gov/drugs-abuse/alcohol

National Institute on Drug Abuse. (2020). Hallucinogens. Retrieved from https://www.drugabuse.gov/drug-topics/hallucinogens

Osborne, V. A., & Benner, K. (2012). Utilizing screening, brief intervention, and referral to treatment: Teaching assessment of substance abuse. *American Journal of Public Health, 12*(7), e37–e38.

Pack, R., Wallander, J. L., & Browne, D. (1998). Health risk behaviors of African American adolescents with mild mental retardation: Prevalence depends on measurement method. *American Journal on Mental Retardation, 102*, 409–420.

Peacock, A., Leung, J., Larney, S., Colledge, S., Hickman, M., Rehm, J., … Degenhardt, L. (2018). Global statistics on alcohol, tobacco and illicit drug use: 2017 status report. *Addiction, 113*(10), 1905–1926. https://doi.org/10.1111/add

Poelen, E. A., Schijven, E. P., Otten, R., & Didden, R. (2017). Personality dimensions and substance use in individuals with mild to borderline intellectual disabilities. *Research in Developmental Disabilities, 63*, 142–150.

Prochaska, J. O., & DiClemente, C. C. (1983). Stages and processes of self-change of smoking: Toward an integrative model of change. *Journal of Consulting and Clinical Psychology, 51*(3), 390–395.

Reis, O., Wetzel, B., & Häßler, 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, J., Sareen, J., Cox, B. J., & Bolton, J. M. (2011). Role of self-medication in the development of comorbid anxiety and substance use disorders: A longitudinal investigation. *Archives of General Psychiatry, 68*(8), 800–807.

Robles, R. R., Colon, H. M., Matos, T. D., Marrero, C. A., & Lopez, C. M. (1990). AIDS risk behavior patterns

among intravenous drug users in Puerto Rico and the United States. *Bol Assoc Med P Rico, 82*(12), 523–527.

Rouse, S. V., Butcher, J. N., & Miller, K. B. (1999). Assessment of substance abuse in psychotherapy clients: The effectiveness of the MMPI-2 substance abuse scales. *Psychological Assessment, 11*(1), 101–107. https://doi.org/10.1037/1040-3590.11.1.101

Saitz, R. (2007). Screening and brief intervention enter their 5th decade. *Substance Abuse, 28*(3), 3–6.

Salavert, J., Clarabuch, A., Fernández-Gómez, M. J., Barrau, V., Giráldez, M. P., & Borràs, J. (2018). Substance use disorders in patients with intellectual disability admitted to psychiatric hospitalisation. *Journal of Intellectual Disability Research, 62*(11), 923–930. https://doi.org/10.1111/jir.12514. Epub 2018 Jul 1.

Schiller, E. Y., & Mechanic, O. J. (2018). *Opioid, overdose*. Treasure Island, FL: StatPearls Publishing. Available from: https://www.ncbi.nlm.nih.gov/books/NBK470415/

Schuckit, M. A. (2006). Comorbidity between substance use disorders and psychiatric conditions. *Addiction, 101*, 76–88.

Sharma, M., & Lakhan, R. (2017). Substance abuse among people with intellectual disabilities: Areas of future research. *Journal of Alcohol and Drug Education, 61*(2), 3–6.

State of Hawaii Health Department. (2018). *Alcohol and drug abuse division*. Retrieved from http://health.hawaii.gov/substance-abuse/prevention-treatment/prevention/risk-factors/

Statista. (2020). Lifetime prevalence of use of any illicit drug for grades 8, 10 and 12 combined from 1991 to 2019. Retrieved from https://www.statista.com/statistics/208420/us-lifetime-prevalence-drug-use-grades-8-10-12-since-1991/

Substance Abuse and Mental Health Services Administration. (2015). *Substance use disorders.* Retrieved from https://www.samhsa.gov/disorders/substance-use

Substance Abuse and Mental Health Services Administration. (2016). *Stimulants.* Retrieved from https://www.samhsa.gov/atod/stimulants

Substance Abuse and Mental Health Services administration [SAMHSA] & Center for Substance Abuse Treatment [CSAT]. (2009). *Treatment Improvement Protocol (TIP) Series, No. 51.*

Swendsen, J., Conway, K. P., Degenhardt, L., Glantz, M., Jin, R., Merikangas, K. R., … Kessler, R. C. (2010). Mental disorders as risk factors for substance use, abuse and dependence: Results from the 10-year follow-up of the National Comorbidity Survey. *Addiction, 105*(6), 1117–1128.

Swerts, C., Vandevelde, S., VanDerNagel, J. E., Vanderplasschen, W., Claes, C., & De Maeyer, J. (2017). Substance use among individuals with intellectual disabilities living independently 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), 1–9.

To, W. T., Vanheule, S., Vanderplasschen, 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, 36*, 498–504. https://doi.org/10.1016/j.ridd.2014.10.046

Tracy, J., & Hoskin, R. (1997). The importance of smoking education and preventative health strategies for people with intellectual disability. *Journal of Intellectual Disability Research, 40*(5), 416–421.

van Duijvenbode, N., VanDerNagel, J. E., Didden, R., Engels, R. C., Buitelaar, J. K., Kiewik, M., & de Jong, C. A. (2015). Substance use disorders in individuals with mild to borderline intellectual disability: Current status and future directions. *Research in Developmental Disabilities, 38*, 319–328.

VanDerNagel, J., Kiewik, M., Buitelaar, J., & DeJong, C. (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*(3), 143–149. https://doi.org/10.1111/j.1741-1130.2011.00304.x

Watson, A. L., Franklin, M. E., Ingram, M. A., & Eilenberg, L. B. (1998). Alcohol and other drug abuse among persons with disabilities. *Journal of Applied Rehabilitation Counseling, 29*(2), 22.

Wendell, A. D. (2013). Overview and epidemiology of substance abuse in pregnancy. *Clinical Obstetrics and Gynecology, 56*(1), 91–96.

Whiteford, H. A., Degenhardt, L., Rehm, J., Baxter, A. J., Ferrari, A. J., Erskine, H. E., … Burstein, R. (2013). Global burden of disease attributable to mental and substance use disorders: Findings from the Global Burden of Disease Study 2010. *The Lancet, 382*(9904), 1575–1586.

Willenbring, M. L. (2005). Integrating care for patients with infectious, psychiatric, and substance use disorders: Concepts and approaches. *AIDS, 19*, S227–S237.

Williams, F., Kouimtsidis, C., & Baldacchino, A. (2018). Alcohol use disorders in people with intellectual disability. *British Journal of Psychiatry Advances*, 1–9.

World Health Organization. (2018) *Substance abuse*. Retrieved from http://www.who.int/topics/substance_abuse/en/

Wu, L., Woody, G. E., Yang, C., Pan, J., & Blazer, D. G. (2011). Racial/ethnic variations IN substance-related disorders among adolescents in the United States. *Archives of General Psychiatry, 68*(11), 1176–1185. https://doi.org/10.1001/archgenpsychiatry.2011.120

Wu, L.-T., Ringwalt, C. L., Mannelli, P., & Patkar, A. A. (2008). Hallucinogen use disorders among adult users of MDMA and other hallucinogens. *The American Journal on Addictions, 17*(5), 354–363. https://doi.org/10.1080/10550490802269064

Aging with Intellectual Disability: Dementia and Cognitive Decline

19

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Introduction

Greater survival rates to late life have been reported among people with an intellectual disability with recent studies reporting averages of 55–66 years (Hartley et al., 2015; Krinsky-Mchale & Silverman, 2013; McCarron, Carroll, Kelly, & McCallion, 2015; Torr & Davis, 2007). This is to be welcomed but, for some, it has brought increased morbidity, both in terms of physical, mental health, and neurocognitive challenges (McCarron et al., 2017). Arguably, this is no different to the experience of those in the general population, but it has been noted that certain age-related health conditions are manifesting at an earlier stage in the intellectual disability population; one such condition is dementia.

In this chapter we will present current perspectives on dementia in the intellectual disability population, noting the nature and pattern of its precocious emergence in both those with and without Down syndrome. However, because of the significant prevalence of Alzheimer's dementia in those with Down syndrome, much of the discussion will draw from the copious literature on that subject. The ever-changing dementia-related terminology will be presented, underlying pathophysiology and aetiology considered and there will be a focus on the importance of – and challenges to – assessment and early diagnosis. Finally, some thoughts will be presented on the specific pharmacological concerns that are pertinent to the management of dementia-related symptoms in this population.

Dementia and Neurocognitive Disorder

As with many medical terms, the word 'dementia' (Latin. *de mens*), which literally means 'out of mind', may bring with it the stigmatising baggage

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of misunderstanding that is often embedded in culture (Mukadam & Livingston, 2012) and reinforced by print and visual media (Parker, 1955). And, whereas dementia remains the dominant term and formed the basis of diagnoses classified in *DSM-IV-TR* (American Psychiatric Association, 2000), the more recent *DSM-V* (American Psychiatric Association, 2013) favoured the term 'major neurocognitive disorder' (NCD). Janicki et al. (2017) note that the new terminology sought to highlight that neurocognitive disorder represented a spectrum of conditions, with variation in aetiology, locus of functional loss, and presentation of symptoms. As with its predecessor, though, these diagnostic criteria, drawn largely from evidence in the general population, do not translate well in the intellectual disability population (Poindexter, Pary, Martin, & Vicari, 2007) and have also found mixed reception in mainstream dementia and Alzheimer communities (Janicki et al., 2017), with older terms still in common usage. For the purposes of this chapter, dementia and NCD are used interchangeably.

The International Classification of Diseases *ICD-10* (World Health Organization, 2004) classifies dementia under mental and behavioural disorders and, although this book focuses on these dual diagnoses, dementia cannot be easily categorised under either of these headings. True, dementia often results in manifestations of behaviours that may be problematic and significantly affect mental health, but these are symptoms of the condition, rather than overarching descriptors. Dementia has been described as a catch-all term for a range of conditions and illnesses that share a common outcome of progressive loss of brain function (Janicki et al., 2017; Krinsky-Mchale & Silverman, 2013). It is a syndrome characterised by a decline in cognitive function, 'involving learning and memory, language, executive function, complex attention, perceptual-motor, social cognition' (Larson, 2018), which impacts on the person's ability to conduct daily living activities and maintain independence (Livingston et al., 2017). This is mediated through neuronal death, neuritic plaques, and neurofibrillary tangles and is described as being clinically progressive with irreversible deterioration in cognition, behaviour, and day-to-day functional ability (Jack & Holtzman, 2013; McKhann et al., 2011).

It has been noted that there are various types of dementia, the three most common of which are dementia in Alzheimer's disease, vascular dementia, and dementia related to other diseases. These are typically related to advancing age and pose major health challenges as an estimated 47 million people are affected worldwide (Corriveau et al., 2017), with Alzheimer's disease accounting for approximately two-thirds of cases (ibid). Such challenges are also posed to persons with an intellectual disability, but the key difference is the age of onset. While, prevalence of dementia in the general population in Western Europe is estimated to be 6.80% in those ages 60+ (Ali, Guerchet, Wu, Prince, & Prina, 2015), the reported prevalence in the intellectual disability population without Down syndrome is 18.3% in those aged 65+ with increased numbers seen in younger cohorts than in the general population (Strydom, Hassiotis, King, & Livingston, 2009). Furthermore, the incidence in this population stands at more than five times that in the general population (Strydom, Chan, King, Hassiotis, & Livingston, 2013) When those with Down syndrome are considered, rates are higher with virtually all having pathophysiological changes typical of the disease (Godfrey & Lee, 2018; McCarron et al., 2017).

Increased dementia in those with intellectual disability have been attributed to reduced brain reserve, head injury-related brain trauma, emotional trauma, diet, obesity, physical inactivity, and poorer physical health (Evans et al., 2013). For those with Down syndrome it is intricately linked to overexpression of the amyloid-beta precursor (APP) gene, resulting in formation of amyloid plaques and neurofibrillary bodies so characteristic of Alzheimer's type dementia. The increased prevalence among those with Down syndrome is also linked to advancing age (Ballard, Mobley, Hardy, Williams, & Corbett, 2016; Bittles & Glasson, 2004; McCarron, McCallion, Reilly, & Mulryan, 2014a), with a

reported incidence five times greater than in the population without intellectual disability (Strydom et al., 2013).

Aetiology and Pathophysiology of Dementia

Alois Alzheimer first reported on 'a peculiar severe disease process of the cerebral cortex' in 1906 (Hippius & Neundorfer, 2003). The patient to whom he referred was a 50-year-old woman who had been admitted to a psychiatric service with paranoia, sleep disturbance, aggression, confusion, and memory disturbance. Following her death, Alzheimer conducted an autopsy and described histologically what later became known as plaques and neurofibrillary tangles. These amyloid plaques and phosphorylated tau tangles became the pathological hallmarks on Alzheimer's disease. Amyloid plaques are extracellular deposits of amyloid-beta (Aβ), and neurofibrillary tangles are composed of paired helical filaments with hyperphosphorylated tau proteins, neuronal loss, and synaptic loss (Anand, Gill, & Mahdi, 2014). With advancing age, there is a build-up of such pathologies with the inevitability of complex neurological anomalies.

Debate on the pathophysiology of Alzheimer's disease is ongoing, and it is posited that the prevailing Aβ theory may not completely account for the complex pathophysiology of the disease (Hardy, 2009).

Critiques do not argue that amyloid has no association with Alzheimer's disease – it is clear from decades of literature (Hardy (2009), Roberson et al. (2007)) that amyloid plaques play a significant role – but that amyloid, by itself, may not be sufficient in explaining the cause of the disease. Recent suggestions that the disease pathophysiology begins years (and possibly decades) before any symptoms of dementia occur (Ritchie & Ritchie, 2012) has moved the focus of research away from *reversing* existing amyloid burden, to, instead, *preventing* the disease process from beginning in the first place. This, however, requires identification of the earliest biomarkers of Alzheimer's disease, so that clinical trials can focus on their manipulation.

Alternative Hypotheses for Alzheimer's Disease

While there is criticism of the amyloid hypothesis, there have not been many credible alternative hypotheses that can explain the genetic data (Hardy, 2009). The *presenilin inhibition hypothesis* suggests that the loss of essential functions of presenilin could better explain Alzheimer's disease and neurodegeneration, whereby conditional inactivation of presenilin has been found to cause progressive memory loss and neurodegeneration in mouse models (Shen & Kelleher, 2007). While this hypothesis is consistent with genetic data, it lacks experimental data to support it (Hardy, 2009). The *double-hit hypothesis* (Small & Duff, 2008) suggests that while the amyloid cascade hypothesis is correct for cases of autosomal dominant Alzheimer's disease, in familial Alzheimer's disease, tau neuropathy also plays a role.

It should again be noted that alternative hypotheses do not discount the role of amyloid in Alzheimer's disease neuropathology but rather suggest additional potential causes.

Other additional processes that have been indicated in the complication process of Alzheimer's disease include:

- Tau
- Failure of neuronal cell cycle control
- Neuro-inflammation
- Oxidative damage
- Mitochondrial function
- Loss of Ca^{2+} homeostasis
- Glucose metabolism
- General metabolic compromise
- Source: Herrup (2015).

Herrup (2015) suggests that it is due to the length and complexity of the possible alternatives to the amyloid hypothesis that has led to the hesitancy to reject it outright.

Although amyloid is implicated in the neurogenesis of Alzheimer's disease, the disease pathway, and the interactions among multiple other potential factors, is not yet understood.

Pathophysiology of Alzheimer's Disease in People with Down Syndrome

Although the amyloid hypothesis has come in for some criticism, it is, however, useful for explaining the pathogenesis of Alzheimer's disease in people with Down syndrome. Down syndrome is caused by a triplication of chromosome 21, or trisomy 21. The amyloid precursor protein (APP) gene is located on the proximal portion of the long arm of chromosome 21 within a critical region (Asim, Kumar, Muthuswamy, Jain, & Agarwal, 2015) of chromosome 21 that causes full Down syndrome phenotypic manifestation when triplicated (Zigman, 2013; Zigman & Lott, 2007). This results in people with Down syndrome having an overexpression of APP as compared to those without Down syndrome (Jennings et al., 2015). This increase in APP leads to an excess in Aβ deposits (Annus et al., 2016). Tau protein has also been indicated as a contributor to the neuropathology of Alzheimer's disease in people with Down syndrome (Di Domenico et al., 2017), with a higher level of tau protein found in people with Down syndrome than in the general population (Head, Lott, Wilcock, & Lemere, 2016).

The theory that APP is a leading cause of Alzheimer's disease pathology in people with Down syndrome is supported by the findings that some older people with Down syndrome, but with only partial trisomy 21 lacking the third copy of chromosome 21, do not have the pathology of Alzheimer's disease (Doran et al., 2017; Prasher, Sajith, Mehta, Zigman, & Schupf, 2010). This highlights the role of triplication of the APP gene in the pathogenesis of Alzheimer's disease in people with Down syndrome (Prasher et al., 2010). Due to the genetically homogenous nature of trisomy 21, and Aβ as a cause for Alzheimer's disease in people with Down syndrome (Strydom et al., 2018), this population allows a unique opportunity to better understand the role of Aβ in the pathology of Alzheimer's disease. The inclusion of this population in dementia research is also vital from an ethical and an equity perspective, due to its extra high risk of developing dementia.

It has been shown that by age 40, practically all individuals with Down syndrome will have the relevant neuropathology – amyloid plaques and tau neurofibrillary tangles – of Alzheimer's disease (Lamar et al., 2011; Schupf et al., 2010). Again, although the neuropathology of Alzheimer's disease will be present in people with Down syndrome, not all such people will actually develop dementia, and this is vital if diagnostic overshadowing – where people with Down syndrome are misdiagnosed with dementia simply because there is the misbelief that dementia is inevitable in this population – is to be avoided. It is also important to note that there are certain risk factors and protective factors that may modify the clinical symptoms of dementia.

Clinical Manifestations

In the general population, the earliest clinical presentation of Alzheimer's disease is memory loss that interferes with everyday life functioning, specifically in the ability to retain new information (Alzheimer's Association, 2014; Lopez, 2011). Additional common presentations include problem-solving impairment, decline in ability to complete familiar daily tasks, disorientation of time, language difficulties, social withdrawal, and evident mood disturbances including apathy, depression, and aggression (Alzheimer's Association, 2014, Lopez, 2011).

Understanding the early clinical manifestations and progression of dementia in people with intellectual disability, and particularly Down syndrome, is critical if a timely diagnosis is to be obtained and presenting symptoms appropriately addressed. This also reduces the potential for dementia being misdiagnosed when there are other causes of the observed symptoms including those associated with mental ill health that could be treated and managed

(O'Caoimh, Clune, & Molloy, 2013). While presentations of the later stages of dementia in people with Down syndrome appear similar to those seen in the general population (McCarron et al., 2018; Strydom et al., 2010), there is currently no consensus description of the early presentations of dementia in this population. The complex cognitive phenotype of individuals with Down syndrome means that while there is usually a general intellectual disability, there are also additional specific deficits, including executive deficiencies in memory, function, and language (Strydom et al., 2018).

In the general population, changes in episodic memory are seen in the prodromal stage of dementia – that is before clinical dementia. A number of studies in a population of people with Down syndrome, however, have suggested that symptoms related to executive dysfunction may be the earliest signs of dementia (Deb & Braganza, 1999). Furthermore, Ball et al. (2006) found that, while there was some overlap in symptoms found in the general population, such as memory loss, confusion, deterioration, speech, personality, and behaviour changes, those associated with executive dysfunction, appeared prior to the typical memory loss in Down syndrome populations. Similarly Holland, Hon, Huppert, Stevens, and Watson (1998), reported that changes in behaviour, such as apathy, withdrawal, and stubbornness, were noticed by carers, when asked retrospectively, *prior* to a clinical diagnosis of dementia. In a systematic review, 15 studies were identified that examined the earliest presentation of dementia in people with Down syndrome (Lautarescu, Holland, & Zaman, 2017). Of these studies, nine identified change in frontal like symptoms, again associated with executive dysfunction, as the earliest symptoms of dementia. This correlates with findings from brain imaging which have shown the frontal region to be an initial site for Aβ deposition (Handen et al., 2012).

In a recent study, drawing on data from almost 300 individuals with Down syndrome and using a data-driven approach with event-based modelling, it was suggested that changes in visuospatial paired associated memory, hand eye coordination, and verbal fluency may be early sensitive signs of Alzheimer's disease. Changes in planning and rule shifting (both associated with executive function) appear later in the process (Firth et al., 2018b). A similar pattern was found in three of the studies included in the Lautarescu et al. (2017) systematic review.

Further longitudinal studies are needed in order to understand the progression of decline in and across a range of cognitive domains, taking into account premorbid level of intellectual disability. There is also a need for research into the presence of concomitant mental illness as this may either be a confounding factor, particularly depression (Strydom, Livingston, King, & Hassiotis, 2007), and may either mimic or mask the clinical manifestations of dementia.

Diagnostic Considerations and Differential Diagnosis

The diagnosis of dementia in people with intellectual disability is accompanied by particular challenges (Krinsky-Mchale & Silverman, 2013). One issue is the lack of recent clear diagnostic criteria, applicable to individuals with intellectual disability; neither ICD-10 (World Health Organization, 2004) nor DSM-IV-TR (American Psychiatric Association, 2000) discusses only the general nonintellectual disability population. Progression of the disease may also be difficult to assess, particularly due to the fact that a baseline of *normal* functioning may not be easily defined in people with intellectual disability (Krinsky-Mchale & Silverman, 2013). It is especially important, therefore, that a comprehensive assessment be undertaken. It is advised that this should, for reference, focus on the ICD-10 criteria, making accommodations for the presence of intellectual disability (Aylward, Burt, Thorpe, Lai, & Dalton, 1997).

One of the key diagnostic considerations is differentiating decline due to dementia from premorbid level of intellectual disability (Rowe, Lavender, & Turk, 2006). Disentangling decline due to dementia from level of intellectual disability is challenging, especially since it has been found that scores on cognitive assessments in

adults with Down syndrome decline by the age of 50, regardless of dementia diagnosis, suggesting that there may be a confounding age-related decline (Carr & Collins, 2018). As previously noted, the onset of mental ill health or presence of a pre-existing mental health concern must also be borne in mind during assessment.

A number of unique challenges have been identified in the assessment of dementia in people with Down syndrome. Difficulties in communication can compromise the administration and interpretation of cognitive test results (Firth et al., 2018a). This said, international guidelines and best practice recommend that individuals with Down syndrome over the age of 35 years should have an annual cognitive screening, to maximise the chance of identifying changes at the earliest possible stage (Burt & Aylward, 2000). While this recommendation has been in place for many years, such screening is not common. This is also the situation for those individuals with intellectual disability unrelated to Down syndrome. Moran, Rafii, Keller, Singh, and Janicki (2013) highlight the importance of having a clear individualised baseline; this is, however, often very hard to ascertain. The lack of baseline scores means that noting change from previous level of functioning is difficult. This is further compounded by high staff turnover in service agencies for, with frequent changes of staff, an individual's deteriorating functioning due to dementia may go unnoticed and may instead be interpreted as baseline functioning (McCarron & Lawlor, 2003; McCarron & McCallion, 2004; Moran et al., 2013). As noted above, another challenge in timely diagnosis is the lack of consensus on what specific early changes should be expected. It is vital, though, that baseline function be established to allow for ongoing monitoring and assessment for any decline (Fig. 19.1).

In the general population, baseline functioning is often done with the individual, drawing on self-rated life history. The difficulties presenting in respect of individuals with an intellectual disability call for the use of both objective and informant measures. The adequacy of assessment tools presents additional problems as instruments used in the general population are often not accurate for this population due to *floor effect* – an individual with an intellectual disability may fail to score on an assessment due to generalised intellectual disability rather than due to dementia. Other aspects that may affect performance include prior skills, prior level of functioning, language skills, sensory impairment, psychiatric illness, and physical or neurological difficulties (National Institute for Clinical Excellence, 2006). Thus, it has been found that 75% of people with an intellectual disability may have been classified as having dementia using the Mini-Mental State Examination (MMSE), who were later found not to have dementia when other criteria were applied (Deb & Braganza, 1999). While there is no agreed battery of assessment for use with this population, a number of instruments have proven valid and reliable (Table 19.1).

Assessments should be repeated annually. Regular screening ensures that any changes in level of functioning are noted at the earliest possible stage. This also means that any other health conditions that may mimic dementia symptoms will be noted and can thus be treated. Jokinen et al. (2013) recommend that a number of actions can be undertaken in the pre-diagnostic stage, to support early identification of dementia:

- Provide for information needs of the person, family, friends, and staff regarding the ongoing diagnostic process and how dementia progresses.
- Undertake regular screenings to identify any early signs of the disease.
- Evaluate the effects of medications in masking or mimicking symptoms of dementia.
- Refer for formal dementia assessment by professionals with relevant expertise, if there are any concerns.
- Ensure that any person accompanying the person to an assessment is knowledgeable about the person being assessed and can provide accurate information.

Source: Jokinen et al. (2013)

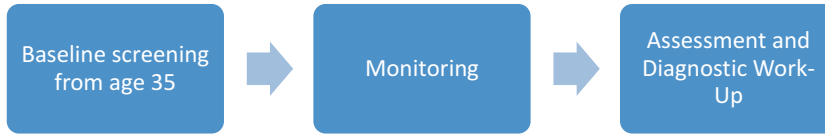

Fig. 19.1 The process for dementia screening and assessment

Table 19.1 Objective and informant measures for dementia, based on Jokinen et al. (2013)

Objective measures		Informant measures	
Test for severe impairment	Albert and Cohen (1992)	Dementia Questionnaire for People with Learning Disability (DLD)	Evenhuis, Kengen, and Eurlings (2007)
Prudhoe Cognitive Function Test	Kay et al. (2003)	Dementia Scale for Down syndrome (DSDS)	Gedye (1995)
Down Syndrome Mental Status Examination (DSMSE)	Haxby (1989)	Cambridge examination for Mental Disorders of the Elderly, modified for use for assessing people with Down syndrome (CAMDEX-DS)	Ball et al. (2004)
CAMCOG-DS	Huppert, Brayne, Gill, Paykel, and Beardsall (1995)	Dementia Screening Questionnaire for Individuals with intellectual Disability (DSQIID)	Deb et al. (2007)
Brief Praxis Test	Dalton, Mehta, Fedor, and Patti (1999)	Adaptive Behaviour Dementia Questionnaire (ABDQ)	Prasher, Adams, and Holder (2003)
Modified FULD Object-Memory Test	Seltzer (1997)		

In order to rule out other conditions that might produce symptoms that could be confused with dementia, physical, psychiatric, neurological, and auditory examinations are needed (McCarron & Lawlor, 2003).

The British Psychological Society (2015) and US Consensus recommendations (Jokinen et al., 2013) recommend that a physical check should include a full blood count, urea and electrolytes, blood sugar, thyroid function, liver function, folate and B12, lipid levels, as well as sensory testing (visual and auditory). The assessment should also take account of relevant history, physical examination, neuropsychological cognitive assessment, and medication review. Considering the potential for concomitant mental illness, particularly depression (Shooshtari, Martens, Burchill, Dik, & Naghipur, 2011; Tsiouris & Patti, 1997; Wark, Hussain, & Parmenter, 2014), psychiatric assessment should also be undertaken to identify any mental health concerns that could be related to or mimicking the symptoms of dementia (Wark et al., 2014).

Modifiable Risk Factors

There is currently no treatment available for Alzheimer's disease. For this reason, there has been an increasing interest in preventing or delaying its onset, particularly through modifiable risk factors. Risk factors (Table 19.2), which could account for 35% of the risk for dementia, have been identified for the general population (Livingston et al., 2017).

Cognitive Reserve

In the general population, it is well reported that, despite having the neuropathological manifestations of dementia, many people do not have the disease and are cognitively unimpaired

Table 19.2 Modifiable risk factors in Alzheimer's disease

Life stage	Risk factor	% modifiable
Early life	Education	8
Midlife	Hearing loss	9
	Hypertension	2
	Obesity	1
Late life	Smoking	5
	Depression	4
	Physical activity	3
	Diabetes	1
	Social interaction	2
Non-modifiable		65

Source: Livingston et al. (2017)

(Cholerton et al., 2016), and this has led to the notion of *cognitive reserve* being considered (Livingston et al., 2017). Cognitive reserve may enable individuals who have Alzheimer's type neuropathology to resist the typical presentation of cognitive and functional decline, meaning the development of the disease process is much slower than typically seen (Stern, 2012). Conversely, people with poor cognitive reserve may be at greater risk for the development of dementia (Livingston et al., 2017), and cognitive resilience with increasing age appears to be closely associated with the continual building and enhancement of the brain's cognitive reserve, earlier in life, through ongoing formal or informal education (Borenstein & Mortimer, 2016; Larson, 2010). One has, therefore, to consider if lower engagement in such educational processes, throughout life, is one reason why people with Down syndrome develop dementia at an earlier age (Strydom et al., 2007).

Various studies have examined how brain reserve may be enhanced and improved throughout the lifespan. Consensus findings point to three key areas: physical exercise, intellectual motivation, and social/leisure activities. Furthermore, brain reserve appears to be heavily associated with reduced incidence of dementia with increasing age, even where there are genetic risk factors for dementia (Wang, Sung, Chang, Wu, & Chuang, 2017). Healthy lifestyles and the notion of a healthy body equating with a healthy mind cannot be ignored. Annus et al. (2016) highlighted that amyloid accumulation appeared to manifest in people with Down syndrome in their early 40s and that the short dormant period between accumulation and cognitive decline could be attributed to poor cognitive reserve. The importance of increasing brain reserve and promoting increased cognitive resilience for people with Down syndrome is an important issue for future generations.

Memory Clinics

Memory clinics were first developed for the general ageing population, in the United States of America in the 1970s. Clinics offered assessment, diagnosis, treatment, and advice for people concerned about impaired memory, as well as post-diagnostic supports for family and carers. In the United Kingdom, the first memory clinics were established in the 1980s and were initially associated with drug trials. Ireland's first such clinic opened in 1991. Memory clinics for people with intellectual disability were, on the other hand, established later in the 1990s (Chicoine, McGuire, & Rublin, 1999) offering a comprehensive assessment service with post-diagnostic supports from a dedicated multidisciplinary dementia team (Bayley, Amoako, & El-Tahir, 2017; Hassiotis, Strydom, Allen, & Walker, 2003).

Memory clinics are independent units, primarily aimed at improving practice in the identification, investigation, and treatment of memory disorders, including dementia (Jolley, Benbow, & Grizzell, 2006; Nagamatsu et al., 2013). Early diagnosis of Alzheimer's disease, or any of the related dementias, is critical to facilitate timely and appropriate treatments, planning of services and access to appropriate supports (Cahill, Moore, & Pierce, 2009).

Specialist dementia clinics for older people with intellectual disability have proven effective in advancing assessment and care management and facilitating continued ageing in place for people with intellectual disability (Chicoine et al., 1999; Chicoine, McGuire, Hebein, & Gilly, 1995; McCreary, Fotheringham, Holden, Quellette-Kuntz, & Robertson, 1993). The key functions of such clinics are set out in Table 19.3.

Table 19.3 Key functions of memory clinics

Screening and follow-up
Diagnosis (including differential diagnosis)
Treatment information such as advice about anti-dementia medications and how they work
Education on ways to improve and maintain cognitive health
Training on diagnosis, treatment, and care of people with intellectual disability who experience symptoms of dementia including:
Understanding of early, middle, and late stage symptoms and behaviours
Understanding the impact of dementia on memory, mood, and communication
Recognition remaining strengths
Response to the behavioural and psychological symptoms of dementia
Preparing for decline in functional ability
Palliative care concerns

Memory clinics with specialised knowledge of dementia in people with intellectual disabilities are of value because the assessment of cognitive decline and subsequent diagnosis of dementia in persons with pre-existing intellectual disability presents significant challenges for staff at all levels in intellectual disability services organisations. Services are familiar with addressing the cognitive impairments associated with intellectual disability and have developed programmes and multidisciplinary responses to promote and support increasing independence and community participation. Symptoms of dementia challenge this approach because they are often masked until decline is quite substantial. With complex co-morbid health conditions frequently encountered, the trajectory is often one of steady decline in memory and functioning. In particular, cognitive implications of anxiety and depression, whether pre-existing or as a psychological symptom of dementia, may seriously impact the nature of this decline (Dodd et al., 2018). Consequently, the changes the person is experiencing may not be diagnosed or understood, programming models may no longer be appropriate, staff may not understand how and why the care provided needs to change, and the very environment in which the person lives may present hazards and barriers to appropriate care. Memory clinics offer the expertise to support better diagnostic processes.

Stages of Dementia

The management of risk factors and maximisation of function must be addressed in the context of the disease progression, from early to late stage dementia, and may form part of a therapeutic regimen aimed at maintaining function for as long as possible. Three main stages are usually identified in this progression: early, middle, and end stages.

Early Stage Dementia

The early stages of dementia are often associated with the appearance of subtle changes, with intermittent lapses in memory, and/or confusion in the conduct of familiar tasks (Jokinen et al., 2013). Changes in personality may become increasingly obvious, with evidence of increased apathy, irritability, and exaggeration of existing personality traits. Decreased empathy for others and functional decline may often be seen before signs of amnesia appear (Wilson, Annus, Zaman, & Holland, 2014). The onset of symptoms, such as memory loss, can be gradual and difficult to identify with decline often attributed to the person's underlying intellectual disability (Aylward et al., 1997). These early warning signs are often unrecognised or overlooked (Jokinen et al., 2013), and referrals delayed until changes in day-to-day behaviours or obvious functional decline are apparent (Adams et al., 2008), thus negating any focus on modifiable risk factors. During this period, there may be evidence of imperceptible decline in memory, particularly of recent events, and a decline in expressive speech and language skills with word finding difficulties becoming increasingly obvious; subtle evidence of agnosia or perceptual difficulties may also be apparent.

A sense of familiarity and maintaining a predictable routine can help compensate for any memory changes or disorientation the person may be experiencing (Janicki, Heller, Seltzer, & Hogg, 1996). The provision of enabling environments, with activities that are free of stress and failure, may sustain the overall well-being of the person (Kalsy-Lillico, 2014), while focusing on maintenance of remaining skills and emphasising a strength-based approach to care. Some early stage care strategies include:

- Maintain current level of functional ability and independence through increased supports and prompting.
- Build on preferred activities.
- Compensate for skills and abilities being lost.
- Provide memory aids/cues to support understanding and aid orientation to time, place, and person.
- Simplify routines and avoid unnecessary changes.
- Employ strategies to manage distress and anxiety associated with cognitive decline and elevated levels of confusion.
- Reduce demands and break tasks down, allowing the person to maintain their independence at their preferred pace.
- Identify environmental challenges to support 'Ageing in Place' through the continuum of the disease process.
- Focus on strengths to promote self-esteem, well-being, and dignity.
- Continuations of community programmes and day activation programmes.
- Provide purposeful engagement and meaningful activities with others to promote and maintain social connections with others.
- Provide education and training to help staff/carers develop skills in working with people with dementia.
- Care for family/carers and peers, helping to plan for the future.
- Ongoing surveillance of change. Record and document changes.

Source: Kalsy-Lillico (2014), Jokinen et al. (2013).

Mid-Stage Dementia

During the middle stages of dementia, symptoms become more pronounced and the decline in cognition and function become more apparent. Aphasia, anomia (inability to recall names of everyday objects) and word-finding difficulties cause difficulty in conversations and social participation. The person usually starts to isolate him/herself and fades into the background. The ability to follow simple conversation will become impaired to point whereby increased prompting is required. Disorientation to time, place and person will become increasingly more evident, and they will require on-going support to make sense of their world. With increased episodes of confusion and memory loss, coping skill begin to deteriorate and, consequently, there are increased incidences of anxiety and frustration (Janicki et al., 1996).

Decline in day-to-day functional ability progresses and ever-higher levels of care support become necessary. Activities of daily living are particularly compromised with dressing, personal care and simple routine chores becoming more difficult for the individual. Furthermore, bowel and bladder incontinence may become significant. Constant supervision and support is required and the person cannot be left alone, as wandering may expose him/her to danger (Janicki, McCallion, & Dalton, 2003).

Many services supporting people with intellectual disability have a strong emphasis on supporting people to promote and increase their levels of independence, autonomy and engagement in their local community (McCarron, McCallion, Reilly, & Mulryan, 2014b). However, in order to support those with dementia, there is need to restructure services, being cognisant of the dynamic changes in levels of need, and providing increased support and environmental modifications, particularly with a view to supporting 'ageing in place' for as long as possible. (Janicki et al., 1996).

People with Down syndrome, who are in the mid stages of Alzheimer's disease, may begin to experience late onset seizures. Such developments are well described in the literature and has been termed *late onset myoclonic epilepsy in*

Down syndrome (LOMEDS) (Sharma, Pandey, Kumawat, & Khandelwal, 2016). Prevalence of epilepsy has been noted to increase with advancing age in people with Down syndrome, particularly in those with Alzheimer's disease (McCarron, O'Dwyer, Burke, McGlinchey, & McCallion, 2014). Such developments may have important implications for the prognosis of the underlying dementia and can result in decline in cognition, language, decline in functional ability, and increased risk of mortality (Robertson, Hatton, Emerson, & Baines, 2015).

In mid-stage dementia, carers must consider diminishing cognitive function and capacity when making decisions regarding future care and treatment. Livingston et al. (2017) highlight that promoting optimal quality of life and maximising overall comfort of the person are the key considerations that should guide practice.

Mid-stage care strategies include:

- Maintain current level of functional ability and independence through increased supports and prompting.
- Keep communication simple and clear, use visual aids/cues.
- Use life story work to enhance communications; to link the person to their past, to ground them in the present and to help them maintain a sense of self.
- Keep to familiar routines and activities.
- Support Ageing in Place in a familiar, safe, calm home setting that is predictable to the individual.
- Care for family/carers and peers – plan for the future.
- Provide purposeful engagement and meaningful activities with others to promote and maintain social connections with others.
- Involvement in stimulating activities including music therapy; art therapy; reminiscence; story telling; drama etc.
- Protection and maintenance of safety.
- Training for carers. All staff should have the prerequisite knowledge to support progressive cognitive and functional decline and to manage the behavioural and psychological symptoms of dementia (BPSD).
- Attention should be given to carer burden and the level of stress carers/family may experience.
- Discussions regarding advance care directives and end of life care should be deliberated.
- Ongoing surveillance of change. Record and document changes.

Source: Kalsy-Lillico (2014), Jokinen et al. (2013).

Late Stage Dementia

While dementia is considered to be a terminal illness, there is often a failure by services to adopt a palliative approach to care (Livingston et al., 2017). Consequently, symptom management may be inadequately addressed, causing untold distress to both the individual and their family members. At this stage, the person will experience substantial dysfunction, requiring specialist attention and compassion. Both long- and short-term memories are lost, as is the ability to recognise familiar faces or their environment (Janicki et al., 1996). Basic skills such as eating and drinking are similarly lost and swallow status are severely compromised. The person is generally immobile and inactive, and concerns regarding skin integrity and risk of aspiration are paramount. Overall physical health can become compromised, and the person is at increased risk from respiratory and urinary tract infections. Pain assessment and its management are essential because, if left untreated it will impact on overall quality of life. Twenty-four-hour care is required, placing immense stress in family members who become the decision makers charged with making the emotionally demanding choices at end of life. Such key decisions relate to nutrition and hydration (percutaneous endoscopic gastrostomy) and resuscitation. A multidisciplinary approach to care is enacted to ensure good end of life care and optimal quality of life, bringing together physicians, clinical nurse specialist (dementia), nurses, psychologists, physiotherapists, occupational therapists, social workers, and spiritual care. Keeping family members informed

through the continuum of the disease from the diagnosis through to end of life can improve knowledge and alleviate concerns.

Key care strategies, at this point, include:

- Twenty-four-hour care
- Multidisciplinary approach
- Psychosocial activities cannot be underestimated in the late stages of dementia – music therapy, therapeutic massage, and touch, reminiscence
- Advance care planning in place, which is reviewed and updated as necessary
- Education and training to help staff/ carers develop skills in working with people with dementia at end of life
- Access to specialist services – palliative care team; spiritual care
- Provision of an environment appropriate to the person's needs

Source: Jokinen et al. (2013), Heller et al. (2018), Watchman et al. (2018).

Dementia-Specific Pharmacological Interventions

While there is no specific cure for dementia, anti-dementia pharmacological interventions have been proposed to treat symptoms. For those who do not have ID in the general population, there is some evidence that the acetylcholinesterase inhibitors, rivastigmine, galantamine or donepezil, and memantine, a non-competitive N-methyl-D-aspartate receptor antagonist, may improve cognitive function, behaviour, and quality of life for those with Alzheimer's disease (McShane, Sastre, & Minakaran, 2006). Adults with Down syndrome or intellectual disability from other aetiologies have often been excluded from clinical trials of drugs for dementia (Eady et al., 2018), meaning evidence regarding the effectiveness of these drugs in slowing cognitive decline adults with Down syndrome remains inconclusive (Hanney et al., 2012; Mohan, Bennett, & Carpenter, 2009a; Mohan, Bennett, & Carpenter, 2009b; Mohan, Carpenter, & Bennett, 2009). A small open-label study by Prasher et al. (2003) of 27 adults with intellectual disability identified significantly less deterioration in global functioning and adaptive behaviour in adults with Down syndrome treated with donepezil compared to a matched non-treated group over a two-year period. However, a Cochrane Review, examining pharmacological interventions for cognitive decline in adults with Down syndrome, concluded that due to low quality of the evidence, it is difficult to draw conclusions about effectiveness (Livingstone, Hanratty, McShane, & Macdonald, 2015).

A recent naturalistic clinical cohort study by Eady et al. (2018) investigated the effect of cholinesterase inhibitors or memantine on survival and function in 145 adults with Down syndrome (representing 47% of their study population who were taking drugs for dementia who had been diagnosed with Alzheimer's disease. Findings from the study indicated that median survival time for those taking drugs for dementia, mainly cholinesterase inhibitors, were significantly greater compared to those who were not prescribed medicines. When subsequent assessments took place, there was a positive effect on maintenance of cognitive function. The authors concluded that cholinesterase inhibitors appeared to be beneficial for adults with Down syndrome who had Alzheimer's disease. Hanney et al. (2012) carried out a prospective, randomised double blind placebo controlled clinical study which included participants with Down syndrome aged 40 years and over from four intellectual disability centres in the United Kingdom and Norway. Participants were randomly assigned to received memantine or placebo over a 52-week period. In this study no significant differences were observed in any of the outcome measures between the treatment groups at 26 or 52 weeks. The authors concluded that memantine treatment in this cohort was not beneficial with significant cognitive decline reported over the course of the study period.

In the general population, the mechanism by which medications for dementia may reduce mortality and increase survival time for those with Alzheimer's disease has not been unequivocally established. It has been hypothesised that the medications may provide protective effects on atherosclerotic cardiovascular risk (Nordström, Religa, Wimo, Winblad, & Eriksdotter, 2013). Given the relatively lower rates of atherosclerotic disease observed in adults with Down syndrome, this mechanism may have less relevance (Eady et al., 2018; Vis et al., 2009). In addition, older adults with intellectual disability who have dementia may be at risk of further cognitive decline due to higher likelihood of exposure to medicines, with anticholinergic properties, to treat other health conditions (Boustani, Campbell, Munger, Maidment, & Fox, 2008).

Clinical Guidelines

Clinical guidance published by the National Institute for Clinical Excellence provides clear, evidence-based guidance on the use of medication for people with dementia and at different stages of dementia, including adults with intellectual disability (National Institute for Clinical Excellence, 2018).

- The acetylcholinesterase (AChE) inhibitors donepezil, galantamine, and rivastigmine, as monotherapy are recommended options for the management of mild/moderate Alzheimer's disease.
- Memantine is recommended as monotherapy for management of Alzheimer's disease in people with moderate Alzheimer's disease who may be intolerant of or have a contraindication to AChE inhibitors of severe Alzheimer's disease.
- For those with established diagnosis of Alzheimer's disease who are already taking an AChE inhibitor, the guidelines recommend consideration of memantine in addition to an AChE inhibitor if there is a diagnosis of moderate disease.
- Memantine may also be prescribed in addition to an AChE inhibitor in the case of severe disease.
- A recommendation is not to stop cholinesterase inhibitors in people with Alzheimer's disease because of disease severity alone.
- Further, for those suffering from vascular dementia, the guideline advises that AChE inhibitors and memantine should not be prescribed; primarily due to lack of licence for use in vascular dementia, but also with regard to the pharmacodynamic potential, given the typical cardiovascular medicines in a person with vascular dementia, though evidence rejecting use of these anti-dementia drugs in persons with vascular dementia, is conflicting.

Source: National Institute for Clinical Excellence (2018).

Anecdotal evidence suggests that adults with Down syndrome respond to lower doses of anti-dementia drugs; therefore it may be feasible to maintain treatment at the lowest effective dose. Doses can be increased if symptoms later re-emerge (British Psychological Society, 2015).

There are few studies examining prevalence and patterns of drugs for dementia among adults with intellectual disability. A cohort study in Sweden compared medicines use between 7936 people with ID who had dementia and 7936 with dementia in the general population (Axmon, Kristensson, Ahlström, & Midlöv, 2017). In this study people with dementia in the ID cohort were less likely to report taking acetylcholinesterase inhibitors compared to the general older population (Axmon et al., 2017). Prevalence of drugs for dementia among older adults with intellectual disability who reported a diagnosis of dementia was also examined in the IDS-TILDA study in Ireland (Burke, McCallion, & McCarron, 2014; McCarron et al., 2011; McCarron, McCallion,

Carroll, et al., 2017). In Wave 1 of the study, 54% of those with dementia were receiving medications for dementia, with 35.4% of those at Wave 2, and 28.8% at Wave 3. Donepezil and memantine were most commonly reported, but there is no data on effectiveness.

Anticholinergic Medicines that May Cause or Contribute to Cognitive Impairment in Adults with Intellectual Disability

Anticholinergic medicines play a significant role in long-term drug-induced functional and cognitive impairment in the general older adult population. Reducing inappropriate use of these medicines represents an important intervention in preventing and reducing cognitive decline. Many medicines used to treat conditions that are prevalent in older adults, for example, antidepressants and drugs for urinary incontinence, possess central and peripheral anticholinergic activity, and may cause adverse effects such as sedation and confusion (Ruxton, Woodman, & Mangoni, 2015). Anticholinergic medicines are considered potentially inappropriate in older adults, particularly those with limited cognitive reserve, including adults who have Alzheimer's disease. A systematic review examining associations between drugs with anticholinergic effects and adverse outcomes in older adults carried out by Ruxton et al. (2015) concluded that exposure to individual medicines with anticholinergic effects or increased overall anticholinergic exposure may increase risk of cognitive impairment and all-cause mortality.

People with intellectual disability who have dementia represent a particularly vulnerable group, often experiencing a much higher anticholinergic burden due to exposure to polypharmacy to treat multiple age-related morbidities (O'Dwyer et al., 2016). In addition, epilepsy commonly co-occurs with dementia in those with Down syndrome, and some antiepileptic medicines have anticholinergic properties. Risk may be compounded when antipsychotics are prescribed for behavioural and psychological symptoms of dementia. With a co-occurring mental health condition, psychosis/schizophrenia where an antipsychotic is indicated, an atypical antipsychotic with a lower anticholinergic load may be a more appropriate choice. Many of these antipsychotics have significant anticholinergic activity. In findings from Wave 2 of the IDS-TILDA study (Burke et al., 2014), when medication use of those with intellectual disability and dementia was examined, 37.5% of those with dementia and Down syndrome reported exposure to antipsychotics, compared to 68% of those with dementia and another cause of intellectual disability ($p < 0.001$). This in part reflects the lower prevalence of mental health conditions reported by adults with Down syndrome (25.8%), compared to reported mental health conditions in those with intellectual disability of other causes (57.5%, $p < 0.001$). This study did not capture information as to whether any of the antipsychotics reported were being prescribed to treat the behavioural and psychological symptoms of dementia (Burke et al., 2014).

Cross-sectional findings by O'Dwyer et al. (2016), from Wave 1 of the IDS-TILDA study identified that 70% of older adults with intellectual disability were exposed to anticholinergic medicines. In addition, 26% had a very high anticholinergic exposure, compared to 2% of older adults in the general older population (Fox et al., 2011). There was a significant association between exposure at a cross-sectional level and adverse effects, e.g. sedation (O'Dwyer et al., 2016). It was not possible to examine those with dementia at a multivariate level, due to small numbers with a diagnosis of dementia. However, it was noted that, in those with dementia, 83.2% had anticholinergic exposure.

The use of anticholinergics represents a modifiable risk factor for the development of dementia and deterioration in cognitive function in adults with intellectual disability. Efforts should be made to reduce and limit use of medications with anticholinergic effects, particularly in those most vulnerable to cognitive decline. The National Task Force on Intellectual Disability and Dementia Practice Consensus Recommendations for Evaluation and

Management of Dementia in Adults with Intellectual Disability recommends reviewing the medication list 'thoroughly' paying special attention to medications that are 'psychoactive, antiepileptic or anticholinergic or those with sedating properties' (Moran et al., 2013). Where psychotropic medications are used for behavioural and psychological symptoms, target symptoms should be clearly recorded; risks and benefits should be discussed with the person and/or carers, and the minimum effective dose should be used for the shortest length of time possible (British Psychological Society, 2015).

Medications have a role to play in the management of dementia, particularly when there are co-occurring psychiatric and other health conditions, and the potential for improved functioning from anti-dementia medications deserves further investigation. However, the potential for unfavourable interactions and for increased anticholinergic burden means that caution in their use and monitoring are necessary.

Conclusion

In recent decades, many developed countries have seen improved social, nutritional, and public health standards, as well as advances in healthcare. These have resulted in many benefits, notably greater longevity, with people achieving lifespans far beyond those of past generations. Such changes in longevity have also been witnessed among people with intellectual disabilities, but they have revealed a propensity for the development of dementia at rates greater than what is seen in the general population. This is particularly the case among those people who have Down syndrome. The occurrence of dementia in people with intellectual disability poses challenges for diagnosis, management, and treatment, but recent work has resulted in the development of consensus statements on nomenclature and diagnostic criteria as well as recommendations for support and management through the progression of the disease. These are leading to ongoing improvements in the healthcare response to those with intellectual disability who are living with dementia.

References

Adams, D., Oliver, C., Kalsy, S., Peters, S., Broquard, M., Basra, T., … McQuillan, S. (2008). Behavioural characteristics associated with dementia assessment referrals in adults with Down syndrome. *Journal of Intellectual Disability Research, 52*, 358–368.

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.

Ali, G.-C., Guerchet, M., Wu, Y.-T., Prince, M., & Prina, M. (2015). The global prevalence of dementia. In M. Prince, A. Wimo, M. Guerchet, G.-C. Ali, Y.-T. Wu, & M. Prina (Eds.), *World Alzheimer report 2015: The global impact of dementia*. London, UK: Alzheimer's Disease International.

Alzheimer's Association. (2014). 2014 Alzheimer's disease facts and figures. *Alzheimers Dement, 10*, e47–e92.

American Psychiatric Association. (2000). *Diagnostic and statistical manual of mental disorders: DSM-IV-TR*. Washington, DC: American Psychiatric Association.

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

Anand, R., Gill, K. D. & Mahdi, A. A. 2014. Therapeutics of Alzheimer's disease: Past, present and future. *Neuropharmacology, 76*(Pt A), 27–50.

Annus, T., Wilson, L. R., Hong, Y. T., Acosta-Cabronero, J., Fryer, T. D., Cardenas-Blanco, A., … Holland, A. J. (2016). The pattern of amyloid accumulation in the brains of adults with Down syndrome. *Alzheimers Dement, 12*, 538–545.

Asim, A., Kumar, A., Muthuswamy, S., Jain, S. & Agarwal, S. 2015. Down syndrome: an insight of the disease. Journal of Biomedical Science, 22, 41.

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.

Aylward, E. H., Burt, D. B., Thorpe, L. U., Lai, F., & Dalton, A. (1997). Diagnosis of dementia in individuals with intellectual disability. *Journal of Intellectual Disability Research, 41*(Pt 2), 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 prospec-

tive population-based study. *International Journal of Geriatric Psychiatry: A Journal of the Psychiatry of Late Life Allied Sciences, 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.

Ballard, C., Mobley, W., Hardy, J., Williams, G., & Corbett, A. (2016). Dementia in Down's syndrome. *The Lancet Neurology, 15*, 622–636.

Bayley, A., Amoako, A., & El-Tahir, M. O. (2017). Service evaluation of a specialist memory clinic for adults with ID in South Wales. *Advances in Mental Health and Intellectual Disabilities, 11*, 145–154.

Bittles, A. H., & Glasson, E. J. (2004). Clinical, social, and ethical implications of changing life expectancy in Down syndrome. *Developmental Medicine and Child Neurology, 46*, 282–286.

Borenstein, A., & Mortimer, J. (2016). *Alzheimers disease – Life course perspectives on risk reduction*. Amsterdam, Netherlands: Academic Press.

Boustani, M., Campbell, N., Munger, S., Maidment, I. & Fox, C. (2008). Impact of anticholinergics on the aging brain: A review and practical application. *Aging Health*, *4*, 311–320.

British Psychological Society (2015). *Dementia and people with intellectual disabilities.* British Psychological Society: Leicester, United Kingdom.

Burke, E., McCallion, P., & McCarron, M. (2014). *Advancing years, different challenges: Wave 2 IDS-TILDA*. Dublin, Ireland: Trinity College Dublin.

Burt, D. B., & Aylward, E. H. (2000). Test battery for the diagnosis of dementia in individuals with intellectual disability. *Journal of Intellectual Disability Research, 44*, 175–180.

Cahill, S., Moore, V., & Pierce, M. (2009). *Memory clinics in Ireland: A guide for family caregivers and health service professionals*. Trinity College Dublin: Dublin, Ireland.

Carr, J., & Collins, S. (2018). 50 years with Down syndrome: A longitudinal study. *Journal of Applied Research in Intellectual Disabilities, 31*, 743–750.

Chicoine, B., McGuire, D., Hebein, S., & Gilly, D. (1995). Use of the community-orientated primary care model for a special needs population. A Clinic for Adults with Down syndrome. *American Journal of Public Health, 85*, 869–870.

Chicoine, B., McGuire, D., & Rublin, S. R. (1999). Speciality clinic perspectives. In M. P. Janicki & A. J. Dalton (Eds.), *Dementia ageing and intellectual disabilities*. Didcot, UK: Taylor and Francis.

Cholerton, B., Larson, E. B., Quinn, J. F., Zabetian, C. P., Mata, I. F., Keene, C. D., … Montine, T. J. (2016). Precision medicine: Clarity for the complexity of dementia. *The American Journal of Pathology, 186*, 500–506.

Corriveau, R. A., Koroshetz, W. J., Gladman, J. T., Jeon, S., Babcock, D., Bennett, D. A., … Holtzman, D. M. (2017). Alzheimer's disease-related dementias summit 2016: National research priorities. *Neurology, 89*, 2381–2391.

Dalton, A., Mehta, P., Fedor, B., & Patti, P. (1999). Cognitive changes in memory precede those in praxis in aging persons with Down syndrome. *Journal of Intellectual Developmental Disability, 24*, 169–187.

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*, 400–407.

Deb, S., Hare, M., Prior, L. & Bhaumik, S. (2007). Dementia Screening Questionnaire for Individuals with Intellectual Disabilities. *British Journal of Psychiatry* 190 (5):440–444.

Di Domenico, F., Tramutola, A., Head, E., Foppolia, C., Perluigi, M., & Butterfield, D. A. (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 & Medicine, 112*, 70.

Dodd, K., Watchman, K., Janicki, M. P., Coppus, A., Gaertner, C., Fortea, J., … Strydom, A. (2018). Consensus statement of the international summit on intellectual disability and dementia related to post-diagnostic support. *Aging & Mental Health, 22*, 1406–1415.

Doran, E., Keator, D., Head, E., Phelan, M. J., Kim, R., Totoiu, M., … Lott, I. T. (2017). Down syndrome, partial trisomy 21, and absence of Alzheimer's disease: The role of APP. *Journal of Alzheimer's Disease: JAD, 56*, 459–470.

Eady, N., Sheehan, R., Rantell, K., Sinai, A., Bernal, J., Bohnen, I., … Gazizova, D. (2018). Impact of cholinesterase inhibitors or memantine on survival in adults with Down syndrome and dementia: Clinical cohort study. *The British Journal of Psychiatry, 212*, 155–160.

Evans, E., Bhardwaj, A., Brodaty, H., Sachdev, P., Draper, B., & Trollor, J. N. (2013). Dementia in people with intellectual disability: Insights and challenges in epidemiological research with an at-risk population. *International Review of Psychiatry (Abingdon, England), 25*, 755–763.

Evenhuis, H. M., Kengen, M. M., & Eurlings, H. A. 2007. *Dementia questionnaire for people with learning disabilities*, Pearson Assessment.

Firth, N. C., Startin, C. M., Hithersay, R., Hamburg, S., Wijeratne, P. A., Mok, K. Y., … Strydom, A. (2018a). Aging related cognitive changes associated with Alzheimer's disease in Down syndrome. *Annals of Clinical Translational Neurology, 5*, 741–751.

Firth, N. C., Startin, C. M., Hithersay, R., Hamburg, S., Wijeratne, P. A., Mok, K. Y., … Consortium, L. (2018b). Sequence of cognitive changes associated with development of Alzheimer's disease in Down syndrome-data driven analysis. *bioRxiv*, 263095.

Fox, C., Richardson, K., Maidment, I. D., Savva, G. M., Matthews, F. E., Smithard, D., … Brayne, C. (2011). Anticholinergic medication use and cognitive impairment in the older population: The medical research

council cognitive function and ageing study. *Journal of the American Geriatrics Society, 59*, 1477–1483.
Gedye, A. (1995). Dementia scale for down syndrome: Manual, A. Gedye.
Godfrey, M., & Lee, N. R. (2018). Memory profiles in Down syndrome across development: A review of memory abilities through the lifespan. *Journal of Neurodevelopmental Disorders, 10*, 5–5.
Handen, B. L., Cohen, A. D., Channamalappa, U., Bulova, P., Cannon, S. A., Cohen, W. I., … Klunk, W. E. (2012). Imaging brain amyloid in nondemented young adults with Down syndrome using Pittsburgh compound B. *Alzheimers Dement, 8*, 496–501.
Hanney, M., Prasher, V., Williams, N., Jones, E. L., Aarsland, D., Corbett, A., … Francis, P. T. (2012). Memantine for dementia in adults older than 40 years with Down's syndrome (MEADOWS): A randomised, double-blind, placebo-controlled trial. *The Lancet, 379*, 528–536.
Hardy, J. (2009). The amyloid hypothesis for Alzheimer's disease: A critical reappraisal. *Journal of Neurochemistry, 110*, 1129–1134.
Hartley, D., Blumenthal, T., Carrillo, M., Dipaolo, G., Esralew, L., Gardiner, K., … Wisniewski, T. (2015). Down syndrome and Alzheimer's disease: Common pathways, common goals. *Alzheimer's & Dementia: The Journal of the Alzheimer's Association, 11*, 700–709.
Hassiotis, A., Strydom, A., Allen, K., & Walker, Z. (2003). A memory clinic for older people with intellectual disabilities. *Aging & Mental Health, 7*, 418–423.
Haxby, J. V. (1989). Neuropsychological evaluation of adults with Down's syndrome: Patterns of selective impairment in non-demented old adults. *Journal of Mental Deficiency Research, 33*(Pt 3), 193–210.
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.
Heller, T., Scott, H. M., Janicki, M. P., Heller, T., Esbensen, A., Fazio, S., … Wheeler, B. (2018). Caregiving, intellectual disability, and dementia: Report of the summit workgroup on caregiving and intellectual and developmental disabilities. *Alzheimer's & Dementia: Translational Research & Clinical Interventions, 4*, 272–282.
Herrup, K. (2015). The case for rejecting the amyloid cascade hypothesis. *Nature Neuroscience, 18*, 794–799.
Hippius, H., & Neundorfer, G. (2003). The discovery of Alzheimer's disease. *Dialogues in Clinical Neuroscience, 5*, 101–108.
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.
Huppert, F. A., Brayne, C., Gill, C., Paykel, E., & Beardsall, L. (1995). CAMCOG—A concise neuropsychological test to assist dementia diagnosis: Socio-demographic determinants in an elderly population sample. *British Journal of Clinical Psychology, 34*, 529–541.
Jack, C. R., Jr., & Holtzman, D. M. (2013). Biomarker modeling of Alzheimer's disease. *Neuron, 80*, 1347–1358.
Janicki, M. P., Heller, T., Seltzer, G. B., & Hogg, J. (1996). Practice guidelines for the clinical assessment and care management of Alzheimer's disease and other dementias among adults with intellectual disability∗. *Journal of Intellectual Disability Research, 40*, 374–382.
Janicki, M. P., McCallion, P., & Dalton, A. J. (2003). Dementia-related care decision-making in group homes for persons with intellectual disabilities AU – Janicki, Matthew P. *Journal of Gerontological Social Work, 38*, 179–195.
Janicki, M. P., McCallion, P., Splaine, M., Santos, F. H., Keller, S. M., & Watchman, K. (2017). Consensus statement of the international summit on intellectual disability and dementia related to nomenclature. *Intellectual and Developmental Disabilities, 55*, 338–346.
Jennings, D., Seibyl, J., Sabbagh, M., Lai, F., Hopkins, W., Bullich, S., … Marek, K. (2015). Age dependence of brain beta-amyloid deposition in Down syndrome: An [18F]florbetaben PET study. *Neurology, 84*, 500–507.
Jokinen, N., Janicki, M. P., Keller, S. M., McCallion, P., Force, L. T., Disabilities, N. T. G. O. I., & Practices, D. (2013). Guidelines for structuring community care and supports for people with intellectual disabilities affected by dementia. *Journal of Policy and Practice in Intellectual Disabilities, 10*, 1–24.
Jolley, D., Benbow, S. M., & Grizzell, M. (2006). Memory clinics. *Postgraduate Medical Journal, 82*, 199–206.
Kalsy-Lillico, S. (2014). Living life with dementia: Enhancing psychological wellbeing. In K. Watchman (Ed.), *Intellectual disability and dementia: Research into practice*. London, UK: Jessica Kingsley.
Kay, D. W. K., Tyrer, S. P., Margallo-Lana, M. L., Moore, P. B., Fletcher, R., Berney, T. P. & Vithayathil E. (2003). Preliminary evaluation of a scale to assess cognitive function in adults with Down's syndrome: the Prudhoe Cognitive Function Test. *Journal of Intellectual Disability Research* 47, 155–168.
Krinsky-Mchale, S. J., & Silverman, W. (2013). Dementia and mild cognitive impairment in adults with intellectual disability: Issues of diagnosis. *Developmental Disabilities Research Reviews, 18*, 31–42.
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.
Larson, E. B. (2010). Prospects for delaying the rising tide of worldwide, late-life dementias. *International Psychogeriatrics, 22*, 1196–1202.
Larson, E. B. (2018). *Evaluation of cognitive impairment and dementia.* [Online]. Wolters Kluwer. Available: https://www.uptodate.com/contents/evaluation-of-cognitive-impairment-and-dementia. Accessed 25th January 2019.

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., … Mukadam, N. (2017). Dementia prevention, intervention, and care. *The Lancet, 390*, 2673–2734.

Livingstone, N., Hanratty, J., McShane, R., & Macdonald, G. (2015). Pharmacological interventions for cognitive decline in people with Down syndrome. *Cochrane Database of Systematic Reviews, 10*. Art. No.: CD011546.

Lopez, O. L. (2011). The growing burden of Alzheimer's disease. *The American Journal of Managed Care, 17*(Suppl 13), S339–S345.

McCarron, M., Carroll, R., Kelly, C., & McCallion, P. (2015). Mortality rates in the general Irish population compared to those with an intellectual disability from 2003 to 2012. *Journal of Applied Research in Intellectual Disabilities, 28*, 406–413.

McCarron, M., & Lawlor, B. A. (2003). Responding to the challenges of ageing and dementia in intellectual disability in Ireland. *Aging & Mental Health, 7*, 413–417.

McCarron, M., & McCallion, P. (2004). Intellectual disabilities and dementia. In K. Doka (Ed.), *Living with grief: Alzheimer's disease*. Washington, DC: Hospice Foundation of America.

McCarron, M., McCallion, P., Carroll, R., Burke, E., McGlinchey, E., O'Donovan, M., … Ryan, J. (2017). *Health, wellbeing and social inclusion: Ageing with an intellectual disability in Ireland*. Dublin, Ireland: Trinity College Dublin.

McCarron, M., McCallion, P., Coppus, A., Fortea, J., Stemp, S., Janicki, M., & Wtachman, 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*, 617–624.

McCarron, M., McCallion, P., Reilly, E., Dunne, P., Carroll, R., & Mulryan, N. (2017). A prospective 20-year longitudinal follow-up of dementia in persons with Down syndrome. *Journal of Intellectual Disability Research: JIDR, 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: JIDR, 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. In K. Watchman (Ed.), *Intellectual disability and dementia: Research into practice*. London, UK: Jessica Kingsley.

McCarron, M., O'Dwyer, M., Burke, E., McGlinchey, E., & McCallion, P. (2014). Epidemiology of epilepsy in older adults with an intellectual disability in Ireland: Associations and service implications. *American Journal on Intellectual and Developmental Disabilities, 119*, 253–260.

McCarron, M., Swinburne, J., Burke, E., McGlinchet, E., Mulryan, N., Andrews, V., … McCallion, P. (2011). *Growing older with an intellectual disability in Ireland in 2011*. Dublin, Ireland: Trinity College Dublin.

McCreary, B. D., Fotheringham, J. B., Holden, J. J. A., Quellette-Kuntz, H., & Robertson, D. M. (1993). Experience in an Alzheimer's clinic for persons with Down syndrome. In J. M. Berg, H. Karlinsky, & A. J. Holland (Eds.), *Alzheimer's disease, Down syndrome and their relationship*. Oxford, UK: Oxford University Press.

McKhann, G. M., Knopman, D. S., Chertkow, H., Hyman, B. T., Jack, C. R., Jr., Kawas, C. H., … Phelps, C. H. (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. *Alzheimers Dement, 7*, 263–269.

McShane, R., Sastre, A. A., & Minakaran, N. (2006). Memantine for dementia. *Cochrane Database of Systematic Reviews*. 2, CD003154.

Mohan, M., Bennett, C., & Carpenter, P. K. (2009a). Galantamine for dementia in people with Down syndrome. *Cochrane Database of Systematic Reviews*. 1, CD007656.

Mohan, M., Bennett, C., & Carpenter, P. K. (2009b). Rivastigmine for dementia in people with Down syndrome. *Cochrane Database of Systematic Reviews*. 1, CD007658.

Mohan, M., Carpenter, P. K., & Bennett, C. (2009). Donepezil for dementia in people with Down syndrome. *Cochrane Database of Systematic Reviews*. 1, CD007178.

Moran, J. A., Rafii, M. S., Keller, S. M., Singh, B. K., & Janicki, M. P. (2013). The National Task Group on Intellectual Disabilities and Dementia Practices consensus recommendations for the evaluation and management of dementia in adults with intellectual disabilities. Mayo Clinic Proceedings, Elsevier, 831–840.

Mukadam, N., & Livingston, G. (2012). Reducing the stigma associated with dementia: Approaches and goals. *Aging Health, 8*, 377–386.

Nagamatsu, L. S., Chan, A., Davis, J. C., Beattie, B. L., Graf, P., Voss, M. W., … Liu-Ambrose, T. (2013). Physical activity improves verbal and spatial memory in older adults with probable mild cognitive impairment: A 6-month randomized controlled trial. *Journal of Aging Research, 2013*, 861893.

National Institute for Clinical Excellence. (2006). *Dementia: supporting people with dementia and their carers in health and social care*. London, UK: NICE.

National Institute for Clinical Excellence. (2018). *Dementia: Assessment, management and support for people living with dementia and their carers*. London, UK: NICE.

Nordström, P., Religa, D., Wimo, A., Winblad, B., & Eriksdotter, M. (2013). The use of cholinesterase

inhibitors and the risk of myocardial infarction and death: A nationwide cohort study in subjects with Alzheimer's disease. *European Heart Journal, 34*, 2585–2591.

O'Caoimh, R., Clune, Y., & Molloy, W. (2013). Screening for Alzheimer's disease in downs syndrome. *Alzheimers Dis Parkinsonism, 7*, 2161–0460. *Dementia,* 1955. Directed by Parker, J. United States of America.

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, 115*, 173971.

Parker, J. (Producer & Director). (1955). *Dementia* [Motion Picture]. United States of America: Cornerstone Media.

Poindexter, A., Pary, R., Martin, M., & Vicari, S. (2007). Delirium, dementia, and amnestic disorders. In R. Fletcher, E. Loschen, C. Stavrakaki, & M. First (Eds.), *Diagnostic manual – Intellectual disability: DM-ID*. New York, NY: NADD Press.

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*, 549–551.

Prasher, V. P., Sajith, S. G., Mehta, P., Zigman, W. B., & Schupf, N. (2010). Plasma beta-amyloid and duration of Alzheimer's disease in adults with Down syndrome. *International Journal of Geriatric Psychiatry, 25*, 202–207.

Ritchie, C. W., & Ritchie, K. (2012). The PREVENT study: a prospective cohort study to identify mid-life biomarkers of late-onset Alzheimer's disease. *British Medical Journal Open, 2, 1-6*:e001893.

Roberson, E. D., Scearce-Levie, K., Palop, J. J., Yan, F., Cheng, I. H., Wu, T., … Mucke, L. (2007). Reducing endogenous tau ameliorates amyloid ß-induced deficits in an Alzheimer's disease mouse model. *Science & Sports, 316*, 750–754.

Robertson, J., Hatton, C., Emerson, E., & Baines, S. (2015). Prevalence of epilepsy among people with intellectual disabilities: A systematic review. *Seizure, 29*, 46–62.

Rowe, J., Lavender, A., & Turk, V. (2006). Cognitive executive function in Down's syndrome. *The British Journal of Clinical Psychology, 45*, 5–17.

Ruxton, K., Woodman, R. J., & Mangoni, A. A. (2015). Drugs with anticholinergic effects and cognitive impairment, falls and all-cause mortality in older adults: A systematic review and meta-analysis. *British Journal of Clinical Pharmacology*. *80,* 921–926.

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. (1997). *Modified Fuld Object Memory Evaluation*. Madison, WI: Waisman Centre, University of Wisconsin Madison.

Sharma, C. M., Pandey, R. K., Kumawat, B. L., & Khandelwal, D. (2016). Late-onset myoclonic epilepsy in Down syndrome (LOMEDS): A spectrum of progressive myoclonic epilepsy – Case report. *Annals of Indian Academy of Neurology, 19*, 267–268.

Shen, J., & Kelleher, R. J., 3rd. (2007). The presenilin hypothesis of Alzheimer's disease: evidence for a loss-of-function pathogenic mechanism. *Proceedings of the National Academy of Sciences of the United States of America, 104*, 403–409.

Shooshtari, S., Martens, P. J., Burchill, C. A., Dik, N., & Naghipur, S. (2011). Prevalence of depression and dementia among adults with developmental disabilities in Manitoba, Canada. *International Journal of Family Medicine, 2011*, 319574–319574.

Small, S. A., & Duff, K. (2008). Linking Aβ and tau in late-onset Alzheimer's disease: A dual pathway hypothesis. *Neuron, 60*, 534–542.

Stern, Y. (2012). Cognitive reserve in ageing and Alzheimer's disease. *The Lancet Neurology, 11*, 1006–1012.

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., Coppus, A., Blesa, R., Danek, A., Fortea, J., Hardy, J., … Zetterberg, H. (2018). Alzheimer's disease in down syndrome: An overlooked population for prevention trials. *Alzheimers Dement (N Y), 4*, 703–713.

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., Livingston, G., King, M., & Hassiotis, A. (2007). Prevalence of dementia in intellectual disability using different diagnostic criteria. *The British Journal of Psychiatry: the Journal of Mental Science, 191*, 150–157.

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 Practice in Intellectual Disabilities, 7*, 96–110.

Strydom A., Startin, C., Hithersay, R., Hamburg, S., Mok, K. Y., Hardy, J. A., Alexander, D. C. & Firth, N. C. 2018. Sequence of cognitive decline in adults with Down syndrome during progression from preclinical to prodromal Alzheimer's disease. Alzheimer's Dementia: The Journal of the Alzheimer's Association, 14**,** P235-P236.

Torr, J., & Davis, R. (2007). Ageing and mental health problems in people with intellectual disability. *Current Opinion in Psychiatry, 20*, 467–471.

Tsiouris, J. A., & Patti, P. J. (1997). Drug treatment of depression associated with dementia or presented as 'Pseudodementia' in older adults with Down syndrome. *Journal of Applied Research in Intellectual Disabilities, 10*, 312–322.

Vis, J., Duffels, M., Winter, M., Weijerman, M., Cobben, J., Huisman, S., & Mulder, B. (2009). Down syn-

drome: A cardiovascular perspective. *Journal of Intellectual Disability Research, 53*, 419–425.

Wang, J.-C., Sung, W.-H., Chang, Y.-L., Wu, S.-H., & Chuang, T.-Y. (2017). Speed and temporal-distance adaptations during non-motorized treadmill walking in stroke and non-disabled individuals. *European Journal of Physical and Rehabilitation Medicine, 53*, 863–869.

Wark, S., Hussain, R., & Parmenter, T. (2014). Down syndrome and dementia:Is depression a confounder for accurate diagnosis and treatment? *Journal of Intellectual Disabilities, 18*, 305–314.

Watchman, K., Janicki, M. P., Udell, L., Hogan, M., Quinn, S., & Beránková, A. (2018). Consensus statement of the international summit on intellectual disability and dementia on valuing the perspectives of persons with intellectual disability. *Journal of Intellectual Disabilities*, 1744629517751817.

Wilson, L. R., Annus, T., Zaman, S. H., & Holland, A. (2014). Understanding the process: Links between Down syndrome and dementia. In K. Watchman (Ed.), *Intellectual disabilities and dementia*. London, UK: Jessica Kingsley Publishers.

World Health Organization. (2004). *ICD-10 : International statistical classification of diseases and related health problems / World Health Organization*. Geneva, Switzerland: World Health Organization.

Zigman, W. B. (2013). Atypical aging in down syndrome. *Developmental Disabilities Research Reviews, 18*, 51–67.

Zigman, W. B., & Lott, I. T. (2007). Alzheimer's disease in Down syndrome: Neurobiology and risk. *Mental Retardation and Developmental Disabilities Research Reviews, 13*, 237–246.

20 Considerations in the Assessment and Treatment of Aggression and Disruption

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Definition and Prevalence

Aggression can generally be defined as any behavior that inflicts bodily harm on another person and may include specific acts such as hitting, kicking, biting, pulling hair, and throwing objects at another person. Disruption can be defined as behavior that results in potential or actual damage to property, such as ripping, throwing, or kicking objects. Disruption includes repetitive or stereotyped behaviors that may interrupt ongoing activities (e.g., screaming, humming, vocal perseveration, and off-task behavior). Note that this general definition includes property destruction, which has sometimes been defined separately from disruption in previous studies. Unless otherwise noted, "disruption" will be used for any of these forms of aberrant behavior throughout this chapter.

Individuals who engage in aggression, in particular, may have more limited access to community services, are more likely to be prescribed antipsychotic medications, and may be referred for specialized behavioral assessment and treatment (Allen, 2000; Crocker et al., 2006; Emerson, Robertson, Gregory, Hatton, Kessissoglou, Hallam, & Hillery, 2000). Further, these behaviors pose significant concerns to staff and other caretakers increasing the costs of treating individuals diagnosed (Gardner & Moffatt, 1990; Wehman & McLaughlin, 1979). Finally, families of individuals who engage in problem behavior are at elevated risk for significant stressors such as financial strain, marital stress, and lack of additional resources to support their child and other children in the home (Smith, Oliver, & Innocenti (2001).

Aggression and disruption have been reported to be more common in individuals with intellectual and developmental disabilities (IDD) and may covary with the severity of impairment (IDD; e.g., Allen, 2000; Dworschak, Ratz, & Wagner, 2016; McClintock, Hall, & Oliver, 2003; Poppes, van der Putten, & Vlaskamp, 2010). That is, individuals with more severe IDD may be more likely to engage in severe forms of challenging behavior such as aggression and disruption (Borthwick-Duffy, Lane, & Widaman, 1997). Aggression may be more common in males and individuals with deficits in expressive communication, whereas both aggression and disruption may be more common in individuals diagnosed with autism spectrum disorder (ASD; McClintock et al., 2003). Therefore, given that aggression and disruption may be more prevalent among individuals with more severe impairment, it is plausible that problem behavior may contact reinforcement contingencies and come to

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co-occur with or replace more appropriate means of communication.

Further, individuals with IDD are more likely to be diagnosed with comorbid psychiatric conditions than typically developing peers (Hurley, 2008; Melville et al., 2016; Richards et al., 2001; Sturmey, 2002). Aggressive and disruptive behaviors may be more common in individuals diagnosed with conduct or mood disorders, attention-deficit hyperactivity disorder (ADHD), and epilepsy, among other psychiatric conditions (Connor, Charlier, Preen, & Kaplan, 2010; Gardner & Moffatt, 1990).

Given that most current diagnostic criteria for psychiatric conditions are largely based upon verbal report of symptoms and this verbal behavior repertoire is often underdeveloped in individuals with IDD, the presence of comorbid psychiatric conditions may be underdiagnosed in this population (Melville et al., 2016). Over the last decade, some researchers have suggested that problem behavior (e.g., aggression, disruption) may be a symptom that may point to underlying psychiatric conditions in the IDD population. This has led to the development of sets of diagnostic criteria, such as the *Diagnostic Manual – Intellectual Disabilities* (DM-ID), which may be useful for clinicians in determining the presence of comorbid psychiatric conditions in a patient with IDD (Fletcher, Barnhill, & Cooper, 2016). Further, assessments such as the Problem Behavior Inventory (PBI) and the Psychiatric Assessment Schedule for Adults with Developmental Disabilities (PAS-ADD) may be useful in diagnosing underlying psychiatric conditions and the presence of problem behavior in individuals with IDD (Sturmey, 2002).

Assessment of Aggression and Disruption

The probability of aggression and disruption is higher in individuals dually diagnosed with IDD and certain genetic and psychiatric conditions, as well as with level of impairment. There is likely an interaction between the physiological characteristics of a given individual and their environment that may lead to an increased risk of aggressive or disruptive behavior in an individual. Therefore, it is important for clinicians working with individuals with IDD to be aware of underlying psychiatric or genetic conditions that may increase this probability, such as fragile X syndrome. However, an understanding of these underlying diagnoses alone does not affect the need for a functional assessment to determine the specific contingencies that may maintain the aggressive or disruptive behavior. Stated differently, despite the particular diagnoses, an individual presenting with severe aggression or disruptive behavior is a good candidate for a thorough functional assessment to determine the specific environmental contingencies that may maintain these problem behaviors as a basis for effective behavioral intervention.

Multiple measures exist to assess problem behavior and may be useful to help determine specific forms of problem behavior that should be targeted for further behavioral assessment. These measures can be administered to parents, teachers, or other caregivers to more efficiently identify problem behaviors of concern. The Aberrant Behavior Checklist (ABC) is a measure designed to assess five sets of behavioral symptoms, including irritability, agitation, crying; lethargy and social withdrawal; stereotypic behavior; hyperactivity; and inappropriate speech (Aman, Singh, Stewart, & Field, 1985). Broad instruments, such as the ABC, may not fully capture specific topographies of problem behavior. Therefore, more specific instruments to identify problem behavior have been developed. For example, the Behavior Problems Inventory (BPI-01) is designed to assess multiple forms of problem behavior (i.e., aggression and disruption, stereotypy, and self-injurious behavior) displayed by individuals with IDD (Rojahn, Matson, Lott, Esbensen, & Smalls, 2001). The BPI-01 is a 52-item instrument that includes 2 scales that address both the frequency and severity of the problem behavior.

As with most forms of problem behavior, aggression and disruption are commonly maintained by contingencies in the environment. That is, aggression and disruption are likely to be

occasioned in certain settings or contexts and maintained by either social positive (i.e., attention, procurement of preferred items/activities) or social negative (i.e., escape from aversive activities/tasks) reinforcers. Functional behavior assessment (FBA) is a process wherein various assessments are conducted to determine the function of an individual's behavior in relation to the environment, thus allowing for appropriate development of interventions (Scott, Nelson, & Zabala, 2003). FBA involves identification of predictable relationships between behavior and the environmental events that occur prior to (i.e., antecedents) and following the behavior (i.e., consequences; Scott & Zabala). The FBA consists of indirect assessments, direct assessments, and functional analysis (FA), which are described in greater detail in previous chapters.

In a review of the literature on FA of problem behavior, Beavers, Iwata, and Lerman (2013) evaluated 158 studies during which a FA of problem behavior was conducted. Of those 158 studies, aggression was the second most common problem behavior targeted, occurring in 48% of the sampled studies. Disruption and property destruction were also commonly assessed, occurring in 36.7% and 26.6% of studies, respectively. Interestingly Beavers et al. found that published reports of assessments of property destruction have proportionally increased in the past 10 years as compared to the previous 20 years. Aggression was most often found to be maintained by social reinforcers (social positive reinforcement in 48.9% of cases and social negative reinforcement in 37.8% of cases). Multiple control (i.e., aggression was maintained by more than one variable) was indicated in 11.1% of cases, while automatic reinforcement was only reported in one case (2.2%). For disruption, the behavior was maintained by social positive reinforcement in 11.8% of cases, social negative reinforcement in 11.8% of cases, and automatic reinforcement in 41.2% of cases. Multiple control was indicated in 35.3% of cases. These findings are important for clinicians, as they may have implications for the most efficient functional assessment to be conducted when serving an individual displaying aggressive or disruptive behavior. Although the assessment of aggression and disruption does not necessarily differ from the assessment of other problem behavior, some specific considerations for FAs of aggression and disruption are provided in more detail below.

Special Considerations in Designing Assessment Conditions For patients presenting with aggressive behavior, it is important to include a therapist in relevant test conditions of the FA in order to provide an opportunity for the aggressive behavior to occur. More specifically, it is reasonable to include an ignore condition rather than an alone condition in the FA of aggression, as this would provide an equal opportunity for the aggressive behavior to occur in all conditions of the FA. In practice, some clinicians and researchers forgo a condition that provides information about automatic reinforcement, assuming that aggression cannot be automatically reinforced (i.e., the act itself requires another person). Although it is unlikely that aggression is maintained by automatic reinforcement (see exceptions below), it may still be important to rule in or out all potential variables. As an alternative, clinicians may wish to include only test conditions for socially maintained aggression (i.e., attention, escape, and tangible) and the toy play condition in the initial FA and then include a series of ignore conditions in a subsequent phase or assessment if patterns of responding in the initial FA support that aggression may be maintained by automatic reinforcement. For example, in the event where rates of aggression are high and undifferentiated across all test and the control conditions of the FA and aggression persists in the reinforcement interval, this may be a case where automatic reinforcement (or other, more idiosyncratic functions) should be assessed further.

For disruptive behavior, it may be necessary to determine safe items that may occasion disruptive behavior and then bait each session of the FA with these items. Inclusion of baited items may be necessary because the presence of items to disrupt with is not consistent in all FA conditions. For example, if the primary topography of

disruptive behavior is tearing objects, differentially higher rates of disruption may be observed in conditions such as the escape condition, where task-related paper materials are present, or the attention condition, if books are included as moderately preferred stimuli, whereas differentially lower rates of disruptive behavior may occur in the alone/ignore, tangible, or toy play conditions, if no objects that can be ripped are present in those conditions. This could be problematic if disruptive behavior was maintained by automatic reinforcement, as the behavior might not occur in all conditions due to unequal access to items.

To alleviate this potential issue, preference for disruptive items could be based on anecdotal observations or descriptive assessment of the type of disruptive behavior the individual usually engages in (e.g., tearing paper, breaking toys, banging on tables, throwing items) or empirically determined through more formal preference assessments for disruptive items. For example, a free operant assessment could be conducted in which the individual is placed in a room alone with the items for a short duration (e.g., 5 min), and data are collected on disruptive behavior that occurs with each of the items (e.g., Ringdahl, Vollmer, Marcus, & Roane, 1997). Items associated with higher levels of disruption during this assessment could then be incorporated across all conditions of the FA sessions to ensure equal opportunities to engage in the specific topography of disruptive behavior are arranged across all conditions. In this way, the items selected for assessment will reflect the topography of the disruptive behavior that is problematic. However safety is of the utmost priority in assessment; thus clinicians should weigh the potential for items included in assessment to cause harm to the individual or those conducting the assessment. For example, if the individual engages in pica, caution should be taken to ensure any items selected for baited disruption sessions do not pose a pica hazard. Similarly, care should be taken to avoid heavy items that might be thrown and injure others, if throwing objects is the specific topography targeted for assessment.

Assessment in School Settings Some of the research on the assessment of aggression and disruption has taken place in the context of classrooms (e.g., Dolezal & Kurtz, 2010; Northup et al., 1995, 1997) or after-school programs (e.g., Noel & Getch, 2016). One reason for this focus on academic settings is that aggressive or disruptive behavior that occurs at school can not only have a negative impact on an individual's ability to learn but also interrupt ongoing instruction for other students.

Northup et al. (1995, 1997) conducted two studies that assessed disruptive behavior in classroom settings for individuals with dual diagnoses, specifically, ADHD and ID. Importantly in both studies, they found that disruptive behavior was more likely when peer attention, rather than teacher attention, followed this behavior for most individuals. The authors also suggest that some behavioral treatments of disruption (e.g., time-out) may be as effective as stimulants in treating this behavior.

Dolezal and Kurtz (2010) conducted a structured descriptive analysis (SDA) to inform the development of the FA conditions in a classroom setting. School staff conducted both demand and toy play sessions during the context of the normal school schedule. Results of the SDA concluded that aggression and disruption only occurred during the demand condition; however, attention reliably followed problem behavior during the demand context. During the FA, single-antecedent conditions (i.e., escape and attention) were compared to a combined-antecedent condition (i.e., escape and divided attention) and a control condition (i.e., toy play). Differentially higher levels of problem behavior were observed in the combined-antecedent test condition of the FA, indicating aggression and disruption were most reliably occasioned in the presence of both antecedent conditions. As both of these variables frequently occur together in a natural classroom setting, their identification is critical to influence the development of effective treatment options. For example, an effective intervention may incorporate choice between the two functional reinforcers (i.e., escape or attention).

Attention Type For some individuals, specific social contexts may occasion aggressive or disruptive behavior. In the attention condition of the traditional FA described by Iwata, Dorsey, Slifer, Bauman, and Richman (1982/1994), the attention condition consists of providing brief reprimands contingent upon the targeted problem behavior. For some individuals, this form of attention may not maintain problem behavior, which may lead to inconclusive FA results. If results from indirect assessments and observations suggest the presence of an attention function, it may be necessary to empirically determine the specific type of attention that maintains aggression. Several studies in the literature have highlighted different types of attention that may maintain problem behavior, such as the content of contingent statements (Fisher, Ninness, Piazza, & Owen-DeSchryver, 1996), physical versus vocal attention (Kodak, Northup, & Kelley, 2007), or peer-delivered attention (Northup et al., 1995). Roscoe, Kindle, and Pence (2010) conducted both a paired stimulus preference assessment of conversational topics and an attention analysis to further investigate the functional relationship between preferred conversational topics and aggression. During this attention analysis, a highly preferred (HP) conversational topic, a low-preferred (LP) conversational topic, and a control condition consisting of noncontingent HP conversation were evaluated. During the HP and LP conversation conditions, aggression resulted in 30 s access to either HP or LP conversational topics. Latency to aggression was differentially lower in the HP conversation condition, suggesting that contingent access to preferred topics maintained aggression. Following the attention analysis, an extended FA was conducted, during which the HP conversation condition was included along with attention, demand, tangible, and the control condition. With the inclusion of the HP conversation condition, a clear attention function could be identified for aggression.

Divided Attention In some individuals, aggression or disruption is maintained by attention during situations in which an adult's attention is divided (e.g., Fisher, Kuhn, & Thompson, 1998; Hagopian, Contrucci-Kuhn, Long, & Rush, 2005: Strohemeier, Pace, & Luiselli, 2014). The inclusion of the divided attention condition may be beneficial because it is possible that observation of ongoing conversation might serve as a discriminative stimulus for attention availability. That is, some individuals may be more likely to engage in aggression or disruption during the attention condition if the therapist's attention is already being delivered to a confederate. Mace, Paige, Ivancic, and O'Brien (1986) described a divided attention condition in a FA in which the therapist engaged in conversation with another adult. Contingent upon aggression, the therapist provided attention to the participant as in the standard attention condition. Fahmie, Iwata, Harper, and Querim (2013) conducted a more systematic assessment of the divided attention condition, specifically to more directly compare rates of problem behavior (including aggression) during the standard and divided attention conditions of the FA. For the majority of participants, rates of problem behavior were similar between the standard and divided attention conditions; however, for a small number of participants, an attention function emerged more quickly during the divided attention condition. Strohemeier et al. (2014) conducted a brief FA of divided attention, which included a preference assessment of staff. The staff member selected most often served as the therapist in all FA sessions, and a confederate was included in the divided attention condition. Contingent upon aggressive behavior toward either the therapist or the confederate, the confederate moved away, and the preferred staff member delivered 30 s of attention. This arrangement produced a clearly differentiated divided attention condition. Given data from these studies, it is plausible that inclusion of both a preference assessment of staff and the divided attention condition in a FA of aggression may be an efficient means of identifying a divided attention function.

Results such as these highlight that it may be necessary to conduct subsequent assessments to better isolate the specific qualities of attention

that may maintain aggression, especially if there is evidence to suggest an attention function is present. That is, although the "standard" attention condition described by Iwata et al. (1982/1994) may be sufficient for the majority of individuals, a small subset of individuals with aggressive behavior may warrant additional assessment to determine whether more idiosyncratic variables maintain the behavior.

Escape from Social Interaction Aggression and disruption may also be maintained by more idiosyncratic variables, such as escaping social interaction, which is a special case of social negative reinforcement wherein an individual engages in aggressive behavior to terminate or delay the onset of aversive social interactions (Carr, 1994; Harper, Iwata, & Camp, 2013; Taylor & Carr, 1992; Taylor & Carr, 1992). Individuals with aggression and disruption maintained by escape from social interaction may engage in differentially higher rates of problem behavior in FA conditions during which there are higher levels of social interaction (i.e., demand and toy play) relative to conditions during which the overall level of social interaction is lower (i.e., attention or ignore; Iwata et al., 1994). Taylor and Carr (1992) assessed whether social interaction could serve as an establishing operation for problem behavior and subsequently demonstrated that the removal of social interaction might function as a reinforcer for some individuals (1992b). A small number of studies in the literature have evaluated a functional relationship between problem behavior and the termination of social interaction (Frea & Hughes, 1997; Hagopian, Wilson, & Wilder, 2001; Harper et al., 2013; Vollmer et al., 1998). In these studies, test conditions during which social interaction was removed contingent upon problem behavior were evaluated. For example, Harper et al. (2013) conducted a modified FA during which a therapist delivered social statements with physical contact every 5 s to the participant and terminated social interaction for 30 s contingent upon aggression. An ignore-plus-toys condition served as the control.

Termination of Interruptions from Preferred Activity Some individuals may display aggression or disruption behavior when activity engagement is interrupted. Hagopian, Bruzek, Bowman, and Jennett (2007) evaluated problem behavior maintained by terminating interruptions. Rates of aggressive behavior during the initial FA (using standard conditions) were low and not representative of rates of aggression that had been observed outside of sessions. In a subsequent interruption analysis, pre-session access to a preferred activity was provided, and then the activity was interrupted with a "do" or "don't" request every 30 s. Problem behavior resulted in contingent access to the activity and termination of the interruption. Although it is difficult to determine whether aggression was maintained by access to the preferred activity (positive reinforcement) or termination of the interruption (negative reinforcement) or a combination of the two, it is possible that aggressive behavior may be occasioned more specifically in a tangible context by interrupting the ongoing activity with a less preferred activity versus simple removal of a toy.

Access to Ritualistic Behavior Some individuals, especially those diagnosed with ASD, may engage in aggression or disruption specifically to gain access to ritualistic behavior. Hausman, Kahng, Farrell, and Mongeon (2008) found that the problem behaviors (including aggression and disruptive behavior) of an individual diagnosed with ASD and ID were maintained by gaining access to a ritualistic behavior (i.e., repeatedly opening and closing doors). The authors were able to treat these behaviors by teaching the individual to appropriately request the ritual, rather than engaging in problem behavior. In a very similar study, Leon, Lazarchick, Rooker, and Deleon (2013) demonstrated that the problem behavior (including aggression and disruption) of one individual diagnosed with autism was maintained by access to straightening and ordering board game pieces. Similarly, the authors were able to treat this behavior by teaching the individual to appropriately request access to the ritual.

Automatic Reinforcement FA results reported in the literature suggest that it is unlikely that aggression is maintained by automatic reinforcement. To date, only three cases of automatically maintained aggressive behavior have been reported in the literature (Ringdahl, Call, Mews, Boelter, & Christensen, 2008; Saini, Greer, & Fisher, 2015; Thompson, Fisher, Piazza, & Kuhn, 1998). In all three cases, the initial FA was inconclusive; however, it should be noted that an ignore condition was not included in any of these initial functional analyses because aggression was one of the primary behaviors targeted. Subsequent manipulations, such as comparing relative rates of aggression during contingent and noncontingent attention conditions (Thompson et al., 1998), extended FAs (i.e., extended ignore or a sequential test-control analysis; Ringdahl et al., 2008), and a single topography FA of aggression and a sensory extinction evaluation (Saini et al., 2015), were conducted to confirm that aggression was maintained by automatic reinforcement. Thus, it may be most efficient to consider evaluating social functions for aggression in the initial FA, followed by subsequent analyses to test for an automatic function if aggression persists across all conditions of the FA, including the control condition (toy play).

Examples of disruption maintained by automatic reinforcement in the published literature are also somewhat rarer; however, it is important to note there is some evidence that automatically maintained disruption, specifically property destruction, may be maintained by the products of this destruction rather than reinforcers embedded in the act of destruction itself. For example, an individual may break a toy into pieces and then throw the pieces of the toy because it produces favorable sensory reinforcement (e.g., visual or auditory input). Fisher, Lindauer, Alterson, and Thompson et al. (1998) reported on two cases where individuals engaged in property destruction to gain access to the products of this destruction, which they subsequently used to engage in stereotypy. In this way, automatically maintained disruption can be distinct from behaviors such as automatically reinforced self-injury, where the response itself is assumed to produce the reinforcer (e.g., sensory stimulation).

Low-Rate, High-Intensity Behavior Assessment of problem behavior that is cyclical in nature is a particular challenge, in that it can be difficult to establish consistent rates of responding. Thus, it is often difficult to determine the function of problem behavior that is more cyclical through the use of brief session durations. For some individuals, problem behavior may present as low-frequency, high-intensity bouts, or severe outbursts, which may be observed in individuals diagnosed with IDD and a cyclical mood disorder (Lewis, Silva, & Silva, 1995). In these cases, modifications to FA methodology may be necessary to assess and treat the target behavior. Kahng, Abt, and Schonbachler (2001) extended the duration of FA sessions such that a single condition of the FA was conducted during the hours of 9:00 am–4:00 pm on an inpatient hospital unit. The order of attention, demand, and toy play sessions were randomly selected, and contingencies were identical to those described by Iwata et al. (1982/1994). During the demand condition, 45 min of demands were presented every hour, followed by a 15 min break with minimal interaction. A clear attention function was identified through the use of this extended FA. The utility of the extended FA was replicated across six individuals and extended to include a tangible condition by Davis, Kahng, Schmidt, Bowman, and Boelter (2012); however, the authors noted that the extended FA did not identify a function for four additional individuals exposed to these contingencies.

Functional Behavioral Phenotype

Taking the perspective that aggressive behavior is likely the outcome of both environmental factors (that occasion and reinforce the behavior) and dysfunction stemming from genetic or medical disorders, some research has examined problem behavior through detailing the functional behavioral phenotype

of a particular syndrome. Put more simply, this research has looked for commonalities in behavior within genetic syndromes and then taken the additional step of examining commonalities in the reasons why these behaviors occur. Although this research is extremely limited, work on dual diagnosis of fragile X and ASD does suggest there may be commonalities in the reason aggression occurs (i.e., the function of aggression) in this population of individuals.

Approximately 20% of individuals diagnosed with fragile X are also diagnosed with ASD (Richards, Jones, Groves, Moss, & Oliver, 2015). Further, approximately 50% of individuals with fragile X engage in aggression. Both Kurtz, Chin, Robinson, O'Connor, and Hagopian (2015) and Machalicek et al. (2014) conducted FAs of problem behavior for groups of individuals with fragile X. Across these two studies, 14 individuals were dually diagnosed with fragile X and ASD. Based on the results of the FA, 64.3% individuals engaged in aggression to access tangible items or requests, and 57.1% individuals engaged in aggression to escape academic work or the presence of other people. Further, access to attention or self-stimulation (i.e., automatic reinforcement) was an unlikely function for aggression in this population. The results of these studies suggest the potential power of functional behavioral phenotyping for a dual diagnosis population, and they may indicate additional risk factors or suggest potential ways to prevent the emergence of aggressive behavior. For example, early intervention with individuals with fragile X might more extensively focus on preemptively training skills such tolerance of delays to highly preferred activities and communication to avoid aversive situations. Following this example, additional functional behavior phenotype studies are needed for other conditions, particularly for individuals where aggression appears quite common (e.g., Angelman's syndrome, cri du chat, and Smith-Magenis; Arron, Oliver, Moss, & Burbidge, 2011).

Conclusion

Individuals with IDD and comorbid psychiatric or genetic conditions are more likely to display aggression and disruptive behavior than typically developing peers, which can have a negative impact on various areas of social functioning. However, the behaviors of individuals with dual diagnoses are heterogeneous, and more research is needed on better understanding how specific groups come to be more or less likely to engage in particular forms of behavior for specific reasons. For example, Cooper et al. (2009) found that individuals with ADHD were more likely to engage in aggression. Further, Neef, Bicard, and Endo (2001) found that individuals with ADHD were more sensitive to immediacy of reinforcement. Speculatively, one can imagine the scenario in which individuals with a dual diagnosis that includes ADHD might engage in aggression or disruption because engaging in these behaviors has a history of shortening the delay to accessing the reinforcer. Similarly, individuals with ASD engage in repetitive, invariant patterns of responding that are less affected by changes in the environment relative to matched controls (Rodriguez & Thompson, 2015). Thus, one might expect these individuals to engage in aggression or disruption more often to gain access to stereotypies or rituals. However, these types of questions have not been examined across a large number of individuals with similar dual diagnoses. Research is needed on common functions for aggression and disruption in genetic and psychiatric conditions, similar to research conducted by Kurtz et al. (2015) and Machalicek et al. (2014) conducted on fragile X. These types of analyses are needed to better understand these populations and develop unique treatments for particular populations.

References

Allen, D. (2000). Recent research on physical aggression in persons with intellectual disability: An overview. *Journal of Intellectual & Developmental Disability, 25*, 41–57. https://doi.org/10.1080/132697800112776

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*, 485–491.

Arron, K., Oliver, C., Moss, J., & Burbidge, C. (2011). The 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

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

Borthwick-Duffy, S. A., Lane, K. L., & Widaman, K. F. (1997). Measuring problem behaviors in children with mental retardation: Dimensions and predictors. *Research in Developmental Disabilities, 18*, 415–433. https://doi.org/10.1016/s0891-4222(97)00020-6

Carr, E. G. (1994). Emerging themes in the functional analysis of problem behavior. *Journal of Applied Behavior Analysis, 46*, 1–2. https://doi.org/10.1901/jaba.1994.27-393

Connor, D. F., Charlier, K. G., Preen, E. C., & Kaplan, R. F. (2010). Impulsive aggression in attention-deficit/hyperactivity disorder: Symptom severity, comorbidity, and attention-deficit/hyperactivity disorder subtype. *Journal of Child and Adolescent Child Psychopharmacology, 20*, 119–126. https://doi.org/10.1089/cap.2009.0078

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*, 217–232. https://doi.org/10.1111/j.1365-2788.2008.01127.x

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*, 652–661. https://doi.org/10.1111/j.1365-2788.2006.00815.x

Davis, B. J., Kahng, S., Schmidt, J., Bowman, L. G., & Boelter, E. W. (2012). Alterations to functional analysis methodology to clarify the functions of low-rate, high-intensity problem behavior. *Behavior Analysis in Practice, 5*, 27–39. https://doi.org/10.1007/bf03391815

Dolezal, D. N., & Kurtz, P. F. (2010). Evaluation of combined-antecedent variables on functional analysis results and treatment of problem behavior in a school setting. *Journal of Applied Behavior Analysis, 43*, 309–314. https://doi.org/10.1901/jaba.43-309

Dworschak, W., Ratz, C., & Wagner, M. (2016). Prevalence and putative risk markers of challenging behavior in students with intellectual disabilities. *Research in Developmental Disabilities, 58*, 94–103. https://doi.org/10.1016/j.ridd.2016.08.006

Emerson, E., Robertson, J., Gregory, N., Hatton, C., Kessissoglou, S., Hallam, A., & Hillery, J. (2000). Treatment and management of challenging behaviours in residential settings. Journal of Applied Research in Intellectual Disabilities, 13, 197–215.

Fahmie, T. A., Iwata, B. A., Harper, J. M., & Querim, A. C. (2013). Evaluation of the divided attention condition during functional analyses. *Journal of Applied Behavior Analysis, 46*, 71–78. https://doi.org/10.1002/jaba.20

Fisher, W. W., Kuhn, D., & Thompson, R. (1998). Establishing discriminative control of responding using functional and alternative reinforcers during functional communication training. *Journal of Applied Behavior Analysis, 31*, 543–560. https://doi.org/10.1901/jaba.1998.31-543

Fisher, W. W., Lindauer, S. E., Alterson, C. J., & Thompson, R. H. (1998). Assessment and treatment of destructive behavior maintained by stereotypic object manipulation. *Journal of Applied Behavior Analysis, 31*, 513–527. https://doi.org/10.1901/jaba.1998.31-513

Fisher, W. W., Ninness, H. A. C., Piazza, C. C., & Owen-DeSchryver, J. S. (1996). On the reinforcing effects of the content of verbal attention. *Journal of Applied Behavior Analysis, 29*, 235–238. https://doi.org/10.1901/jaba.1996.29-235

Fletcher, R. J., Barnhill, J., & Cooper, S.-A. (2016). *Diagnostic manual – Intellectual disability (DMID-2): A textbook of diagnosis of mental disorders in persons with intellectual disability*. Kingston, NY: NADD Press.

Frea, W. D., & Hughes, C. (1997). Functional analysis and treatment of social-communicative behavior of adolescents with developmental disabilities. *Journal of Applied Behavior Analysis, 30*, 701–704.

Gardner, W. I., & Moffatt, C. W. (1990). Aggressive behavior: Definition, assessment, and treatment. *International Review of Psychiatry, 2*, 91–100.

Hagopian, L. P., Bruzek, J. L., Bowman, L. G., & Jennett, H. J. (2007). Assessment and treatment of problem behavior maintained by interruption of free-operant behavior. *Journal of Applied Behavior Analysis, 40*, 89–103. https://doi.org/10.1901/jaba.2007.63-05

Hagopian, L. P., Contrucci-Kuhn, S. A., Long, E. S., & Rush, K. A. (2005). Schedule thinning following functional communication training: Using competing stimuli to enhance tolerance to decrements in reinforcer density. *Journal of Applied Behavior Analysis, 38*, 177–193. https://doi.org/10.1901/jaba.2005.43-04

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*, 229–232. https://doi.org/10.1901/jaba.2001.34-229

Harper, J. M., Iwata, B. A., & Camp, E. M. (2013). Assessment and treatment of social avoidance. *Journal of Applied Behavior Analysis, 46*, 147–160. https://doi.org/10.1002/jaba.18

Hausman, N., Kahng, S. W., Farrell, E., & Mongeon, C. (2008). Idiosyncratic functions: Severe problem behavior maintained by access to ritualistic behaviors. *Education and Treatment of Children, 32*, 77–87. https://doi.org/10.1353/etc.0.0051

Hurley, A. D. (2008). Depression in adults with intellectual disability: Symptoms and challenging behaviour. *Journal of Intellectual Disability Research, 52*, 905–916. https://doi.org/10.1111/j.1365-2788.2008.01113.x

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)

Kahng, S., Abt, K. A., & Schonbachler, H. E. (2001). Assessment and treatment of low-rate high-intensity problem behavior. *Journal of Applied Behavior Analysis, 34*, 225–228. https://doi.org/10.1901/jaba.2001.34-225

Kodak, T., Northup, J., & Kelley, M. E. (2007). An evaluation of the types of attention that maintain problem behavior. *Journal of Applied Behavior Analysis, 46*, 71–78. https://doi.org/10.1901/jaba.2007.43-06

Kurtz, P. F., Chin, M. D., Robinson, A. N., O'Connor, J. T., & Hagopian, L. P. (2015). Functional analysis and treatment of problem behavior exhibited by children with fragile X syndrome. *Research in Developmental Disabilities, 43-44*, 150–166. https://doi.org/10.1016/j.ridd.2015.06.010

Leon, Y., Lazarchick, W. N., Rooker, G. W., & DeLeon, I. G. (2013). Assessment of problem behavior evoked by disruption of ritualistic toy arrangement in a child with autism. *Journal of Applied Behavior Analysis, 46*, 507–511. https://doi.org/10.1002/jaba.41

Lewis, M. H., Silva, J. R., & Silva, S. G. (1995). Cyclicity of aggression and self-injurious behavior in individuals with mental retardation. *American Journal on Mental Retardation, 99*, 436–444.

Mace, F., Paige, T., Ivancic, M., & O'Brien, S. (1986). Analysis of environmental determinants of aggression and disruption in mentally retarded children. *Applied Research in Mental Retardation, 7*, 203–221. https://doi.org/10.1016/0270-3092(86)90006-8

Machalicek, W., McDuffie, A., Oakes, A., Ma, M., Thurman, A. J., Rispoli, M. J., & Abbeduto, L. (2014). Examining the operant function of challenging behavior in young males with fragile X syndrome: A summary of 12 cases. *Research in Developmental Disabilities, 35*, 1694–1704. https://doi.org/10.1016/j.ridd.2014.03.014

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. https://doi.org/10.1046/j.1365-2788.2003.00517.x

Melville, C. A., Johnson, P. C. D., 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. https://doi.org/10.1016/j.ridd.2016.03.007

Neef, N. A., Bicard, D. F., & Endo, S. (2001). Assessment of impulsivity and the development of self-control in students with attention deficit hyperactivity disorder. *Journal of Applied Behavior Analysis, 34*, 397–408. https://doi.org/10.1901/jaba.2001.34-397

Noel, C. R., & Getch, Y. Q. (2016). Non-contingent reinforcement in after-school settings to decrease classroom disruptive behavior for students with autism spectrum disorder. *Behavior Analysis in Practice, 9*, 261–265. https://doi.org/10.1007/s40617-0616-0117-0

Northup, J., Broussard, C., Jones, K., George, T., Vollmer, T. R., & Herring, M. (1995). The differential effects of teacher and peer attention on the disruptive classroom behavior of three children with a diagnosis of attention deficit hyperactivity disorder. *Journal of Applied Behavior Analysis, 28*, 227–228. https://doi.org/10.1901/jaba.1995.28-227

Northup, J., Jones, K., Broussard, C., DiGiovanni, G., Herring, M., Fusilier, I., & Hanchey, A. (1997). A preliminary analysis of interactive effects between common classroom contingencies and methylphenidate. *Journal of Applied Behavior Analysis, 30*(1), 121–125.

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*, 1269–1275. https://doi.org/10.1016/j.ridd.2010.07.017

Rodriguez, N. M., & Thompson, R. H. (2015). Behavioral variability and autism spectrum disorder. Journal of Applied Behavior Analysis, 48(1), 167–187.

Richards, C., Jones, C., Groves, L., Moss, J., & Oliver, C. (2015). Prevalence of autism spectrum disorder phenomenology in genetic disorders: A systematic review and meta-analysis. *The Lancet Psychiatry, 2*, 909–916. https://doi.org/10.1016/s2215-0366(15)00376-4

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*, 523–527. https://doi.org/10.1192//bjp.179.6.523

Ringdahl, J. E., Call, N. A., Mews, J. B., Boelter, E. W., & Christensen, T. J. (2008). Assessment and treatment of aggressive behavior without a clear social function. *Research in Developmental Disabilities, 29*, 351–362. https://doi.org/10.1016/j.ridd.2007.06.003

Ringdahl, J. E., Vollmer, T. R., Marcus, B. A., & Roane, H. S. (1997). An analogue evaluation of environmental enrichment: The role of stimulus preference. *Journal of Applied Behavior Analysis, 30*, 203–216. https://doi.org/10.1901/jaba.1997.30-203

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*, 577–588.

Roscoe, E. M., Kindle, A. E., & Pence, S. T. (2010). Functional analysis and treatment of aggression maintained by preferred conversational topics. *Journal of Applied Behavior Analysis, 43*, 723–727. https://doi.org/10.1901/jaba.2010.43-723

Saini, V., Greer, B. D., & Fisher, W. W. (2015). Clarifying inconclusive functional analysis results: Assessment and treatment of automatically reinforced aggression. *Journal of Applied Behavior Analysis, 48*, 315–330. https://doi.org/10.1002/jaba.203

Scott, T. M., Nelson, C. M., & Zabala, J. (2003). Functional behavior assessment training in public schools: Facilitating systemic change. *Journal of Positive Behavior Interventions, 5*(4), 216–224.

Strohemeier, C., Pace, G. M., & Luiselli, J. K. (2014). Brief (test-control) functional analysis and treatment evaluation of aggressive behavior evoked by divided attention. *Behavioral Interventions, 29*, 331–338. https://doi.org/10.1002/bin.1394

Sturmey, P. (2002). Mental retardation and concurrent psychiatric disorder: Assessment and treatment. *Current Opinion in Psychiatry, 15*, 489–495. https://doi.org/10.1097/00001504-200209000-00005

Smith, T. B., Oliver, M. N., & Innocenti, M. S. (2001). Parenting stress in families of children with disabilities. American journal of orthopsychiatry, 71, 257.

Taylor, J. C., & Carr, E. G. (1992). Severe problem behaviors related to social interaction I: Attention seeking and social avoidance. *Behavior Modification, 16*, 305–335. https://doi.org/10.1177/01454455920163002

Thompson, R. H., Fisher, W. W., Piazza, C. C., & Kuhn, D. E. (1998). The evaluation and treatment of aggression maintained by attention and automatic reinforcement. *Journal of Applied Behavior Analysis, 31*, 103–116. https://doi.org/10.1901/jaba.1998.31-103

Vollmer, T. R., Progar, P. R., Lalli, J. S., Van Camp, C. M., Sierp, B. J., Wright, C. S., … Eisenschink, K. J. (1998). Fixed-time schedules attenuate extinction-induced phenomena in the treatment of severe aberrant behavior. *Journal of Applied Behavior Analysis, 31*, 529–542. https://doi.org/10.1901/jaba.1998.31-529

Wehman, P., & McLaughlin, P. J. (1979). Teachers' perceptions of behavior problems with severely and profoundly handicapped students. *Mental Retardation, 17*, 20–21.

Self-Injurious Behavior, Rituals, and Stereotypies

21

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Introduction

Individuals diagnosed with intellectual disability (ID) are at an increased risk for development of behavioral challenges relative to the general population. In fact, 10–20% of the ID population displays challenging forms of behavior (Blair, Lloyd, Craig & Kennedy, 2014; McClintock, Hall, & Oliver, 2003). The prevalence of challenging behavior in individuals with ID often increases throughout childhood into the teenage years, before plateauing during adulthood (Davies & Oliver, 2013). Furthermore, the consequences of challenging behavior are often detrimental to the lives of individuals with ID, resulting in social isolation, impaired adaptive skills, placement in residential facilities, denial of healthcare services, physical abuse, injury, and even death (Emerson, 2000; Hyman, Fisher, Mercugliano, & Cataldo, 1990; Robertson et al., 2004). Thus, the need for prevention of, and effective treatment for, challenging behavior is essential for improving the lives of individuals with ID.

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Self-injurious behavior (SIB), rituals, and stereotypies are three common forms of challenging behavior presented by individuals with ID. Historically, treatment for such challenging behavior in this population has involved pharmacological interventions or application of arbitrarily selected reinforcers and/or punishers, based primarily on clinical intuition rather than consideration of patient or environmental variables that directly affected the occurrence of the behavior. Such treatments produced outcomes that were intermittently effective, at best (Loyd & Kennedy, 2014). However, the advent of methodologies aimed at evaluating the factors that affect the occurrence of challenging behavior (e.g., functional assessment) paved the way for development of more consistently effective treatments (Edward, Carr, 1977; Derby et al., 1992; Iwata, Dorsey, Slifer, Bauman, & Richman, 1982/1994; McClintock et al., 2003).

One of the most critical components of treatment development is implementation of appropriate behavioral assessment strategies that help caregivers and practitioners best understand the symptoms (e.g., behavioral topographies) desired for change and the relevant variables that most likely occasion the targeted challenging behavior. However, it is also crucial to have an understanding of the risk factors associated with SIB, rituals, and stereotypies to help aid in the development of preventative techniques. Thus, in the present chapter, we will begin by discussing definitions and

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risk factors associated with SIB, rituals, and stereotypies. We will describe the unique characteristics of each of these three topographies, the similarities that often exist between them, and how they may be closely intertwined. We will conclude by reviewing common assessment approaches that are often used to facilitate treatment development and/or planning. Thus, the overview provided in this chapter is aimed to equip the reader with a better understanding of the specified behavioral topographies and how one may best go about informing treatment development.

Definitions and Risk Factors for SIB, Rituals, and Stereotypies

Self-injury, rituals, and stereotypies have been found to be positively correlated (i.e., increases in SIB tend to be associated with increases in rituals and stereotypies) across a number of studies within the literature (e.g., Bodfish, Crawford, Powell, & Parker, 1995; Bodfish & Lewis, 2002; Bodfish, Symons, Parker, & Lewis, 2000; Collacott, Cooper, Branford, & McGrother, 1998), suggesting that these forms of challenging behavior tend to co-occur within the ID population. Beyond the covariation among these behavioral patterns, there has also been considerable overlap in the way in which they have been conceptualized and defined. For example, SIB has been defined as behavior directed toward oneself that produces physical damage (Tate & Baroff, 1966). Although SIB can take on many different forms (e.g., self-biting (Iwata et al., 1982/1994), face slapping (Lang et al., 2010), skin picking (Cooper et al., 2009)), it can occur in a repetitive manner – a defining feature of rituals and stereotypies. More specifically, rituals have been defined as *repetitive* adherence to self-imposed rules (Zohar & Felz, 2001) and stereotypies as *repetitive*, topographically invariant, and rhythmical behavioral patterns (Powell, Newman, Pendergast, & Lewis, 1999; Wunderlich & Vollmer, 2015). In either case, rituals and stereotypies are classified as repetitive behaviors and may manifest as SIB within the ID population when these behavioral patterns have the potential to cause physical damage to the individual performing the behavior.

Given the similarities across these behavioral patterns (e.g., they may occur repetitively), differentiating between them within the literature can be challenging. Bodfish and Lewis (2002) noted two issues that may contribute to this difficulty. First, the use of poor operational definitions may significantly impede efforts to differentiate between behavior that is self-injurious, ritualistic, or stereotypic. For example, a behavioral therapist may define self-injury for a given client as "contact between the client's head and an open palm or closed fist." This definition is sufficiently broad to describe the forceful strike that leaves lasting soft tissue damage, which one may readily classify as self-injury, and light, repetitive taps to the cheek which one may classify instead as stereotypy. The scientific discipline of the individual who is describing the behavior may also influence the manner in which that behavior is defined and the etiology that is ascribed to the behavior. Trichotillomania (i.e., repeatedly pulling out one's own hair), for example, may be conceptualized as SIB by a practitioner focusing on behavior-environment interactions, and this individual may focus more heavily on the possible mediation of the behavior by social consequences (e.g., delivery of caregiver attention for bouts of hair pulling). Conversely, a practitioner focusing on more cognitive explanations of behavior may define trichotillomania as a ritual and focus on the potential underlying neurobiological mechanisms of the behavior.

The challenges in distinguishing between SIB, rituals, and stereotypies described above may have serious negative collateral effects. For example, if researchers differentially characterize these behavioral patterns based on their scientific perspectives, consuming the scoping literature on these behavioral patterns to identify effective assessment and treatment strategies may be difficult. For example, if one were to define chronic nail biting as a minor form of self-injury (see, e.g., Ballinger, 1971), she or he may inadvertently overlook effective nail-biting treatments found in the literatures on rituals and stereotypies

(e.g., Barmann, 1979; Ladouceur, 1979). Further, within an individual who displays SIB, rituals, and stereotypies, these behavioral patterns may superficially *look* similar but may be *functionally* distinct. That is, the behaviors may fit under the same insufficiently precise operational definition but may occur for very different reasons. If clinicians were unable to distinguish between them based on a dubious definition, assessment and treatment of any individual behavior may be difficult. Thus, in the following section, we will provide guidance for differentiating between SIB, rituals, and stereotypies to aid in efficient assessment strategies and ultimately the development of effective intervention.

Self-Injurious Behavior

Self-injurious behavior has been defined a number of ways within the literature, and the definition has evolved over time. As previously stated, Tate and Baroff (1966) defined SIB as repetitive acts directed toward oneself that produce physical damage. This definition was then further refined by Winchel and Stanley (1991) and Klonsky and Meuhlenkamp (2007) to exclude any form of self-injury associated with suicide, sexual arousal, or sociocultural practices. That is, the *intent* of SIB was included in these definitions (i.e., excluding SIB that occurs with a specific goal). This method for defining SIB may be problematic because, in the absence of direct evidence about the goal of a behavior, the goal so ascribed may lead to erroneous conclusions regarding the causes of SIB (e.g., one may assert that SIB is caused by depression, when more careful analysis may reveal that SIB is communicative and maintained by social consequences). For this reason, Rojahn, Whittaker, Hoch, and Gonzáles (2007) argued that definitions of SIB should not include inferences regarding the intent of the behavior. However, for the purposes of this chapter, we will adopt an integrated definition of SIB put forth by Didden et al. (2012) and define it as repetitive acts that have the potential to cause harm to the individual and exclude any acts associated with suicide, sexual arousal, or sociocultural practices. This definition most closely aligns with the forms of SIB displayed by individuals with ID. Moreover, it acknowledges the distinctiveness of other forms of self-injury associated with suicide, sexual arousal, and sociocultural practices, as these specific forms of self-injury are etiologically separable and may require approaches to assessment and treatment that are outside the purview of this chapter (Didden et al., 2012; Weiss, 2003).

Given this broad definition of SIB, there are many topographies of behavior that may fit into this classification (e.g., hair pulling, eye poking, self-scratching, rumination, face rubbing, etc.). However, the most common topographies of SIB within the ID population are head banging, head hitting, and self-biting (Kahng, Iwata, & Lewin, 2002). These specific forms of SIB are common among individuals in the ID population and are of social significance, as they may put the individual at an increased risk for permanent physical injury (e.g., brain injury; Anderson & Ernst, 1994), loss of sensory function (e.g., loss of vision; Barrera, Violo, & Graver, 2007), or even death (Nissen & Haveman, 1997).

Beyond the potential for SIB to cause physical damage to the individual, it may also impact other aspects of their daily lives. For instance, Baghdadli, Pascal, Grisi, and Aussilloux (2003) assessed, among other variables, the levels of adaptive functioning (e.g., communication, daily living skills, socialization) displayed by a sample of 222 young children (aged 2–7 years old) with autism spectrum disorder (ASD; 96% with comorbid ID), 53% of whom engaged in SIB. These authors found that development of adaptive skills was significantly delayed in those children who engaged in SIB relative to those who did not. Engagement in SIB in conjunction with low levels of adaptive functioning can impede individuals' social and educational development, potentially leading to restricted living situations and/or school placements (Khang, et al., 2002). Furthermore, SIB has been linked to increased stress and depression in caregivers (Baxter, Cummins, & Yiolitis, 2000), illustrating the widespread negative collateral effects SIB may have beyond detriments to the physical state

of the individual who engages in the behavior. For these reasons, SIB has been described as one of the most destructive forms of problem behavior exhibited by individuals with ID (Minshawi, Hurwitz, Morriss, & McDougle, 2015a; b).

Regarding its prevalence, it has been estimated that approximately 10% of the ID population displays some form of SIB (Emerson et al., 2001; Holden & Gitlesen, 2006; Lowe et al., 2007; Murphy, Oliver, & Corbett, 1993; Oliver, Murphy, & Corbett, 1987). However, when certain risk factors are also present, these prevalence estimates increase dramatically within the ID population. For instance, prevalence estimates of SIB in the ASD population have ranged from 33% to 71% (Baghdadli et al., 2003; Dominick, Davis, Lainhart, Tager-Flusberg, & Folstein, 2007; Murphy, Healy, & Leader, 2009; McTiernan, Leader, Healy, & Mannion, 2011; Richards, Davies, & Oliver, 2017; Richards, Oliver, Nelson, & Moss, 2012; Soke et al., 2017). On the surface, these elevated prevalence rates, relative to prevalence in the general ID population, suggest that a comorbid diagnosis of ASD is a risk factor for SIB. This relation, however, becomes much more complicated as the level of intellectual and adaptive functioning is accounted for in the ASD population. For example, Soke et al. (2017) assessed the association between SIB and a variety of risk factors across two large samples of children with ASD ($n = 8065$ from the Autism and Developmental Disabilities Monitoring Network; $n = 5102$ from the Autism Speaks-Autism Treatment Network). Findings suggested that lower levels of intellectual and adaptive functioning (i.e., higher levels of ID symptomology) within the ASD population were associated with higher levels of SIB. This general relation between SIB and level of intellectual/adaptive functioning has been corroborated elsewhere in the literature (e.g., Baghdadli et al., 2003; Bodfish & Lewis, 2002; Johnson & Day, 1992; Kahng et al., 2002; Rojahn & Esbensen, 2002; Rattaz, Michelon, & Baghdadli, 2015; Saloviita, 2000). However, when examining SIB in a sample of individuals with severe to profound ID that either did or did not carry an ASD diagnosis, Matson et al. (1996) failed to find a meaningful difference in SIB prevalence between these groups. Thus, among those with severe to profound ID, the likelihood of displaying SIB is high independent of whether or not the individual has ASD. Therefore, it appears that carrying an ASD diagnosis increases the risk of engaging in SIB, and the severity of ID symptomology (i.e., levels of intellectual and adaptive functioning) modulates that relation.

Other risk factors for engagement in SIB that have been reported in the literature include (a) the presence of other forms of challenging behavior (e.g., aggression (Carroll, Metcalfe, & Gunnell, 2014; Soke et al., 2017), rituals and stereotypies (Oliver, Petty, Ruddick, & Bacarese-Hamilton, 2012), and impulsivity and hyperactivity (Richman et al., 2013; Rattaz et al., 2015)), (b) sensory-processing issues (Deurden et al., 2012; Soke et al., 2017), (c) comorbid psychopathology and genetic syndromes (e.g., depression, bipolar disorder (Matson & LoVullo, 2008; Sigafoos, O'Reilly, Lancioni, Lang, & Didden, 2014), Lesch-Nyhan and Cornelia de Lange syndromes (Winchel & Stanley, 1991; Fragile X; Hall, Lighthouse, & Reiss, 2008)), (d) the severity of ASD symptomatology (i.e., deficits in social communication and repetitive behavioral patterns, e.g., Baghdadli et al., 2003; Matson & Rivet, 2008; Rattaz et al., 2015), (e) younger age (Baghdadli et al., 2003; Soke et al., 2017), and (f) lower socioeconomic status (SES; Soke et al., 2017). However, these findings have not been consistently reported within the literature. That is, other researchers have failed to find significant associations between SIB and the severity of ASD symptomology (e.g., Duerden et al., 2012; Soke et al., 2017), age (e.g., Deurden et al., 2012; Murphy et al., 2009), and SES (Baghdadli et al., 2003; Schroeder et al., 2014). Thus, the extent to which these factors may increase the risk of SIB within the ID population remains unclear and warrants future research.

Finally, from an environmental perspective, it has been reported that individuals with ID living in residential placements, relative to those living in community settings, have a higher probability of engaging in SIB (Borthwick-Duffy, 1994; Holden & Gitlesen, 2006; Lowe et al., 2007). As

noted previously, however, individuals who engage in SIB and other forms of challenging behavior are at an increased risk for institutional placement (Emerson, 2000; Hyman et al., 1990). Thus, the nature of any putative cause-effect relation between these variables remains uncertain. On the one hand, it could be the case that residential placement increases the risk that individuals in the ID population will engage in SIB. On the other hand, those placed in residential facilities may have received those placements *because* they engaged in SIB. It is also possible that those individuals living in restricted placements have lower levels of intellectual and/or adaptive functioning compared to their peers living in the community. Again, lower levels of intellectual/adaptive functioning are risk factors for engagement in SIB, independently of residential placement. Thus, it is unclear to what extent an individual's living situation independently contributes to the presence of SIB within the ID population, and additional research is needed on this topic.

Rituals and Stereotypies

Similar to SIB, both rituals and stereotypies may be defined generally as atypical patterns of repetitive behavior. Ritualistic behavior has been defined more specifically as repetitive adherence to self-imposed rules (Zohar & Felz, 2001) and may also be conceptualized as compulsions (i.e., repetitive behavior the individual feels driven to complete in response to an obsession or that occurs in accordance to a set of self-imposed rules that must be followed rigidly; American Psychiatric Association [APA], 2013). Examples of rituals may include repetitive hand washing, repeated checking (e.g., confirming that the door is locked over and over), and lining up or arranging objects in a specific manner. However, it is worth noting that ritualistic behavior can be a part of a child's typical development (e.g., repeating nursery rhymes; Bolton, 1996; Evans & Gray, 2000). These behaviors become disorderly when they cause clinically significant impairment (APA, 2013).

As for stereotypy, there has been disagreement within the literature on a precise definition. Broadly speaking, however, stereotypy has been defined as repetitive, topographically invariant, rhythmical behavioral patterns that do not appear to have a clear social function (Powell et al., 1999; Wunderlich & Vollmer, 2015). Stereotypic behaviors often include repetitive motor movements (e.g., body rocking, hand flapping) or repetitive vocalizations (e.g., recitation of words or production of sounds). In some cases, nonrepetitive behavior such as posturing (Rapp & Vollmer, 2005) may be defined as stereotypy. Similar to ritualistic behavior, stereotyped responding may be part of a child's typical development (e.g., foot kicking; Symons, Sperry, Dropik, & Bodfish, 2005) but can become disorderly when these behaviors are displayed at high rates, appear atypical in their manifestation, and/or cause clinically significant impairment such as interfering with the individual's ability to socialize and acquire new skills (APA, 2013; Didden et al., 2012; Lanovaz, Robertson, Soerono, & Watkins, 2013; Reese, Richman, Belmont, & Morse, 2005).

Based on reports from the literature, rituals and stereotypies are common within the ID population (e.g., Matson & Dempsey, 2008; Matson & Dempsey, 2009; Matson & Rivet, 2008), with prevalence estimates of 40% and 60%, respectively (Bodfish et al., 1995). In regard to the risk factors associated with rituals and stereotypies, both have been found to be negatively correlated with intellectual functioning (Berkson, Rafaeli-Mor, & Tarnovsky, 1999; Bodfish et al., 1995; Lundqvist, 2013). That is, among those with an ID, the severity of ID symptomatology (i.e., lower levels of intellectual and adaptive functioning) is a risk factor for rituals and stereotypies. In addition, a comorbid ASD diagnosis is a strong risk factor for rituals and stereotypies (e.g., Bodfish et al., 2000; Matson & Dempsey, 2009). This finding may not be surprising, given that repetitive behaviors such as rituals and stereotypies are defining features of ASD (APA, 2013).

Nonetheless, to examine the relation between ID and the occurrence of rituals and stereotypies and the extent to which a comor-

bid ASD diagnosis moderated this relation, Matson et al. (1996) compared levels of rituals and stereotypies displayed by a sample of 185 individuals with severe or profound ID, with and without ASD. Results indicated that these repetitive behavioral patterns were more prevalent in the ASD group (i.e., 75% of participants with ASD exhibited these patterns) compared to the non-autistic group (7% of participants without ASD). As another example, Matson and Dempsey (2008) conducted a study that examined the presence of rituals and stereotypies in a sample of 336 adults across two groups: adults with a comorbid ASD and ID diagnosis and adults with an ID alone. Outcomes again suggested that those with a comorbid diagnosis of ASD were more likely to engage in rituals and stereotypies (73% of adults with ASD and ID engaged in these behaviors; 45% of adults with ID alone). Collectively, these findings indicate that the presence of ASD symptomatology is a strong risk factor for rituals and stereotypies within the ID population.

Finally, similar to what was described with SIB, environmental factors have also been found to play a role in the occurrence of rituals and stereotypies within the ID population. That is, ritualistic or stereotyped behavioral patterns have been linked to impoverished environments or environments that provide excessive levels of stimulation (Repp, Karsh, Deitz, & Singh, 1992). On the one hand, it has been postulated that barren environments (i.e., those lacking stimulation) may not provide adequate opportunities for these individuals to acquire more adaptive behavioral patterns (Didden et al., 2012) and may inadvertently support the maintenance of ritualistic and stereotyped behavior. On the other hand, some have argued that excessively stimulating environments (e.g., grocery store) may produce anxiety or may be otherwise aversive and that stereotypies may reduce these anxiogenic or aversive properties (see Lutz, 2014; Péter, Oliphant, & Fernandez, 2017). Thus, environments that offer levels of stimulation at either extreme (i.e., impoverished or overly abundant) may increase the likelihood that individuals engage in stereotyped behavior.

Assessment of Self-Injury, Stereotypies, and Rituals

The previous sections of this chapter focused on understanding the etiology of SIB, rituals, and stereotypies. They also offered guidance in terms of defining the topography, or form, of these behaviors. Although information regarding topography is important for understanding the behavior desired for change, additional information is warranted for aiding in the development of an effective treatment for these behaviors (Cunningham & Schreibman, 2008). That is, treatment development is best informed by understanding the functional properties of the behavior or *why* it occurs. This functional approach to the assessment and treatment of SIB, rituals, and stereotypies in individuals with ID is rooted in behavioral/learning theory or more specifically applied behavior analysis (ABA; Minshawi et al., 2015a, b). Note that, in this section, we often will refer to SIB, rituals, and stereotypies simply as "behavior" to reduce our repetitive, topographically invariant, rhythmical typing behavior. We also use the term "behavior" as the same assessment strategies may be used to determine the functional properties of many different types of challenging behaviors (e.g., aggression, property destruction).

Utilization of ABA methodologies allows practitioners and caregivers to gain a better understanding of behavior that is targeted for treatment by not only observing the physical properties of the behavior but also by understanding the environmental variables that affect its occurrence and maintenance. That is, by analyzing the events that precede (i.e., antecedents) and those that follow (i.e., consequences) a behavior, we can understand more precisely the environmental circumstances that cause behavior to occur. Further, we can arrange reinforcement and/or punishment contingencies to promote more socially appropriate forms of behavior

(e.g., communication) and decrease the occurrence of the targeted behavior (e.g., self-injury).

Given the heterogeneity of the behaviors of concern discussed in this chapter, a function-based assessment approach, informed by an individual's clinical presentation, rather than a topographical approach is a crucial step in treatment development. There are various methods for assessing the occurrence of SIB, rituals, and stereotypies in order to develop effective treatment procedures for these behaviors. However, the ease of implementation, as well as the accuracy of outcomes, varies across different assessment procedures. In the remaining sections of this chapter, we will briefly discuss various assessment methodologies, highlighting the advantages and disadvantages of each.

Functional Analysis

A functional analysis (FA), considered the "gold standard" of behavioral assessment, involves experimentally manipulating the environmental variables hypothesized to affect the occurrence of problem behavior such as SIB, rituals, and stereotypies. Potential reinforcement contingencies for problem behavior are arranged within test conditions, and outcomes from test conditions are compared to those from a control condition in which one would not expect to observe the behavior in question. Observing high levels of behavior in one or more test condition(s), relative to the control condition, helps caregivers and practitioners confirm the operant function of the behavior. For example, one could conclude a negative reinforcement function of SIB, if high rates of behavior were observed during a condition in which academic work was presented and subsequently discontinued contingent on the occurrence of SIB, relative to behavior in a control condition during which academic work was not presented.

The use of an FA to determine the operant function of challenging behavior was first recognized in the scientific literature by Iwata et al. (1982/1994), which is commonly termed the "standard" FA. Iwata and colleagues' study demonstrated the utility of FA methodology for effectively assessing the variables that affected the occurrence of SIB across nine participants. That is, the experimenters arranged conditions to test the effects of social positive reinforcement (e.g., contingent attention), social negative reinforcement (e.g., contingent escape from academic demands), and automatic reinforcement (e.g., minimal stimulation context) on the effects of SIB and compared the outcomes to a control condition (e.g., unstructured play). Results of the study showed variability both within and between subjects and, for the majority of participants, correspondence between high levels of SIB with specific environmental stimulus conditions (e.g., academic demands). Thus, these study outcomes highlighted two important considerations for treatment development: (1) Treatment should be developed based on each individual's clinical presentation (given between-subject variability), and (2) experimental manipulations can clearly identify environmental stimuli that occasion problem behavior (given within-subject variability).

The FA methodology offers a variety of advantages over less systematic assessment approaches (e.g., indirect measures – to be described below), namely, confirmation of behavioral function to best inform treatment development. Given the benefits of FA methodology, decades of research have focused on variations of the "standard" FA procedures to best match the needs of the individual, behavior, and context (e.g., public school, private clinic) in which assessment occurs. That is, these procedures have been extended in a number of ways to increase ease of implementation across various contexts (e.g., trial-based functional analysis may be more appropriate when the resources required to conduct a "standard" functional analysis are absent) and reduce the negative collateral outcomes associated with dangerous forms of challenging behavior (e.g., discontinuing assessment conditions following the first occurrence of behavior and measuring latency to engage in the targeted behavior). A full review of FA methodologies is outside the scope of this chapter, but we direct readers to Hanley, Iwata, and McCord (2003) and Beavers, Iwata, and Lerman (2013) for reviews.

Despite the significant benefits FA methodology provides for developing effective treatments for challenging behaviors, there are some considerations caregivers should take into account prior to conducting an FA. Implementing FA procedures requires extensive training and expertise, in addition to significant resources. For example, it can often be challenging to identify a location to conduct a functional analysis that allows for complete control over all the variables present that may affect the occurrence of behavior. Caregivers must also consider the potential risks associated with implementation of an FA, such as possible injury (e.g., occurrence of SIB) that may occur given the intentional arrangement of variables hypothesized to evoke and maintain problem behavior. If conducting an FA is not feasible (e.g., lack of trained professionals), implementation of other, albeit less precise, behavioral assessment approaches can be considered.

Indirect and Direct Assessment

Indirect assessment measures involve reporting of information related to the occurrence of problem behavior(s) like SIB, rituals, and stereotypies through rating scales and questionnaires, as well as structured or unstructured interviews. Information is often gathered from caregivers (e.g., parent, teacher) of the individual whose behavior is being evaluated but can also be obtained from the individual himself/herself, when feasible. Indirect assessment measures may be most useful when problem behavior occurs infrequently or at high intensities (Lloyd & Kennedy, 2014). Furthermore, indirect assessments can be useful as a starting point for gathering information to conduct more formal assessments. For example, if caregivers endorse a specific function of problem behavior (i.e., to gain access to attention or tangible items, to escape aversive situations), FA methodologies may then be applied to experimentally analyze and confirm/disconfirm the presence of that particular function. Confirmation of outcomes using multiple assessment strategies generally increases the degree of confidence one may have when making data-based decisions about the likely function of the behavior being assessed (see Kazdin, 2011).

There are several published rating scales, checklists, and structured interviews with varying evidence of reliability and validity. Two commonly used measures include the Questions About Behavioral Function (QABF; Matson & Vollmer, 1995) and the Functional Analysis Interview (FAI; O'Neill et al., 1997). The QABF is a checklist aimed at gathering information regarding environmental correlates of target behavior to aid in the conclusion of the function of the behavior. One potential advantage of the QABF, over similar rating scales or checklists, is that this tool assesses the presence of seemingly less common functions of challenging behavior (e.g., social avoidance, physical discomfort) in addition to the more readily assessed functions (e.g., negative reinforcement in the form of escape from academic tasks).

The FAI is a structured interview aimed at quick (i.e., 45–90-min administration) identification of environmental variables that potentially affect the occurrence of challenging behavior. The interview is designed to assist the clinician in development of an effective intervention by identifying operational definitions of the target behavior, setting events, antecedent events that occasion behavior, consequent events that maintain behavior, the efficiency of the behavior, the individual's adaptive and communicative repertoire, potential reinforcers, and previously implemented treatment strategies. Outcomes from the FAI generally provide a summary statement that includes the situation and context in which behavior typically occurs, a description of the target behavior, and the function of the behavior.

Advantages of implementing indirect assessment methods include the procedures being time- and cost-efficient, in addition to the need for less training and expertise relative to more formal methods (e.g., FA; Kelley, LaRue, Roane, & Gadaire, 2011). However, the validity of the information gleaned from indirect assessments is limited. Self-report measures and reports solicited from stakeholders in the care of an individual may not accurately reflect the intensity, fre-

quency, or severity of the individual's behavior and may be influenced by extraneous factors (Kazdin, 2011). For example, parental stress may influence the extent to which parents report their child's behavior as concerning or deviant. Further, research has demonstrated a lack of correspondence between the outcomes from indirect and experimental behavioral assessments.

Direct assessment measures involve obtaining a descriptive account of the problem behavior as it is occurring in the natural environment. That is, data are gathered during observations of the individual within the context in which problem behavior like SIB, rituals, and stereotypies is reported to occur. Data can be obtained through anecdotal notes recorded during the observation or from more structured data collection methods, such as behavioral recording sheets or checklists.

Similar to indirect assessments, there are several tools used to complete direct assessments. However, unlike with indirect, direct assessments involve observation of the target behavior within contrived or natural situations. These observations lack the actual manipulation of environmental variables to confirm behavioral function characteristic of FA methods. Furthermore, direct assessments involve some form of data collection on the target behavior. Two forms of direct assessment tools that are readily used in practice, likely due to the ease of implementation, include the Antecedent-Behavior-Consequence (ABC) Checklist (Miltenberger, 2004) and the scatterplot. Descriptive analysis given an ABC checklist involves conducting direct observations of the individual and placing a check mark next to predefined (based on information from caregiver interviews, etc.) antecedent and consequent events that were present at the time the target problem behavior occurred. Information gleaned from an ABC checklist may be insufficient for confirming behavioral function, as the temporal relation between the behavior and the noted environmental events is unknown. Furthermore, frequency and/or duration of the target behavior may not be readily available given data provided by an ABC checklist.

The scatterplot is another direct assessment strategy that involves observation of the individual and recording the frequency of the target behavior during specified time intervals (e.g., 15-min intervals) within a given observation period (e.g., throughout the school day). However, the scatterplot does not include recording of environmental events that precede or follow the behavior. Overall, the scatterplot can assist with identifying temporal patterns of the target behavior that may aid in the implementation of treatment strategies during specified times and/or isolate times to conduct further assessment of the environmental variables that affect the occurrence of the target behavior.

Direct assessment methods, relative to indirect methods, require more training, time, and resources to effectively implement. However, direct assessment can allow for a better understanding of the relationship between the problem behavior and specific environmental variables than indirect assessments. Despite this advantage, direct observations alone cannot *confirm* the operant function of the problem behavior, as in an FA. However, given obtained conditional probabilities regarding behavior-environment relations, direct assessment measures may also be a useful tool in the development of an FA for target behavior.

Summary

Self-injury, rituals, and stereotypies are common behavioral concerns in the ID population. The goals of this chapter were to (a) define these behaviors to provide readers with some guidance in distinguishing between them; (b) review the risk factors associated with SIB, rituals, and stereotypies; and (c) review the assessment strategies that may usefully be applied to best understand why individuals engage in these behaviors to ultimately aid in the development of effective treatment. A brief overview of these elements is provided below.

While these behaviors may share some general features (e.g., they all may be repetitive in nature), SIB, rituals, and stereotypies may be dif-

ferentiated based on other characteristics. For example, SIB refers more specifically to behaviors that cause harm to the individual engaging in the behavior; rituals are behaviors that are controlled, at least in part, by self-imposed rules; and stereotypies refer to repetitive motor or vocal behaviors that lack the components of SIB and rituals just described. That is not to say that these behavioral patterns always are distinct – stereotypies and rituals may produce physical harm (e.g., repeated forceful vocalizations may damage vocal cords), and topographies of behavior often considered to be self-injurious may not always produce physical harm (e.g., head hitting may not always occur with sufficient force to cause soft tissue damage). Careful operational definitions of these behaviors may help caregivers and practitioners distinguish between them in clinical and community settings.

The risk factors associated with SIB, rituals, and stereotypies share substantial overlap. Specifically, severity of ID symptomology is positively associated with each of these patterns, and ASD diagnostic status moderates this relationship (i.e., individuals with ID and ASD diagnoses tend to engage in SIB, rituals, and stereotypies more frequently than those without ASD). Environmental factors may also play a role in the development and maintenance of these behavioral patterns. Specifically, institutionalization may result in more frequent SIB, though the direction of the cause-effect relation between these variables remains uncertain. Further, excessively stimulating environments, or environments devoid of external stimulation, may be associated with increased incidence of rituals and stereotypies.

Behavior-analytic assessment strategies (e.g., FA) result in confirmation of the variables that maintain SIB, rituals, and stereotypies, which aids in the development of effective treatments. The functional analysis approach utilizes an experimental perspective to identify the variables that maintain SIB, rituals, and stereotypies. Though FA methodologies may clearly isolate these variables, their implementation is not always feasible (i.e., substantial training and environmental resources may be required for their application). Direct (e.g., structured observation) and indirect (e.g., parent interviews or interviews with the individual who engages in the behavior) assessments may provide critical information about the determinants of behavioral patterns, which can also aid in the development of treatment strategies. Overall, implementation of the most appropriate assessment strategies to inform treatment development should follow a best-practice approach and thus be guided by a practitioner's clinical expertise, knowledge of the literature base, and understanding of the patient, student, or client's characteristics.

References

American Psychiatric Association. (2013). *Diagnostic and statistical manual for mental disorders* (5th ed.). Arlington, VA: Author. https://doi.org/10.1176/appi.books.9780890425596

Anderson, L. T., & Ernst, M. (1994). Self-injury in Lesch-Nyhan disease. *Journal of Autism and Developmental Disorders, 24*(1), 67–81. https://doi.org/10.1007/BF02172213

Baghdadli, A., Pascal, C., Grisi, S., & Aussilloux, C. (2003). Risk factors for self-injurious behaviors among 222 young children with autistic disorders. *Journal of Intellectual Disability Research, 47*(8), 622–627. https://doi.org/10.1046/j.1365-2788.2003.00507.x

Ballinger, B. R. (1971). Minor self-injury. *The British Journal of Psychiatry, 118*, 535–538. https://doi.org/10.1192/bjp.118.546.535

Barmann, B. C. (1979). The use of overcorrection with artificial nails in the treatment of chronic fingernail-biting. *Mental Retardation, 17*, 309–311.

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. https://doi.org/10.1002/bin.228

Baxter, C., Cummins, R. A., & Yiolitis, L. (2000). Parental stress attributed to family members with and without intellectual disability: A longitudinal study. *Journal of Intellectual and Developmental Disability, 25*(2), 105–118. https://doi.org/10.1080/13269780050033526

Brian A. Iwata, Michael F. Dorsey, Keith J. Slifer, Kenneth E. Bauman, Gina S. Richman, (1994) TOWARD A FUNCTIONAL ANALYSIS OF SELF-INJURY. *Journal of Applied Behavior Analysis, 27* (2):197-209

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

Berkson, G., Rafaeli-Mor, N., & Tarnovsky, S. (1999). Body-rocking and other habits of college students and persons with mental retardation. *American Journal on Mental Retardation, 104*(2), 107–116. https://doi.org/10.1352/0895-8017(1999)104<0107:BAOHOC>2.0.CO;2

Bodfish, J. W., Crawford, T. W., Powell, S. B., & Parker, D. E. (1995). Compulsions in adults with mental retardation: Prevalence, phenomenology, and comorbidity with stereotypy and self-injury. *American Journal on Mental Retardation, 100*(2), 183–192.

Bodfish, J. W., & Lewis, M. H. (2002). Self-injury and comorbid behaviors in developmental, neurological, psychiatric, and genetic disorders. In S. R. Schroeder, M. L. Oster-Granite, & T. Thompson (Eds.), *Self-Injurious Behavior: Gene-Brain-Behavior Relationships* (pp. 23–39). Washington, DC: US: American Psychological Association. https://doi.org/10.1037/10457-002

Bodfish, J. W., Symons, F. J., Parker, D. E., & Lewis, M. H. (2000). Varieties of repetitive behavior in autism: Comparisons to mental retardation. *Journal of Autism and Developmental Disorders, 30*(3), 237–243. https://doi.org/10.1023/A:100559650

Blair P. Lloyd, Craig H. Kennedy, (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

Bolton, D. (1996). Annotation: Developmental issues in obsessive-compulsive disorder. *Journal of Child Psychology and Psychiatry, 37*(2), 131–137. https://doi.org/10.1111/j.1469-7610.1996.tb01384.x

Borthwick-Duffy, S. A. (1994). Prevalence of destructive behaviors: A study of aggression, self-injury, and property destruction. In T. Thompson & D. B. Gray (Eds.), *Destructive behavior in developmental disabilities: Diagnosis and treatment* (pp. 3–23). Thousand Oaks, CA: Sage Publishing.

Carroll, R., Metcalfe, C., & Gunnell, D. (2014). Hospital management of self-harm patients and risk of repetition: Systematic review and meta-analysis. *Journal of Affective Disorders, 168*, 476–483. https://doi.org/10.1016/j.jad.2014.06.027

Collacott, R. A., Cooper, S. A., Branford, D., & McGrother, C. (1998). Epidemiology of self-injurious behaviour in adults with learning disabilities. *The British Journal of Psychiatry, 173*(5), 428–432. https://doi.org/10.1192/bjp.173.5.428

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*. https://doi.org/10.1111/j.1365-2788.2008.01060.x

Cunningham, A. B., & Schreibman, 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

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*, 764–765. https://doi.org/10.1016/j.ridd.2012.10.004

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. https://doi.org/10.1007/978-1-4614-3037-7_3

Dominick, K. C., Davis, N. O., Lainhart, J., Tager-Flusberg, H., & Folstein, S. (2007). Atypical behaviors in children with autism and children with a history of language impairment. *Research in Developmental Disabilities, 28*(2), 145–162. https://doi.org/10.1016/j.ridd.2006.02.003

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*, 2460–2470

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. https://doi.org/10.1007/s10803-012-1497-9

Emerson, E. (2000). Developmental disability and behaviour. Gillberg, C. & O'Brien, G (eds.). London: MacKeith Press.

Edward G. Carr, (1977) The motivation of self-injurious behavior: A review of some hypotheses.. *Psychological Bulletin, 84*(4):800–816

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

Evans, D. W., & Gray, F. L. (2000). Compulsive-like behavior in individuals with down syndrome: Its relation to mental age level, adaptive and maladaptive behavior. *Child Development, 71*(2), 288–300. https://doi.org/10.1111/1467-8624.00144

Hall, S. S., Lighthouse, A. A., & Reiss, A. L. (2008). Compulsive, self-injurious, and autistic behavior in children and adolescents with fragile x syndrome. *American Journal of Mental Retardation, 113*, 44–53. https://doi.org/10.1352/0895-8017(2008)113[44:CSAABI]2.0.CO

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

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

Hyman, S. L., Fisher, W., Mercugliano, M., & Cataldo, M. F. (1990). Children with self-injurious behavior. *Pediatrics, 85*, 437–441.

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*, 197–209. https://doi.org/10.1901/jaba.1994.27-197. (Reprinted from Analysis and Intervention in Developmental Disabilities, 2, 3-20, 1982). https://doi.org/10.1901/jaba.1994.27-197

Johnson, W. L., & Day, R. M. (1992). The incidence and prevalence of self-injurious behavior. In J. K. Luiselli, J. L. Matson, & N. N. Singh (Eds.), *Self-injurious behavior: Analysis, assessment, and treatment* (pp. 21–56). New York, NY: Springer. https://doi.org/10.1007/978-1-4613-9130-2_2

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. https://doi.org/10.1352/0895-8017(2002)107<0212:BTOSIT>2.0.CO;2

Kazdin, A. E. (2011). *Single-case research designs: Methods for clinical and applied settings* (2nd ed.). New York, NY: Oxford University Press. https://doi.org/10.1192/S0007125000200706

Kelley, M. E., LaRue, R. H., Roane, H. S., & Gadaire, D. M. (2011). Indirect behavioral assessments: Interviews and rating scales. In W. W. Fisher, C. C. Piazza, & H. S. Roane (Eds.), *Handbook of applied behavior analysis* (pp. 182–190). New York, NY: The Guilford Press.

Klonsky, E. D., & Meuhlenkamp, J. J. (2007). Self-injury: A research review for the practitioner. *Journal of Clinical Psychology: In Session, 63*, 1045–1056. https://doi.org/10.1002/jclp.20412

Ladouceur, R. (1979). Habit reversal treatment: Learning an incompatible response or increasing subject awareness? *Behavior Research and Therapy, 17*, 313–316. https://doi.org/10.1016/0005-7967(79)90003-2

Lang, R., Didden, R., Machalicek, W., Rispoli, M., Sigafoos, J., Lancioni, G., … Kang, S. (2010). Behavioral treatment of chronic skin-picking in individuals with developmental disabilities: A systematic review. *Research in Developmental Disabilities, 31*(2), 304–315. https://doi.org/10.1016/j.ridd.2009.10.017

Lanovaz, M. J., Robertson, K., Soerono, K., & Watkins, N. (2013). Effects of reducing stereotypy on other behaviors: A systematic review. *Research in Autism Spectrum Disorders, 7*, 1234–1243. https://doi.org/10.1016/j.rasd.2013.07.009

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

Lundqvist, L. O. (2013). Prevalence and risk markers of behavior problems among adults with intellectual disabilities: A total population study in Orebro County, Sweden. *Research in Developmental Disabilities, 34*, 1346–1356. https://doi.org/10.1016/j.ridd.2013.01.010

Lutz, C. K. (2014). Stereotypic behavior in nonhuman primates as a model for the human condition. *ILAR Journal, 55*, 284–296. https://doi.org/10.1093/ilar/ilu016

K. McClintock, S. Hall, C. Oliver, (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

K. Mark Derby, David P. Wacker, Gary Sasso, Mark Steege, John Northup, Karla Cigrand, Jennifer Asmus, (1992) brief functional assessment techniques to evaluate aberrant behavior in an outpatient setting: a summary of 79 cases. *Journal of Applied Behavior Analysis, 25* (3):713–721

Miltenberger, R. G., (2004). Behavior Modification: Principles and Procedures (4th ed.) Belmont, CA: Wadsworth

Matson, J. L., & Vollmer, T. R. (1995). User's guide: Questions About Behavioral Function (QABF). Baton Rouge, LA: Scientific Publishers, Inc.

Matson, J. L., Baglio, C. S., Smiroldo, B. B., Hamilton, M., Packlowskyj, T., Williams, D., & Kirkpatrick-Sanchez, S. (1996). Characteristics of autism as assessed by the diagnostic assessment for the severely handicapped-II (DASH-II). *Research and Developmental Disabilities, 17*, 135–143. https://doi.org/10.1016/0891-4222(95)00044-5

Matson, J. L., & Dempsey, T. (2008). Stereotypy in adults with autism spectrum disorders: Relationship and diagnostic fidelity. *Journal of Developmental and Physical Disabilities, 20*(2), 155–165. https://doi.org/10.1007/s10882-007-9086-0

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. https://doi.org/10.1016/j.ridd.2008.10.001

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. https://doi.org/10.1177/0145445507304581

Matson, J. L., & Rivet, T. T. (2008). Characteristics of challenging behaviours in adults with autistic disorder, PDD-NOS, and intellectual disability. *Journal of Intellectual and Developmental Disability, 33*(4), 323–329. https://doi.org/10.1080/13668250802492600

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 Disorders, 5*(3), 1215–1222. https://doi.org/10.1016/j.rasd.2011.01.009

Minshawi, N. F., Hurwitz, S., Morriss, D., & McDougle, C. J. (2015a). 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*, 1541–1568. https://doi.org/10.1007/s10803-014-2307-3

Minshawi, N. F., Hurwitz, S., Morriss, D., & McDougle, C. J. (2015b). 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

Murphy, G. H., Oliver, C., & Corbett, J. (1993). Epidemiology of self-injury, characteristics of people with severe self-injury and initial treatment outcome. In C. Kiernan (Ed.), *Research to practice? Implication of research on challenging behavior with learning disability* (pp. 1–35). Avon: British Institute of Learning Disabilities.

Murphy, O., Healy, O., & Leader, G. (2009). Risk factors for challenging behaviors among 157 children with autism spectrum disorder in Ireland. *Research in Autism Spectrum Disorders, 3*(2), 474–482. https://doi.org/10.1016/j.rasd.2008.09.008

O'Neill, R. E., Horner, R. H., Albin, R. W., Sprague, J. R., Storey, K., & Newton, J. S. (1997). Functional assessment and program development for problem behavior: A practical handbook. Pacific Grove, CA: Brooks/Cole Publishing.

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/10.1046/j.1365-2788.1997.04545.x

Oliver, C., Murphy, G. H., & Corbett, J. A. (1987). Self-injurious behaviour in people with mental handicap: A total population study. *Journal of Intellectual Disability Research, 31*(2), 147–162. https://doi.org/10.1111/j.1365-2788.1987.tb01351.x

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*, 910–919. https://doi.org/10.1007/s10803-011-1320-z

Péter, Z., Oliphant, M. E., & Fernandez, T. V. (2017). Motor stereotypies: A pathophysiological review. *Frontiers in Neuroscience, 11*, 171. https://doi.org/10.2289/fnins.2017.00171

Powell, S. B., Newman, H. A., Pendergast, J. F., & Lewis, M. H. (1999). A rodent model of spontaneous stereotypy: Initial characterization of developmental, environmental, and neurobiological factors. *Physiology & Behavior, 66*(2), 355–363. https://doi.org/10.1016/s0031-9384(98)00303-5

Rapp, J. T., & Vollmer, T. R. (2005). Stereotypy I: A review of behavioral assessment and treatment. *Research in Developmental Disabilities, 26*, 257–547. https://doi.org/10.1016/j.ridd.2004.11.005

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*(8), 730–741. https://doi.org/10.1111/jir.12177

Reese, R. M., Richman, D. M., Belmont, J. M., & Morse, P. (2005). Functional characteristics of disruptive behavior in developmentally disabled children with and without autism. *Journal of Autism and Developmental Disorders, 35*, 419–428. https://doi.org/10.1007/s10803-005-5032-0

Repp, A. C., Karsh, K. G., Deitz, D. E. D., & Singh, N. N. (1992). A study of the homeostatic level of stereotypy and other motor movements of persons with mental handicaps. *Journal of Intellectual Disability Research, 36*(1), 61–75. https://doi.org/10.1111/j.1365-2788.1992.tb00471.x

Richards, C., Davies, L., & Oliver, C. (2017). Predictors of self-injurious behavior and self-restraint in autism spectrum disorder: Towards a hypothesis of impaired behavioral control. *Journal of Autism and Developmental Disorders, 47*(3), 701–713. https://doi.org/10.1007/s10803-016-3000-5

Richards, C., Oliver, C., Nelson, L., & Moss, J. (2012). Self-injurious behaviour in individuals with autism spectrum disorder and intellectual disability. *Journal of Intellectual Disability Research, 56*(5), 476–489. https://doi.org/10.1111/j.1365-2788.2012.01537.x

Richman, D., 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, 57*, 429–439. https://doi.org/10.1111/j.1365-2788.2012.01628.x

Robertson, J., Emerson, E., Pinkney, L., Caesar, E., Felce, D., Meek, A., … Hallman, A. (2004). Quality and costs of community-based residential supports for people with mental retardation and challenging behavior. *American Journal on Mental Retardation, 109*, 332–344. https://doi.org/10.1352/0895-8017(2004)109<332:QACOCR>2.0CO;2

Rojahn, J., & Esbensen, A. J. (2002). Epidemiology of self-injurious behavior in mental retardation: A review. In S. R. Schroeder, M. L. Oster-Granite, & T. Thompson (Eds.), *Self-injurious behavior: Gene-brain-behavior relationships* (pp. 41–77). Washington, DC: American Psychological Association. https://doi.org/10.1037/10457-003

Rojahn, J., Whittaker, K., Hoch, T. A., & Gonzáles, M. L. (2007). Assessment of self-injurious and aggressive behavior. In J. L. Matson (Ed.), *Handbook of assessment in persons with intellectual disability* (pp. 281–319). San Diego: Academic. https://doi.org/10.1016/S0074-7750(07)34009-3

Saloviita, T. (2000). The structure and correlates of self-injurious behaviour in an institutional setting. *Research in Developmental Disabilities, 21*, 501–511. https://doi.org/10.1016/S0891-4222(00)00055-X

Schroeder, S. R., Marquis, J. G., Reese, R. 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 on Intellectual and Developmental Disabilities, 119*(4), 351–370. https://doi.org/10.1352/1944-7558-119.4.351

Sigafoos, J., O'Reilly, M. F., Lancioni, G. E., Lang, R., & Didden, R. (2014). Self-injurious behavior. In P. Sturmey & R. Didden (Eds.), *Evidence-based practice and intellectual disabilities* (pp. 133–162). https://doi.org/10.1002/9781118326077.ch6

Soke, G. N., Rosenberg, S. A., Hamman, R. F., Fingerlin, T., Rosenberg, C. R., Carpenter, L., … Reynolds, A. (2017). Factors associated with self-injurious behaviors in children with autism spectrum disorder: Findings from two large national samples. *Journal of Autism and Developmental Disorders, 47*(2), 285–296. https://doi.org/10.1007/s10803-016-2951-x

Symons, F. J., Sperry, L. A., Dropik, P. L., & Bodfish, J. W. (2005). The early development of stereotypy and self-injury: A review of research methods. *Journal of Intellectual Disability Research, 49*(2), 144–158. https://doi.org/10.1111/j.1365-2788.2004.00632.x

Tate, B. G., & Baroff, G. S. (1966). Aversive control of self-injurious behavior in a psychotic boy. *Behavior Research and Therapy, 4*, 281–287. https://doi.org/10.1016/0005-7967(66)90024-6

Weiss, J. (2003). Self-injurious behaviours in autism: A literature review. *Journal of Developmental Disabilities, 9*, 129–143.

Winchel, R. M., & Stanley, M. (1991). Self-injurious behavior: A review of the behavior and biology of self-mutilation. *American Journal of Psychiatry, 148*, 306–317. https://doi.org/10.1176/ajp.148.3.306

Wunderlich, K. L., & Vollmer, T. R. (2015). Data analysis of response interruption and redirection as a treatment for vocal stereotypy. *Journal of Applied Behavior Analysis, 48*, 749–764. https://doi.org/10.1002/jaba.227

Zohar, A. H., & Felz, L. (2001). Ritualistic behavior in young children. *Journal of Abnormal Child Psychology, 29*(2), 121–128. https://doi.org/10.1023/A:1005231912747

Zohar, A. H., & Felz, L. (2001). Ritualistic behavior in young children. Journal of Abnormal Child Psychology, 29, 121-128

22 Feeding Problems and Assessment in Individuals with Intellectual Disability

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Feeding Problems and Assessment in Individuals with Intellectual Disability

Children and adults with intellectual disability (ID) face a variety of challenges regarding the development of and participation in activities of daily living. One of the most significant challenges relates to the high comorbidity of feeding problems. Prevalence estimates of feeding problems vary but occur in 25–45% of typically developing children (Forsyth, Leventhal, & McCarthy, 1985). However, an estimated 80–90% of children with an identified developmental disability present with some level of feeding concern (Lefton-Greif & Arvedson, 2007), and the prevalence and severity of feeding problems increases with severity of intellectual impairment (Arvedson & Lefton-Greif, 2007). Numerous feeding and eating difficulties occur in individuals with ID but generally fall into one of the following four categories: (1) lack of independent skills needed for eating, (2) eating too much/too little, (3) disruptive mealtime behavior, and (4) selectivity by type or texture (Linscheid, 1983; Sisson & Van Hasselt, 1989). Numerous overlapping and co-occurring etiological pathways contribute to the development of eating difficulties including medical, nutritional, behavioral, psychological, and environmental factors.

Addressing feeding problems in individuals with ID and other neurodevelopmental conditions is vital in fostering growth, cognitive development and learning, and appropriate life span. Often, signs and symptoms of disordered feeding become apparent in the first 1–3 years of the child's life but can remain ongoing into and throughout adulthood if not addressed appropriately (Silverman, 2015). Many healthcare and allied providers are able to provide specific treatment of feeding disorders including those trained in medicine, psychology, speech and language pathology, nutrition, and occupational therapy. While professionals may work alone in the assessment and treatment of such problems, a growing trend in recent decades has involved the use of interdisciplinary teams to assess, track, and treat feeding problems in individuals with intellectual and other neurodevelopmental disabilities (Silverman, 2010). This approach can be particularly efficacious when both medical and physiological concerns co-occur with behavioral feeding challenges.

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This chapter will present an overview of common mealtime and eating concerns often observed in those with ID. Guidelines are provided to assist the clinician in the assessment of behavioral, medical, and physiological factors related to feeding difficulties. Finally, the importance of an interdisciplinary team and collaborative care is discussed as an optimal solution to achieve better outcomes for the individual.

Physiological and Medically-Related Feeding Problems

The initial identification of a medically-related feeding disorder is often first noted when a young child has been identified as "failure to thrive" due to height or weight measurements significantly below what is expected for his or her age. The main reason a young child may experience a deceleration or halt in growth is inadequate nutrition (Larson-Nath & Biank, 2016). If an infant is not offered enough food, is not willing or able to eat enough food, or vomits repeatedly (in the case of severe gastroesophageal reflux), there will not be enough calories to support growth. In the case where the young child cannot absorb enough calories (e.g., severe allergies, celiac disease, or a medical condition like cystic fibrosis), he or she will not grow as anticipated. As can be expected, due to the importance of nutrition in the early developmental period, a diagnosis of failure to thrive can have deleterious outcomes and lead to developmental delays if not treated.

While failure to thrive is certainly a very serious condition, this classification does not adequately capture many of the feeding and mealtime difficulties most often observed in the clinical setting for those with ID. Difficulties in eating occur in those with ID well into adulthood and are most often associated with deficits in feeding skills (Fodstad & Matson, 2008; Linscheid, 1983; Schwarz, 2003; Schwarz, Corredor, Fisher-Medina, Cohen, & Rabinowitz, 2001). Second, many individuals with significant feeding problems do not have growth failure. For example, a child fed through a gastrostomy tube will gain weight adequately but consume nothing by mouth (Wright, Smith, & Morrison, 2011). Similarly, an individual who has severe food selectivity (e.g., only eats junk food) may consume enough food/calories to gain weight adequately, although he or she might be at risk for nutritional deficiency (Johnson et al., 2014; Lane, Geraghty, Young, & Rostorfer, 2014; Schmitt, Heiss, & Campbell, 2008). Third, many feeding problems are associated or precipitated by a concomitant medical condition(s) (Rommel, De Meyer, Feenstra, & Veereman-Wauters, 2003). Fourth, feeding disorders may not be severe until well into adolescence or adulthood (Bryant-Waugh, 2013; Gravestock, 2000).

Numerous general medical conditions may contribute to or account for feeding difficulties (see Table 22.1). For those with ID, gastroenterological problems (GI) occur at a rate higher than typically developing peers, with some estimates indicated as much as 70% are affected (Buie et al., 2010). Commonly reported GI issues include chronic constipation, encopresis, gastroesophageal reflux disease (GERD), abdominal pain, bloating, and diarrhea. In situations where eating is associated with nausea or pain (e.g., food allergies, GERD, gastritis, esophagitis), food refusal or significant aversions to specific tastes may develop (Bernstein, 1999; Birch, 1999). This type of pain-related food refusal may continue to persist even after the medical issue is resolved due to a pain-related history with food and limited experience of non-painful eating. Similarly, individuals with complex medical histories (e.g., medically fragile, cancer, cardiac problems) may also develop oral aversions, avoidance of mealtimes, and texture selectivity issues due to repeated procedures (e.g., intubation), lengthy hospitalizations, being unable to eat by mouth, or medication side effects (Rommel et al., 2003).

For individuals with ID, difficulties in basic feeding skills are often a significant barrier to achieving independence in eating (Fodstad & Matson, 2008). When a person is unable to complete basic skills (e.g., utensil use, neatness, table manners, proper pacing, and oral-motor skills), the ability to eat properly decreases, and the risk for developing a feeding problem increases. The

Table 22.1 Medical conditions commonly associated with dysfunctional feeding/eating patterns

Prematurity/low birth weight
Mitochondrial disease
Metabolic disorders
Niemann-Pick disease
Tay-Sachs disease
Muscular disorders
Cerebral palsy
Muscular dystrophy
Anatomical abnormalities of the mouth or oropharyngeal regions
Cleft palate
Cleft lip
Ankyloglossia (i.e., *tongue-tied)*
Oral-motor dysfunction
Dysphagia
Oral apraxia
Esophageal spasms
Aspiration
Gastrointestinal diseases
Gastroesophageal reflux disease (GERD)
Gastroparesis
Crohn's disease
Food allergies
Lactose intolerance
Celiac disease
Other medical diseases/conditions
Liver disease
Cancer/leukemia
Heart disease
GI inflammation (e.g., *gastritis, duodenitis, esophagitis)*
Infections
Central nervous system infections
Adrenal hyperplasia
Constipation/impaction of bowels

problems associated with poor feeding skills include difficulty swallowing, chewing, and accessing food. Structural abnormalities of the oral/pharyngeal area (e.g., oral/facial clefts, atresia, dental malocclusions, short frenulum, etc.) are more common in those with ID and can make mealtime success difficult. Furthermore, certain neurological conditions often associated with ID place the individual with ID at an even higher likelihood for having feeding skill difficulties. Dysphagia (e.g., oral-motor disorders including sucking, chewing, lingual movement, and swallowing discoordination) occurs in more than half of individuals with Down's syndrome, severe ID, and cerebral palsy (Schwarz, 2003; Robertson, Chadwick, Baines, Emerson, & Hatton, 2017). Further, mealtime difficulties can occur when there are motor control difficulties in other areas of the body that may be involved in eating – for example, positioning, grasping and using utensils, recognizing internal sensations of fullness/hunger, and pacing/endurance during mealtime. When an individual is unable, from an inability or unwillingness, to complete these tasks for a period of time, consequences may occur including malnutrition and starvation. Similarly, if a person is unable to eat at a regular pace (i.e., eating too fast), the risk of choking or aspirating increases dramatically and can cause a potentially life-threatening situation.

The nutritional status of those with ID is also an area of concern linked to healthy eating habits. For example, restrictive eating habits (whether imposed by the individual or due to medical reasons) can lead to nutritional deficits or malnutrition which in the most severe cases may result in a feeding tube to ensure adequate caloric and nutritional intake. Additionally, those with ID are highly likely to engage in behaviors that may lead to significant medical complications including pica (i.e., eating non-food items) and rumination (i.e., repeated regurgitation, chewing, and re-swallowing of previously ingested foods). These behaviors may be behaviorally oriented (e.g., low self-leisure skills/inability to self-stimulate; related to environmental contingencies) but are often related to underlying medical factors (e.g., gastroesophageal reflux [GERD], vitamin/nutrient imbalance). Finally, weight-related concerns (both being underweight and overweight) and sedentary lifestyle are a major concern for those with ID, which is a major contributor to increased medical complications and early death.

Physiological and Medical Feeding Assessment

Given that feeding difficulties in those with ID are a multifactorial and heterogeneous problem, the clinician should ensure that a thorough

assessment is completed. The identification of a medical, motor, or other physiological problem can assist in the treatment or approach to treatment of feeding problems. For example, determining that an individual refuses to eat certain foods (e.g., tomatoes) because it exacerbates his GERD would suggest a medical intervention (e.g., acid-suppressing medication) to suppress the reflux. A medical assessment may involve many components, with the primary goal of determining important factors such as safety to consume solid food, what the individual currently consumes to meet nutritional needs, his or her ability to self-feed or learn self-feeding skills, and if the individual is receiving proper nutrition absorption for weight gain and overall health (Kleinert, 2017).

Before intervention can begin, the individual must be cleared as being able to safely consume solid food. To assess the integrity of the GI systems involved in chewing and swallowing (e.g., hypopharynx and other upper gastrointestinal anatomy) and to ensure that the individual can protect his or her airway during swallowing, a barium swallow study would be indicated (Babbitt et al., 1994). This procedure also provides information regarding the movement of the food/bolus through the upper gastrointestinal tract, which may demonstrate that the individual is bringing food up from the stomach or esophagus back into the mouth (i.e., rumination). It is also useful in diagnosing severe grades of GERD (Ott, 1994). An upper gastrointestinal tract (GI) endoscopy provides information about whether medical conditions exist (e.g., esophagitis) and about the mucosal lining of the esophagus, stomach, and duodenum (Babbitt et al., 1994; Böhmer et al., 1999). The presence of esophageal reflux is also identified using this technique (Kuruvilla & Trewby, 1989). A gastric emptying scan is useful in evaluating motility in the upper gastrointestinal tract (Babbitt et al., 1994). Aberrant results may be associated with a poor appetite. Esophageal manometry is a relatively new technique that measures intraesophageal pressure that provides information about peristalsis and thus the esophageal motility (Patti, Diener, Tamburini, Molena, & Way, 2001).

Nutritionists can also provide valuable information pertaining to feeding problems (O'Brien, Repp, Williams, & Christophersen, 1991). An evaluation of the individual's weight indicates whether he or she is overweight or underweight. An evaluation of an individual's diet ensures that all necessary nutrients are consumed. A nutritionist can assess food allergies that contribute to the presenting problem or identify syndromes that are the basis for problems, such as the inability to digest or metabolize certain proteins.

Instrumental in the evaluation of behavioral feeding problems is an evaluation of the individual's coordination and physical ability to perform various tasks essential to achieving independence in self-feeding. An evaluation of the individual's abilities during mealtime can be conducted by a speech-language pathologist or by an occupational therapist, depending on the nature of the assessment (Kleinert, 2017; O'Brien et al., 1991). The skills evaluated that are necessary for self-feeding include gross reflexive movements, hand-eye coordination, and motor development. The skills evaluated for oral feeding include oral pharyngeal reflexes and oral-motor skills including sucking, swallowing, chewing, and tongue control.

Behavioral Feeding Problems

The importance in evaluating for medically or physiologically related feeding concerns cannot be overemphasized. However, feeding difficulties in individuals with ID are often behavioral in nature. While many toddlers and young children struggle with "picky eating," the prevalence of significant behavioral feeding problems may increase to 70% or above for individuals with significant ID (Rezaei, Rashedi, Gharib, & Lotfi, 2011; Cermak, Curtin, & Bandini, 2010). Several different behavioral feeding difficulties can be seen in individuals with ID, ranging from food selectivity, rumination/vomiting, food refusal, and other negative mealtime behavior problems (Matson & Fodstad, 2009). These children often still consume enough food to gain weight appropriately but are at risk for nutritional deficiency

due to the limited nature of their diet (Johnson et al., 2014; Lane et al., 2014; Schmitt et al., 2008).

Individuals with ID have been noted to have a wide range of behavioral feeding challenges. This may include poor table manners, strong emotional responses elicited by the presentation of new foods, food selectivity (by type, texture, and/or presentation), mealtime problem behaviors (e.g., aggression, self-injury, batting at utensils, or other disruptive behaviors), food refusal, rituals regarding food preparation/presentation, rumination or vomiting, pica (see Chap. 28), abnormal eating pace (i.e., eating too quickly or too slowly), and/or overeating (Fodstad & Matson, 2008; Ledford & Gast, 2006; Schreck, Williams, & Smith, 2004). Due to these feeding problems, these individuals are less likely to eat the family diet (Collins et al., 2003) and are at risk for eating less than 20 total foods (Cornish, 1998). Many individuals with ID who struggle with eating may be diagnosed with a behavioral feeding disorder such as avoidant/restrictive food intake disorder (ARFID). Diagnostic and Statistical Manual of Mental Disorders, Fifth Edition (DSM-5), defines ARFID as a feeding disturbance that results in one or more of the following: significant weight loss, significant nutritional deficiency, dependence on enteral feeding or oral nutrition supplements, or marked interference with psychosocial functioning (American Psychiatric Association, 2013, p. 334).

Food selectivity and feeding skill problems both contribute to the development of significant behavioral feeding problems. While feeding skill problems can be taught, food selectivity is often more difficult to address. Food selectivity is often by type or by texture (Field, Garland, & Williams, 2003). The common consequence of food selectivity is significant food refusal, resulting in a diet limited in its nutritional value. Typically, food refusal is not a complete refusal to eat anything but rather results in an extremely limited diet. Similarly, this type of food refusal is more significant than "picky eating," as this term is typically used to describe food selectivity in a mild form that does not result in any nutritional or psychosocial disturbance and is more consistent with feeding problems in typically developing individuals.

An individual with ID will more typically display selectivity based on food brand (e.g., only McDonald's chicken nuggets, only Kraft Macaroni and Cheese), temperature (e.g., will refuse to eat frozen foods), smell, or texture (e.g., will not eat slimy foods). This may also include selectivity based on the presentation of the food, such as refusing to eat if someone different feeds them, if the "look" of the food environment changes (e.g., sitting in a different seat at the table), or if the "look" of the food changes (e.g., sandwich cut in halves instead of fourths, food touching on the plate). For most individuals, it is a combination of these factors that result in significant restriction of food intake (Hubbard, Anderson, Curtin, Must, & Bandini, 2014; Kozlowski, Matson, Fodstad, & Moree, 2011; Schreck et al., 2004). Unfortunately, insufficient food intake due to behavioral feeding challenges may result in the need for medical intervention discussed earlier in this chapter, including the possibility of feeding tubes or nasogastric or gastronomy tubes (Manno, Fox, Eicher, & Kerwin, 2005). While this intervention increases an individual's food intake, it does not address the behavioral nature of the feeding problem and, thus, may not solve this problem for the long term.

An additional behavioral deficit may be an actual deficit in feeding skills. For example, Page and Bouchert (1998) found that 79% of their sample of children displayed oral-motor deficits, 55% had fine motor skills deficits, and 17% had impairments in gross motor skills. Delays in utensil use and overall oral-motor skill development can contribute to the developmental of feeding problems. Many children with ID are delayed in the development of their chewing skills (Collins et al., 2003), which can impact the pacing or rate of a meal (e.g., eating too fast or slow) and can increase the risk of choking or aspirating. A history of choking or aspiration can contribute to the development of significant feeding problems due to increased anxiety around eating in the future (Nicholls & Bryant-Waugh, 2009).

Behavioral Feeding Assessment

Assuming that medical and/or physiological concerns have been ruled out, feeding problems may be behavioral in nature and can be assessed using behavioral assessment techniques. From this viewpoint, appropriate feeding can be understood as a behavior shaped by systematic teaching. Factors such as food type, individual behavior, caregiver behavior, type of challenging behavior (e.g., verbal or physical), and level of severity of behavior are all considered (Kleinert, 2017). Strategies to address targeted feeding behaviors are dependent on the function of the problematic feeding behavior (e.g., tangible, escape, attention, or automatic) but may include systematic presentation of food, positive reinforcement, and escape extinction. The function of problematic feeding behavior is often first determined through a functional behavior assessment, caregiver interview and/or log, or screening tools or assessments.

A first step in the assessment of feeding problems may be through an informal, clinical observation of a mealtime that involves both the individual and the primary caregiver. This observation can be used to determine both antecedent and consequent factors that may be affecting the feeding behaviors. Preferred and non-preferred foods are presented while the clinician records specific mealtime behaviors, including foods accepted or refused, the individual's behavior, and caregiver responses (Silverman, 2015). A functional behavior assessment (FBA), on the other hand, is a formalized, evidence-based, clinician-led process of observing and collecting data on an individual's behaviors and interactions with others. It is used to determine the relationship between a behavior and the context in which that behavior occurs (O'Neill, Horner, Albin, Storey, & Sprague, 1997). The FBA is a means of identifying the reason or function of such behaviors, as well as the factors maintaining relevant behaviors. Such an assessment then facilitates the development of tailored interventions. Antecedent interventions involve the elimination or manipulation of environmental variables that may interfere with feeding compliance, whereas consequence-based strategies involve caregiver responses to the behaviors that impact or interfere with feeding. Often, challenging feeding behaviors serve the function of "escaping" the mealtime or the demand of consuming the target food, though other sensory and physiologically-related concerns should be considered.

Another helpful tool that can be easily accessed by caregivers is a food log or journal, which is used to document information regarding the individual's feeding habits, including time of meals, duration of meals, foods accepted or rejected, and quantity of food ingested. Additional information may also include any notes documenting external factors that may have impacted the mealtime (e.g., distractions in the home, siblings, media or television, etc.). Best practice suggests a data collection period of consecutive days lasting 1–2 weeks at the onset of treatment. This information is a useful component of the FBA and provides information that is often forgotten or is difficult to witness in the clinical setting (Harvey, Bryant-Waugh, Watkins, & Meyer, 2015).

Caregiver-led data collection is an essential component of most behavioral feeding programs because it provides a valuable snapshot of the individual's behavior and the common factors that impact feeding in the most common mealtime environment, the home. Data collection in the clinic setting is also essential. Many screening tools and instruments are available and have been found valuable across disciplines. One such screening tool found beneficial within the field of psychology is the *Screening Tool of Feeding Problems* (STEP; Matson & Kuhn, 2001), which is a brief screener to identify feeding and mealtime behavior problems in individuals with intellectual disabilities. This instrument and other such screening and measurement tools are used to collect baseline data about the target feeding behavior and can be used to track progress or setbacks overtime. Such screening tools are particularly beneficial when measuring treatment outcomes.

The Utility of Interdisciplinary Assessment

Due to the complexity of factors that may affect feeding challenges in individuals with ID, one key to assessing and treating these challenges is the coordination of clinicians from multiple disciplines (Miller et al., 2001). There are many advantages to the team approach. One advantage includes saving the individual and family from multiple trips to meet with different providers. In addition, an interdisciplinary approach increases efficiency for providers due to shared evaluation, planning, and documentation and can, therefore, overall improve the delivery of care. Disciplines that may be included in interdisciplinary feeding assessment include nutrition or dietetics, occupational therapy, speech-language pathology, pediatrics, psychology, gastroenterology, and/or social work (see Table 22.2 for a list of relevant tasks based on provider).

The work of an interdisciplinary feeding team evaluation helps the family or caregiver team to develop achievable and appropriate goals for the individual in one visit (Silverman, 2010). For example, a dietician or nutritionist may be able to evaluate nutritional concerns by examining growth and nutrient intake and identifying other nutrition concerns. A specialized speech and language pathologist or occupational therapist is able to assess oral-motor feeding skills and can consider the extent in which textures, temperature, food volume, and/or sensory processing difficulties are contributing to feeding difficulties. A psychologist can help to identify and assess behavioral causes for difficulties related to comorbid diagnoses, developmental delays, inappropriate mealtime interactions, learned feeding avoidance or refusal, and/or family and cultural expectations. Intensive interdisciplinary feeding programs for individuals with intellectual and developmental disabilities have been shown to be effective in improving feeding behaviors, reducing caregiver stress, and increasing weight and caloric intake (Laud, Girolami, Boscoe, & Gulotta, 2009; Greer, Gulotta, Masler, & Laud, 2007). Individuals participating in an interdisciplinary feeding program benefit from receiving a comprehensive approach that can address the multifaceted challenges of feeding problems in individuals with intellectual and developmental disabilities.

Table 22.2 Roles and responsibilities of interdisciplinary feeding team members

Provider	Responsibilities
Gastroenterologist	Assess, treat, and monitor GI medical problems
Pediatrician	Assess developmental status based on age norms; assess, treat, and monitor general medical problems
Nutritionist or Dietician	Assess nutritional status, recommend calories, educate caregivers in nutrition
Psychologist	Assess behaviors interfering with feeding, develop and implement behavioral intervention, and conduct caregiver training
Occupational Therapist/Speech-Language Pathologist	Assess and treat oral-motor, fine motor, and sensory factors; assess and modify seating; perform barium swallow study
Social Worker	Assess family functioning and resources, secure services, case management

Conclusion

Feeding problems are a common concern among individuals with ID, and research has shown that these issues are often due to and compounded by a number of complex and overlapping factors. Physiological and medically complicated factors, such as GERD and dysphagia, are associated with feeding problems, and these conditions are more common among this population. When indicated, referrals should be made to appropriate medical specialists for treatment. However, behaviorally-rooted feeding problems, including over-selectivity and avoidant/restrictive feeding patterns, are among other common concerns and often require intensive behavioral intervention.

Before an intervention can be tailored to the individual's specific needs, an assessment should occur and, when possible, should include both

direct observation of the individual during mealtime and caregiver report and input. Psychologists, speech pathologists, and occupational therapists are among those trained in behavioral intervention for feeding problems. In most recent years, research has begun to suggest that some of the most effective clinical assessments and interventions are conducted by an interdisciplinary team, including other disciplines such as pediatrics, nutrition, gastroenterology, and social work. Through this method, physiological, medical, behavioral, and environmental factors can all be considered in the assessment and treatment planning process. As we strive to provide the best standard of care to all individuals with ID, the use of interdisciplinary teams is becoming increasingly advantageous in the assessment of many other comorbid concerns and has particular utility in monitoring and assessing concerns across the life span.

References

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

Arvedson, J. C., & Lefton-Greif, M. A. (2007). Ethical and legal challenges in feeding and swallowing intervention for infants and children. *Seminars in Speech and Language, 28*(3), 232–238.

Babbitt, R. L., Hoch, T. A., Coe, D. A., Cataldo, M. F., Kelly, K. J., Stackhouse, C., & Perman, J. A. (1994). Behavioral assessment and treatment of pediatric feeding disorders. *Journal of Developmental & Behavioral Pediatrics, 15*, 278–291.

Bernstein, I. L. (1999). Taste aversion learning: A contemporary perspective. *Nutrition, 15*, 229–234.

Birch, L. L. (1999). Development of food preferences. *Annual Review of Nutrition, 19*, 41–62.

Böhmer, C. J. M., Niezen-de Boer, M. C., Klinkenberg-Knol, E. C., Devillé, W. L. J. M., Nadorp, J. H. S. M., & Meuwissen, S. G. M. (1999). The prevalence of gastroesophageal reflux disease in institutionalized intellectually disabled individuals. *The American Journal of Gastroenterology, 94*(3), 804.

Bryant-Waugh, R. (2013). Feeding and eating disorders in children. *Current Opinion in Psychiatry, 26*, 537–542.

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 1), S1–S18.

Cermak, S. A., Curtin, C., & Bandini, L. G. (2010). Food selectivity and sensory sensitivity in children with autism spectrum disorders. *Journal of the American Dietetic Association, 110*(2), 238–246.

Cornish, E. (1998). A balanced approach towards healthy eating in autism. *Journal of Human Nutrition and Dietetics, 11*, 501–509.

Collins, M. S., Kyle, R., Smith, S., Laverty, A., Roberts, S., & Eaton Evans, J. (2003). Coping with the unusual family diet: Eating behaviour and food choices of children with Down's syndrome, autistic spectrum disorders or cri du chat syndrome and comparison groups of siblings. *Journal of Learning Disabilities, 7*, 137–155.

Field, D., Garland, M., & Williams, K. (2003). Correlates of specific childhood feeding problems. *Journal of Pediatrics and Child Health, 39*, 299–304.

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*, 541–550.

Forsyth, B. W. C., Leventhal, J. M., & McCarthy, P. L. (1985). Mothers' perceptions of feeding problems and crying behaviors: A prospective study. *American Journal of Diseases of Children, 139*, 269–272.

Harvey, L., Bryant-Waugh, R., Watkins, B., & Meyer, C. (2015). Parental perceptions of childhood feeding problems. *Journal of Child Healthcare, 19*(3), 392–401.

Gravestock, S. (2000). Eating disorders in adults with intellectual disability. *Journal of Intellectual Disability Research, 44*, 625–637.

Greer, A. J., Gulotta, C. S., Masler, E. A., & Laud, R. B. (2007). Caregiver stress and outcomes of children with pediatric feeding disorders treated in an intensive interdisciplinary program. *Journal of Pediatric Psychology, 33*(6), 612–620.

Hubbard, K. L., Anderson, S. E., Curtin, C., Must, A., & Bandini, L. G. (2014). A comparison of food refusal related to characteristics of food in children with autism Spectrum disorder and typically developing children. *Journal of the Academy of Nutrition and Dietetics, 114*(12), 1981–1987.

Johnson, C. R., Turner, K., Stewart, P. A., Schmidt, B., Shui, A., Macklin, E., & Hyman, S. L. (2014). Relationships between feeding problems, behavioral characteristics and nutritional quality in children with ASD. *Journal of Autism and Developmental Disorders, 44*(9), 1–10.

Kleinert, J. O. (2017, April). Pediatric feeding disorders and severe developmental disabilities. In *Seminars in speech and language* (Vol. 38(02), pp. 116–125). Thieme Medical Publishers.

Kozlowski, A. M., Matson, J. L., Fodstad, J. C., & Moree, B. N. (2011). Feeding therapy in a child with autistic disorder: Sequential food presentation. *Clinical Case Studies, 10*, 236–246.

Kuruvilla, J., & Trewby, P. N. (1989). Gastro-oesophageal disorders in adults with severe mental impairment. *British Medical Journal, 299*(6691), 95.

Lane, A. E., Geraghty, M. E., Young, G. S., & Rostorfer, J. L. (2014). Problem eating behaviors in autism spectrum disorder are associated with suboptimal daily nutrient intake and taste/smell sensitivity. *ICAN: Infant, Child, & Adolescent Nutrition, 6*(3), 172–180.

Larson-Nath, C., & Biank, V. F. (2016). Clinical review of failure to thrive in pediatric patients. *Pediatric Annals, 45*(2), e46–e49.

Laud, R. B., Girolami, P. A., Boscoe, J. H., & Gulotta, C. S. (2009). Treatment outcomes for severe feeding problems in children with autism spectrum disorder. *Behavior Modification, 33*(5), 520–536.

Ledford, J. R., & Gast, D. L. (2006). Feeding problems in children with autism spectrum disorders: A review. *Focus on Autism and Other Developmental Disabilities, 21*, 153–166.

Lefton-Greif, M. A., & Arvedson, J. C. (2007). Pediatric feeding and swallowing disorders: State of health, population trends, and application of the international classification of functioning, disability, and health. *Seminars in Speech and Language, 28*(3), 161–165.

Linscheid, T. R. (1983). Eating problems in children. In C. E. Walker & M. C. Roberts (Eds.), *Handbook of clinical child psychology* (pp. 616–639). New York, NY: Wiley.

Manno, C. J., Fox, C., Eicher, P. S., & Kerwin, M. E. (2005). Early oral-motor interventions for pediatric feeding problems: What, when and how. *Journal of Early and Intensive Behavior Intervention, 2*, 145–159.

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, 22*, 165–172.

Matson, J. L., & Fodstad, J. C. (2009). The treatment of food selectivity and other feeding problems in children with autism spectrum disorders. *Research in Autism Spectrum Disorders, 3*(2), 455–461.

Miller, C. K., Burklow, K. A., Santoro, K., Kirby, E., Mason, D., & Rudolph, C. D. (2001). An interdisciplinary team approach to the management of pediatric feeding and swallowing disorders. *Children's Health Care, 30*(3), 201–218.

Nicholls, D., & Bryant-Waugh, R. (2009). Eating disorders of infancy and childhood: Definition, symptomatology, epidemiology, and comorbidity. *Child and Adolescent Psychiatric Clinics of North America, 18*, 17–30.

O'Brien, S., Repp, A. C., Williams, G. E., & Christophersen, E. R. (1991). Pediatric feeding disorders. *Behavior Modification, 15*(3), 394–418.

O'Neill, R. E., Horner, R., Albin, R., Storey, K., & Sprague, J. (1997). *Functional analysis of problem behavior: A practical assessment guide* (2nd ed.). Pacific Grove, CA: Brookes/Cole.

Ott, D. J. (1994). Gastroesophageal reflux: What is the role of barium studies? *American Journal of Roentgenology, 162*(3), 627–629.

Page, J., & Boucher, J. (1998). Motor impairments in children with autistic disorder. *Child Language Teaching and Therapy, 14*, 233–259.

Patti, M. G., Diener, U., Tamburini, A., Molena, D., & Way, L. W. (2001). Role of esophageal function tests in diagnosis of gastroesophageal reflux disease. *Digestive Diseases and Sciences, 46*(3), 597–602.

Rezaei, M., Rashedi, V., Gharib, M., & Lotfi, G. (2011). Prevalence of feeding problems in children with intellectual disability. *Iranian Rehabilitation Journal, 9*, 56–59.

Robertson, J., Chadwick, D., Baines, S., Emerson, E., & Hatton, C. (2017). Prevalence of dysphagia in people with intellectual disability: A systematic review. *Intellectual and Developmental Disabilities, 55*(6), 377–391.

Rommel, N., De Meyer, A. M., Feenstra, L., & Veereman-Wauters, G. (2003). The complexity of feeding problems in 700 infants and young children presenting to a tertiary care institution. *Journal of Pediatric Gastroenterology and Nutrition, 37*, 75–84.

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.

Schmitt, L., Heiss, C. J., & Campbell, E. E. (2008). A comparison of nutrient intake and eating behaviors of boys with and without autism. *Topics in Clinical Nutrition, 23*, 23–31.

Schwarz, S. M. (2003). Feeding disorders in children with developmental disabilities. *Infants & Young Children, 16*, 317–330.

Schwarz, S. M., Corredor, J., Fisher-Medina, J., Cohen, J., & Rabinowitz, S. (2001). Diagnosis and treatment of feeding disorders in children with developmental disabilities. *Pediatrics, 108*, 671–676.

Silverman, A. H. (2010). Interdisciplinary care for feeding problems in children. *Nutrition in Clinical Practice, 25*(2), 160–165.

Silverman, A. (2015). Behavioral Management of Feeding Disorders of childhood. *Annuals of Nutrition and Metabolism, 66*, 33–42.

Sisson, L. A., & Van Hasselt, V. B. (1989). Feeding disorders. In *Behavioral medicine and developmental disabilities* (pp. 45–73). New York, NY: Springer.

Wright, C. M., Smith, K. H., & Morrison, J. (2011). Withdrawing feeds from children on long term enteral feeding: Factors associated with success and failure. *Archives of Disease in Childhood, 96*, 433–439.

The Assessment of Sleep Disorders in Dually Diagnosed Individuals

23

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Introduction

Sleep has a complex and reciprocal relationship with neurodevelopment. As the brain matures across the life span, endogenous and environmental factors dictate sleep duration and patterns. Sleep-wake cycles begin in utero when fetal and maternal factors establish circadian rhythms at approximately 30 weeks (McKenna & Reiss, 2018). Infant sleep-wake cycles typically progress from polyphasic sleep, to the biphasic napping of the toddler, to a monophasic sleep by 4–5 years of age (Staton, Smith, Hurst, Pattinson, & Thorpe, 2017) with total sleep time decreasing to about 9 hours per night by puberty (Stores, 2014). The trend toward later sleep and wake times in adolescence is well documented (Paksarian, Rudolph, He, & Merikangas, 2015) and gives way to mature sleep in the young adult. With aging, sleep disturbances typically increase (Mander, Winer, & Walker, 2017; Skeldon, Derks, & Dijk, 2016).

The neurophysiology of wakefulness and sleep as reoccurring brain states provides a foundation for understanding sleep disorders (Aserinsky & Kleitman, 1953; Herice, Patel, & Sakata, 2018; Weber & Dan, 2016). Altered brain neurophysiology and/or anatomy may contribute to the well-documented increased incidence of sleep disturbances in persons with intellectual disability (Esbensen & Schwichtenberg, 2016; Fletcher, Barnhill, & McCarthy, 2016). A meta-analysis including 1377 individuals found poorer sleep quality and shorter sleep duration in persons with intellectual disability (Surtees, Oliver, Jones, Evans, & Richards, 2018). Night wakening and middle insomnia show a significant relationship to behavioral problems in children with autism spectrum disorder (ASD) (Mazurek & Sohl, 2016). A study of 107 youth with Down syndrome (DS) found 65% with significant sleep disturbances on screening, yet 66% of parents reported no sleep problems in their child (Hoffmire, Magyar, Connolly, Fernandez, & van Wijngaarden, 2014). The importance of sleep across the life span of persons with intellectual disability is receiving increasing attention in part due to the association of sleep disturbance with physical and mental illness (Fletcher et al., 2016; Watson et al., 2015). This chapter briefly reviews the science of sleep before discussing DSM-5 sleep disorders and the assessment of sleep disturbances in persons with intellectual disability.

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A Brief Review of Sleep Science

Through the ages, philosophers, scientists, psychologists, and physicians have theorized about the process of sleep. Prior to the modern era, the *passive process theory* prevailed, espousing the idea that the brain was at rest and inactive during sleep (Kryger, Roth, & Dement, 2017). The modern era of sleep science began with the physiological sleep studies of Piéron (Morrison, 2014), but it was Berger's invention of the EEG in 1924 and documentation of the differences in electrical activity between awake and asleep states that provided a basis for the technology still in use today for the assessment of sleep disturbances (Burger, 1929). Davis, Davis, Loomis, Harvey, and Hobart (1937) further defined the electrical activity as the brain wave patterns familiar in modern sleep studies. Rapid eye movement (REM) sleep was first described by Aserinsky and Kleitman (1953). REM was studied using electrooculograms (EOG) to measure eye motility while sleeping. The hypothesis that REM sleep represented a lightening of sleep or a dream state was confirmed when sleep subjects were awakened during REM and could recall vivid dreams, while participants awakened at non-REM (NREM) phases could not recall their dreams (Kryger et al., 2017). Aserinsky and Kleitman (1953) described the patterns of slow wave sleep followed by REM sleep which are repeated throughout the night in 90–100-minute cycles. Dement and Kleitman (1957) conducted studies over a full night and documented the sleep cycle. These early studies lead to the founding of sleep clinics and modern computerized polysomnography which includes EEG and EOG readings as well as measures of heart rhythm (ECG), muscle activity (EMG), and respiratory functions (Deak & Epstein, 2009).

Sleep Cycles

The work of Aserinsky and Kleitman (1953) informs the American Academy of Sleep Medicine (AASM) descriptions of sleep staging and sleep architecture. The AASM breaks NREM and REM sleep into stages. NREM sleep is further defined into three distinct stages, N1, N2, and N3. N1 is defined as light sleep, very similar to wakefulness with the exception that theta rhythms (5–8 Hz) predominate and alpha rhythms (8–14 Hz) are absent. N2 on EEG is notable for K-complexes or spikes and sinusoidal waveforms called spindles. N3 sleep is slow wave (0.5–2 Hz) or delta wave sleep. In contrast, REM sleep is markedly different, with muscle atonia and twitches, rapid eye movements, and an EEG with low amplitude waves similar to what is seen in an awake but drowsy individual (Kryger et al., 2017). Persons with intellectual disability have less REM sleep (Stores, 2014). Recent research suggests REM sleep is necessary for memory consolidation (Esposito & Carotenuto, 2013; Peever & Fuller, 2017). The normal sleep cycle consists of an alternating pattern of NREM, REM sleep, and arousals throughout the sleep period, with a cycle lasting between 70 and 90 minutes. Sleep cycles undergo change across the life span with NREM sleep stages established by 6 months (MacLean, Fitzgerald, & Waters, 2015). Over half of fetal and newborn sleep is REM (Stores, 2014) with REM sleep decreasing with age (Ohayon, Carskadon, Guilleminault, & Vitiello, 2004). The pattern of these cycles is called sleep architecture and is graphed as a hypnogram. The structure of a hypnogram varies across the life cycle and shows characteristic changes for certain sleep disorders, medications, and mental disorders. The hypnogram captures the electrical manifestation of a highly regulated, active physiological process with multiple brain regions and neurotransmitters playing a role (Kryger et al., 2017).

Neurotransmitters

Over a dozen neurotransmitters, neuropeptides, and hormones play a role in sleep-wakefulness regulation (Schwartz & Kilduff, 2015). Alteration in a single neurotransmitter may impact sleep-wake regulation. For example, a deficiency of hypocretin is a biomarker for narcolepsy (Holst & Landolt, 2018). The dysregula-

Table 23.1 Major neurotransmitters involved in sleep-wakefulness

Neurochemical	Source	Target	Promotes	Reference
Acetylcholine	Basal forebrain	Cortex	Wakefulness	Schwartz and Kilduff (2015)
Adenosine	Extracellular	Ventral forebrain	Sleep	Strecker et al. (2000)
Dopamine	Ventral tegmental area	Laterodorsal tegmental pedunculopontine nuclei	Wakefulness	Monti and Monti (2007)
	Substantia nigra pars compacta	Dorsal raphe nucleus, locus coeruleus, hypothalamus, basal forebrain, thalamus		
GABA	Ventral lateral preoptic area	Lateral hypothalamus	Sleep	Brady, Siegel, Albers, and Price (2012)
	Median preoptic area	Locus coeruleus, tuberomammillary nuclei, dorsal raphe nuclei		
Glutamate	Parabrachial nucleus, pedunculopontine tegmental nucleus	Basal forebrain, cortex	Wakefulness	Saper and Fuller (2017)
Histamine	Tuberomammillary nuclei	Prefrontal cortex, anterior hypothalamus	Wakefulness	Holst and Landolt (2018)
Hypocretin	Posterior lateral hypothalamus	Basal forebrain, tuberomammillary nuclei, dorsal raphe nuclei, locus coeruleus, laterodorsal tegmental/ pedunculopontine nuclei	Wakefulness	Schwartz and Kilduff (2015)
Norepinephrine	Locus coeruleus	Ventral lateral preoptic area, median preoptic area	Wakefulness	Stahl (2013)
Serotonin	Dorsal raphe nucleus	Cortex	Wakefulness	Stahl (2013)
Melatonin	Pineal gland	Suprachiasmatic nuclei	Sleep	Holst and Landolt (2018)

tion of many of these same neurotransmitters is implicated in mental disorders. While the neurochemistry of the sleep-wake cycles of persons with intellectual disability requires further research, specific neurotransmitter abnormalities have been associated with specific developmental disorders including Smith-Magenis syndrome and Norrie disease (Stores, 2014). Table 23.1 highlights the major chemical messengers and brain areas involved in the regulation of sleep and arousal.

Acetylcholine, first identified by Dale, Feldberg, and Vogt (1936) as the neurotransmitter responsible for signal transmission at the neuromuscular junction, has projections not only in the peripheral nervous system but also within the central nervous system. Acetylcholine promotes wakefulness by stimulating the cortex via neurons originating in the basal forebrain (Schwartz & Kilduff, 2015).

Adenosine has been shown to play a role in sleep-wakefulness. Adenosine is a breakdown product of ATP (adenosine triphosphate) and ADP (adenosine diphosphate), which power cellular metabolism. Extracellular adenosine levels rise during periods of wakefulness and decline during sleep. Adenosine helps to induce slow wave sleep by inhibiting wake-promoting neurons in the brain stem (Strecker et al., 2000).

Dopamine is associated with motor activity, motivation, and reward systems and is a target for psychotropic medications treating psychiatric disorders including schizophrenia, bipolar disorder, depression, tic disorders, and attention deficit hyperactivity disorder (ADHD) (Stahl, 2013). Dopamine plays a role in sleep-wakefulness, through projections (both efferent and afferent) connecting the ventral tegmental area (VTA) and the substantia nigra pars compacta (SNc) to the laterodorsal tegmental/pedunculopontine nuclei (LDT/PPT), dorsal raphe nucleus, locus coeruleus (LC), the hypothalamus, basal forebrain, and the thalamus (Monti & Monti, 2007). In general terms, dopamine activity promotes wakefulness,

although dopamine's effect is actually receptor dependent. Five distinct dopamine receptor types have been identified (D_1–D_5). Stimulation of D_1 and D_2 increases wakefulness, while D_3 activation decreases locomotion and possibly leads to sleep (Brady et al., 2012).

γ-Aminobutyric acid (GABA) is a major inhibitory neurotransmitter. GABA receptor activation decreases the likelihood of neuronal firing. Medications that target GABA receptors in the brain reduce seizure activity and anxiety, and promote sedation and sleep. GABAergic medications are also used for general anesthesia (Brady et al., 2012).

Glutamate, a nonessential amino acid, is an excitatory neurotransmitter. Glutamatergic projections from the parabrachial (PB) and the pedunculopontine tegmental nucleus (PPT) activate the basal forebrain which in turn activates the cortex (Saper & Fuller, 2017).

Histamine-producing neurons are located in the posterior hypothalamus in a region called the tuberomammillary nucleus (TMN). The TMN has projections to a variety of areas of the brain, including the prefrontal cortex and the anterior hypothalamus (Brady et al., 2012). Histamine activity is much greater during periods of wakefulness. Clinically, antihistamine medications that block histamine receptors (H_1) are sedating (Holst & Landolt, 2018).

Hypocretins, also referred to as orexins, are excitatory neuropeptides promoting wakefulness, located in the posterior lateral hypothalamus (PLH). Hypocretin-producing neurons have projections throughout the brain as well as the spinal cord, including other wakefulness-promoting regions such as the basal forebrain, tuberomammillary nucleus, dorsal raphe nucleus, locus coeruleus, and lateral dorsal tegmental nucleus/pedunculopontine tegmentum nuclei (Schwartz & Kilduff, 2015). Hypocretin neurons are inactive during both NREM and REM sleep. Hypocretins are of particular interest as they have been implicated as a possible cause of narcolepsy (Holst & Landolt, 2018).

Norepinephrine, an excitatory neurotransmitter, is primarily produced by the neurons of the locus coeruleus, a component of the ascending reticular activating system (ARAS). Norepinephrine promotes wakefulness and attention. Medications which raise the extracellular levels of norepinephrine are used to treat depression (Stahl, 2013). Norepinephrine may play a role REM sleep induction although this theory has not been confirmed (Brady et al., 2012).

Serotonin (5-HT) is produced by neurons of the dorsal raphe nuclei (DRN). Serotonin's role in sleep-wakefulness is complex, as the DRN projects to a multitude of regions in the brain and has multiple receptor types. Currently, serotonin is thought to promote wakefulness and to inhibit REM sleep (Holst & Landolt, 2018). Serotonergic medications which increase extracellular levels of serotonin or target serotonin receptors in the brain are used to treat a variety of disorders including schizophrenia, depression, and anxiety (Stahl, 2013).

Melatonin, a hormone produced by the pineal gland, plays an important role in the entrainment of the sleep-wake cycle to dark-light signals. Melatonin levels are ten times higher during periods of darkness than in light. The suprachiasmatic nuclei (SCN), the location of the circadian rhythm clock in the brain, has numerous melatonin receptors (Holst & Landolt, 2018). The actual mechanism of how melatonin impacts the SCN is unknown, but one proposed theory is that melatonin activity attenuates the SCN alerting signals, thus allowing the sleep-promoting signals to induce sleep (Stahl, 2013). Exogenous melatonin, available as an over-the-counter medication, is effective in inducing sleep in humans. In addition, a melatonin receptor agonist has been approved for insomnia, and another has been effective in resynchronizing circadian rhythms (Holst & Landolt, 2018).

Neuroanatomy of Sleep

Structures known to be involved in NREM sleep include the ventral lateral preoptic area (VPLO), median preoptic area (MnPO), basal forebrain, midbrain, raphe nucleus, and thalamus (Schwartz & Kilduff, 2015) (Fig. 23.1).

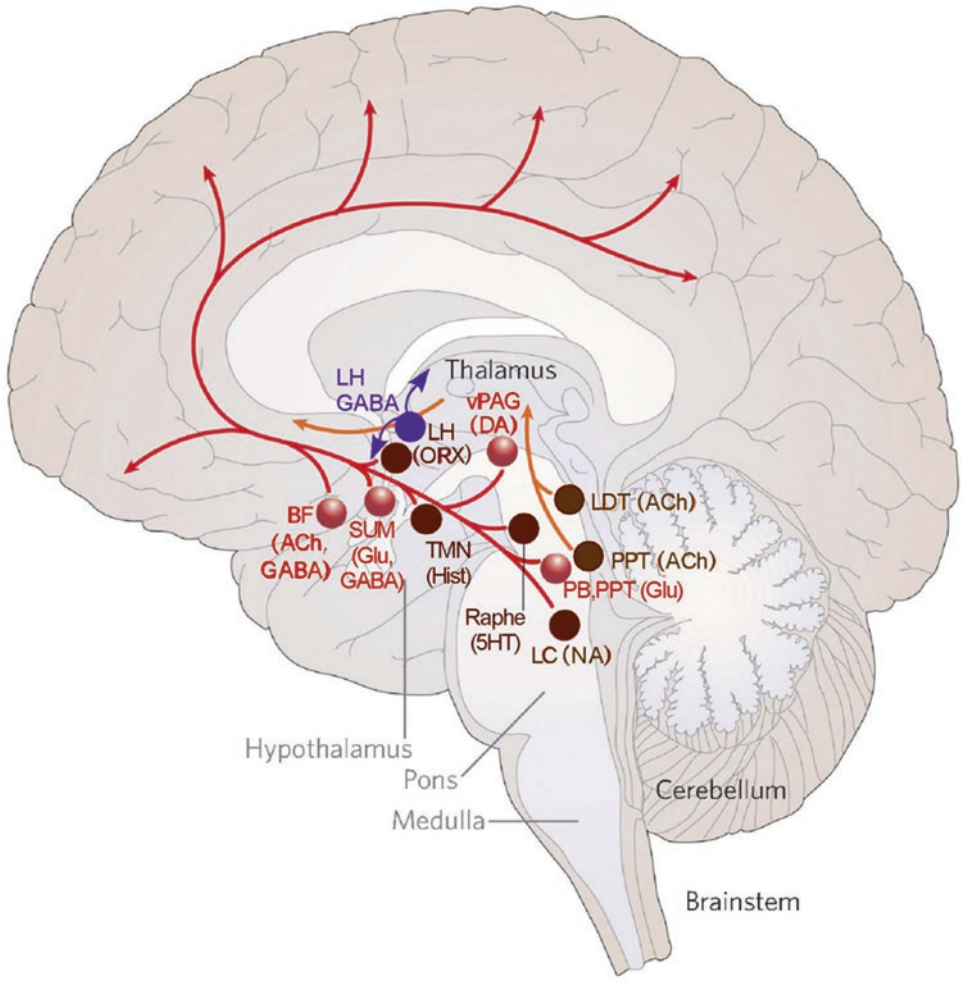

Fig. 23.1 Primary areas of the brain involved in sleep-wakefulness. (Figure from Saper and Fuller (2017). Used with permission)

The VLPO and MnPO are located in the anterior hypothalamus and, through their GABAergic projections, inhibit the cortex indirectly through the inhibition of cortical activating neurons in the lateral hypothalamus, locus coeruleus, tuberomammillary nuclei, and the dorsal raphe nuclei. During periods of wakefulness, the VLPO and MnPO are themselves inhibited by norandrenergic projections from the ascending reticular activating system (ARAS), which is a collection of nuclei located in the brain stem (Schwartz & Kilduff, 2015).

The basal forebrain, a major source of acetylcholine, promotes wakefulness and REM sleep with projections extending throughout the cortex. Inhibition of the basal forebrain resulting in slow wave sleep occurs indirectly through the inhibition of the parabrachial nucleus and the lateral hypothalamus which, unless inhibited, stimulate the basal forebrain (Schwartz & Kilduff, 2015).

The thalamus, specifically the reticular nucleus, is responsible for generating bursts of spindles (7–12 Hz) seen on EEG during N2 NREM sleep. During periods of wakefulness and REM sleep, the reticular neurons operate tonically (continuously); the burst activity (spindles) is inhibited by the locus coeruleus (norepinephrine) and the dorsal raphe nuclei (serotonin) (Brown, Basheer, McKenna, Strecker, & McCarley, 2012).

REM sleep is generated by the dorsolateral pons, through acetylcholine projections in the laterodorsal tegmental nucleus (LDT) and the pedunculopontine tegmentum nuclei (PPT) to glutamatergic pontine reticular formation areas in the brain stem. This activity ultimately results in the manifestations of REM sleep including theta activity, REM sleep, and muscle atonia as well as the synchronized electrical activity in the pons, lateral geniculate nucleus, and the occipital cortex (PGO Waves) (Brown et al., 2012).

McCarley and Hobson proposed a model of NREM/REM sleep control in which REM-on cells in the LDT/PPT not only generate REM sleep but inhibit REM-off cells by exciting GABAergic interneurons located adjacent to them. REM sleep ends when REM-on cells start to excite REM-off cells (Brown et al., 2012).

Sleep Homeostasis and Circadian Rhythms

Sleep is controlled through regulation of intensity and timing. The neurochemical physiological or homeostatic process (Process S) regulates sleep intensity, and the circadian system (Process C) regulates the timing of sleep. These processes are interrelated and may be intentionally overridden. When Process S is at a minimum, it triggers awakening; at its maximum level, it triggers sleep, if Process C is in the correct circadian phase (Borbely, Daan, Wirz-Justice, & Deboer, 2016; Schwartz & Kilduff, 2015). Process S gauges the body's need for sleep versus arousal. It is conceptualized as a *sleep debt* which increases during periods of wakefulness and declines during sleep (Borbely et al., 2016). Process S is measured by examining EEG delta waves (0.5–4.0 Hz). The delta wave activity is a direct result of the individual's prior periods of sleep-wakefulness (Schwartz & Kilduff, 2015).

Process C is regulated by the circadian pace maker, commonly called the biological clock, located in the hypothalamic suprachiasmatic nuclei (SCN). The SCN coordinates input from multiple neural pathways and sends signals to the thalamus, hypothalamus, and epithalamus to regulate peripheral clocks, ultimately leading to the rhythm of behaviors such as food intake, activity level, and body temperature (Rosenwasser & Turek, 2015). The major contributor of SCN input is the retinohypothalamic tract (RHT). The RHT originates with specialized retinal ganglion cells. Interestingly, these retinal cells are not responsible for vision but respond to the daily light-dark cycle entraining the SCN to express the gene identified as *Per* (PERIOD). The expression of *Per* combined with signals from other brain regions regulates the circadian cycle (Rosenwasser & Turek, 2015). Circadian rhythms are established before birth and stabilize in childhood before resetting during the teen years and restabilizing in the young adult. As individuals age, the timing of circadian rhythms for temperature, melatonin, and cortisol levels advance (occur earlier) which is reflected in the earlier sleep times common in the elderly (Duffy, Zitting, & Chinoy, 2015). Circadian rhythm sleep-wake disorders (CRSWD) occur when Process C (circadian pace maker) is not functioning properly or there is poor synchronization between Process C and the external 24-hour environment. Sollars and Pickard (2015) have written a comprehensive exploration of circadian rhythm neurobiology. Chronobiology studies the effects of exposure to light on the circadian system. Chronomedicine, the application of chronobiology to the treatment of disease, has implications for CRSWD as well as diabetes, asthma, heart disease, and endocrine disorders (Cortelli, 2015).

The American Academy of Sleep Medicine has issued consensus statements with sleep recommendations for sleep time, decreasing as children age (Paruthi et al., 2016), to at least 7 hours for adults and a slight increase after 60 (Watson et al., 2015). Failure to maintain necessary sleep time and the resulting behavioral issues are common challenges in persons with intellectual disability (Stores, 2014). Given the complexity of sleep regulation, the number of brain structures involved, and many environmental factors influencing sleep, the association of sleep disturbances with most mental disorders and the high rate of sleep disturbances in persons with intellectual disability is not surprising. It is this robust asso-

ciation that makes knowledge of sleep disorders and the assessment of sleep during each patient contact critical in clinical practice.

Sleep Disorders

Classification Systems for Sleep Disorders

While mental health clinicians are familiar with the *Diagnostic and Statistical Manual of the American Psychiatric Association* (APA, 2013) and the World Health Organization *International Classification of Diseases* (WHO, 2007), the leading classification system for sleep disorders is the American Academy of Sleep Medicine *International Classification of Sleep Disorders* (ICSD), currently in its third edition (AASM, 2014). The AASM provides resources for the public at sleepeducation.org. The application of DSM criteria to the diagnosis of sleep disorders in persons with intellectual disability is explored in the *Diagnostic Manual-Intellectual Disability: A Textbook of Diagnosis of Mental Disorders in Persons with Intellectual Disability* (DM-ID 2) (Fletcher et al., 2016). The DM-ID 2 provides a useful overview of sleep disorders in persons with intellectual disability but does not recommend significant modifications to DSM-5 criteria. In addition, the National Institutes of Mental Health *Research Domain Criteria* (RDoC) offers a transdiagnostic framework designed to promote comparison of domain characteristics within and between DSM/ICD categories. The RDoC does not offer diagnostic guidance, but the domain, *Arousal and Regulatory Systems*, offers a research framework for arousal, circadian rhythms and sleep-wakefulness (Cuthbert, 2014). The *Diagnostic Classification of Mental Health and Developmental Disorders of Infancy and Early Childhood* (DC:0–5) provides diagnostic guidance for children under six (ZERO TO THREE, 2016). (See Chap. 2 "Diagnostic Systems" for further details on classification systems.)

The ICSD and ICD categorize sleep disorders under insomnia, sleep-related breathing disorders, central disorders of hypersomnolence, circadian rhythm sleep-wake disorders, parasomnias, sleep-related movement disorders, and other sleep disorders with 60 diagnoses described in the ICSD 3 (AASM, 2014; WHO, 2007). The DSM-5 includes 11 diagnostic groups (APA, 2013). Research has informed revisions of the ICSD, DSM, and ICD with a goal of identifying the underlying pathophysiology of sleep disorders (Thorpy, 2012). The description of sleep disorders in this chapter will follow DSM-5 nosology. In addition to the specific sleep disorders discussed here, the DSM-5 allows the modifiers *other specified* or *unspecified* for insomnia disorder, hypersomnolence disorder, and sleep-wake disorder.

Insomnia Disorder

Insomnia disorder is the most common sleep disorder among individuals with intellectual or developmental disabilities and among the general population (Barnhill, Soda, Poindexter, & Hollway, 2016). While insomnia is commonly used broadly to connotate difficulty sleeping, the diagnosis and treatment of insomnia disorder can be challenging with the differential diagnosis including other sleep disorders as well as medical and mental illness. The DSM-5 defines insomnia disorder as

> a predominant complaint of dissatisfaction with either sleep quantity or sleep quality present despite adequate opportunity for sleep. (p. 362, APA, 2013)

This complaint may reflect trouble falling asleep (initial insomnia), staying asleep (middle insomnia), or waking too early (early morning awakening). The diagnostic criteria include duration requirements, three nights per weeks for 3 months, and the presence of significant distress or impairment. Specifiers are listed for comorbid mental, medical, or other sleep disorders and duration – episodic, persistent, or recurrent (APA, 2013). The DM-ID-2 does not offer modification of DSM-5 criteria for persons with intellectual disability (Fletcher et al., 2016). The DC:0–5 offers guidance for the diagnosis of insomnia in

young children with the diagnoses of sleep onset disorder and night waking disorder (pp. 92–93, ZERO TO THREE, 2016).

The differential diagnosis of insomnia is wide ranging. Physiological, behavioral, and environmental factors must be considered. Physiological causes of insomnia, indicated by the specifier *other medical comorbidity* (p. 362, APA, 2013), may include genetic disorders, pain, gastrointestinal disturbances, epilepsy, and heart disease; the specifier *nonsleep disorder mental comorbidity* (p. 362, APA, 2013) applies to insomnia with depression, schizophrenia, substance caffeine or nicotine use, or other mental disorder (Esbensen & Schwichtenberg, 2016). In addition, some individuals are physiological short sleepers (5–6 hours per night) or very short sleepers (less than 5 hours per night). This may reflect a genetic variation of clock genes which govern sleep duration and homeostasis (Pellegrino et al., 2014). While behavioral insomnia is not a DSM-5 diagnosis, it a common presenting complaint in clinical practice. Behavioral causes of insomnia include bedtime resistance due to inability to calm, anxiety, fears, or other factors (Esbensen & Schwichtenberg, 2016). Behavioral challenges are the most frequent sleep concern expressed by the caregivers of children with intellectual disability (Köse, Yılmaz, Ocakoğlu, & Özbaran, 2017). Behavioral challenges may be precipitated by environmental factors such as the use of television or electronic equipment at bedtime as well as lights, noise, and temperature variations which may be especially disturbing to persons with sensory issues. Blue spectrum lighting, found in electronics, LEDs, and compact fluorescent bulbs, increases alertness and can disrupt circadian rhythms (Tosini, Ferguson, & Tsubota, 2016).

The US Centers for Disease Control National Health Interview Survey for the years 2002 and 2012 reported an unadjusted prevalence of insomnia in adults as 17.5% and 19.2%, respectively (Ford, Cunningham, Giles, & Croft, 2015). In a longitudinal mailed survey study of 2363 individuals conducted in the UK, 37% reported symptoms consistent with insomnia with older individuals and those with anxiety, depression, or pain more likely to endorse symptoms of insomnia (Morphy et al., 2007). In a study of primary care patients with 41% reporting difficulty sleeping, primary insomnia was diagnosed in 12%, obstructive sleep apnea (OSA) in 9%, and delayed sleep phase in 2%. Chronic medical conditions were high (43%) as were depression (50%) and anxiety (43%) (Arroll et al., 2012). A cross-sectional study of 700 children found 19% with insomnia symptoms with a greater prevalence in girls aged 11–12 (Calhoun et al., 2014). The CDC Youth Risk Behavior Survey of over 67,000 high school students conducted between 2007 and 2015 found less than 6 hours sleep a day was associated with suicide and risk-taking behaviors (Weaver et al., 2018).

Difficulty initiating sleep, maintaining sleep, and reduced sleep duration are the most prevalent sleep problems among children with ASD (Grigg-Damberger & Rawls, 2013; Miano et al., 2007), with insomnia being the most frequent sleep disorder (Köse et al., 2017). Mayes and Calhoun (2009) report that as many as 60% of children with autism spectrum disorder experience difficulty falling asleep. Sleep disorders may exacerbate daytime behavior problems such as mood instability, aggression, and self-injury in children with ASD (Blackmer & Feinstein, 2016; Ming, Gordon, Kang, & Wagner, 2008). Insomnia in persons with ASD does not necessarily resolve in adulthood. Baker and Richdale (2015) conducted a cross-sectional study comparing sleep patterns and sleep problems and their relationship with daytime functioning in 36 adults diagnosed with autism spectrum disorder and no comorbid intellectual disability (high-functioning autism spectrum disorder) to 36 neurotypical adults and found that the adults with autism spectrum disorder had significantly more general sleep disturbances, longer sleep onset latencies, and poorer sleep efficiency. Clinicians should be aware that some neurodevelopmental disabilities may increase the risk for early-onset dementia and that these individuals are at risk for a range of sleep disorders including behavioral insomnia stemming from impulsivity, aggression, and confusion with the onset and progres-

sion of dementia. For example, by the age of 40 years, virtually all persons with Down syndrome have neuropathological markers of dementia and experience a decline in cognitive function with age which may trigger or worsen sleep disturbances (Hill et al., 2016). The diagnosis of behavioral insomnia requires ruling out sleep disorders or medical conditions as an etiology for the disturbance.

Hypersomnolence Disorder

The DSM-5 defines hypersomnolence disorder as excessive daytime sleepiness despite adequate duration of sleep (at least 7 hours) if one of the following is present: additional sleep periods, more than 9 hours of sleep that is not experienced as restorative, or not feeling alert on awakening, sometimes called *sleep inertia*. Duration (three times a week for at least 3 months), distress/impairment, and exclusionary criteria apply as well. Specifiers include comorbidities, duration, and severity (APA, 2013). The DM-ID-2 does not offer modification of DSM-5 criteria for persons with intellectual disability (Fletcher et al., 2016). The ICSD corresponding diagnosis is idiopathic hypersomnolence disorder (AASM, 2014). A systematic review reported that complaints of daytime sleepiness are common but hypersomnolence disorder is rare and thought to occur in less than 1% of the population although persons with intellectual disability are at increased risk (Sowa, 2016). Persons with Kleine-Levin syndrome and *PURA*-related neurodevelopmental disorders may experience hypersomnia (Kotagal, 2018; Reijnders et al., 2017). GABA appears to be involved, but the etiology of hypersomnolence disorder is unknown (Kotagal, 2018). About a third of patients report similar issues in a family member (Trotti, 2017). Objective observation of sleep and multiple sleep latency tests (MSLT) aid in the diagnosis of hypersomnolence disorder and assist in ruling out other sleep disorders (Sowa, 2016; Trotti, 2017). A careful history is necessary to identify health, head injury, or medications as the cause of excessive sleepiness (Billiard & Sonka, 2016).

Narcolepsy

A second disorder of excessive sleepiness, narcolepsy, is characterized by an uncontrollable urge to sleep. In addition, cataplexy, hypocretin deficiency, or characteristic REM latency on sleep study must be present. Duration (three times a week for at least 3 months), distress/impairment, and exclusionary criteria apply as well. Five types of narcolepsy are specified by the DSM-5 (APA, 2013). The ICSD 3 classifies narcolepsy as type 1 (with cataplexy) or type 2 (without cataplexy), with type 1 requiring characteristic PSG and MSLT and/or decreased CSF hypocretin-1 concentration and type 2 requiring characteristic PSG and MSLT and increased CSF hypocretin-1 concentration, if measured (AASM, 2014).

The DSM-5 (2013) defines cataplexy as:

> Episodes of bilateral loss of muscle tone resulting in the individual collapsing, often occurring in association with intense emotions such as laughter, anger, fear, or surprise. (p. 818)

The onset of cataplexy may occur months or years after the initial presentation of excessive daytime sleepiness (Ruoff & Rye, 2016). In persons with developmental disabilities and poor coordination or hypotonia, cataplexy can be more challenging to identify. In children, early cataplexy may begin as a complex movement disorder with an associated "cataplectic facies associated with hypotonia, ptosis, tongue protrusion or unsteady gait" (p. 3481) (Plazzi et al., 2011; Rocca, Pizza, Ricci, & Plazzi, 2015). Although not diagnostic, nonpsychotic hypnogogic (at sleep onset) and hypnopompic (on waking) hallucinations and the belief that events from dreams are real (dream delusions) are associated with narcolepsy (Wamsley et al., 2014). Narcolepsy sleep spells are typically less than 20 minutes although sedation may be increased when individuals are sedentary or living in environments with limited stimulation (Barnhill et al., 2016). Ruoff and Rye (2016) offer suggestions for assessment with comprehensive evaluation requiring referral to a sleep specialist.

Narcolepsy-cataplexy affects less than 0.05% of the general population with onset of narco-

lepsy typically between the ages of 15 and 25 (Kumar & Sagili, 2014). Sedky, Bennett, and Pumariega (2014) reviewed six studies that assessed children with Prader-Willi syndrome (total of 42 affected children) using multiple sleep latency testing and/or screening for cataplexy symptoms and found that the prevalence of signs/symptoms suggestive of narcolepsy was 35.71%. A case series by Prihodova, Dudova, Mohaplova, Hrdlicka, and Nevsimalova (2018) identified four children with narcolepsy type 1 and Asperger's syndrome and three for whom Asperger's syndrome was suspected or diagnosed at the time of onset of narcolepsy type 1, suggesting that children with autism spectrum disorder may be at increased risk for narcolepsy. Narcolepsy can persist into adulthood in persons with neurodevelopmental disabilities and may require special accommodations in work or group living settings (Nevsimalova, 2014).

Liblau, Vassalli, Seifinejad, and Tafti (2015) demonstrated a hypocretin signaling deficiency in type 1 narcolepsy. In addition, complex genetic and autoimmune reactions are thought to contribute to the onset of narcolepsy which may be triggered by infection, pesticides, heavy metals, or other environmental factors (Kumar & Sagili, 2014). An association between narcolepsy and the H1N1 vaccinations or infections has been suggested (Han et al., 2011).

Breathing-Related Sleep Disorders

The DSM-5 recognizes three sleep-related breathing disorders: obstructive sleep apnea (OSA), central sleep apnea (CSA), and sleep-related hypoventilation. Apnea indicates a complete lack of airflow; hypopnea, a reduced airflow; and hypoventilation, slowed respiration. The breathing-related sleep disorders are diagnosed by disorder-specific characteristics on polysomnography (APA, 2013). Referral to a sleep specialist for objective evaluations is indicated if sleep-disordered breathing is suspected.

In OSA, the tongue relaxes occluding the upper airway leading to apneic episodes. The craniofacial abnormalities, poor muscle tone, and obesity which are all risk factors for OSA are common in persons with developmental disabilities (Javaheri et al., 2017). Down, Apert, Crouzon, and Pfeiffer are all syndromes commonly associated with craniofacial abnormalities and OSA (Stores, 2014). Low muscle tone is associated with Down, Prader-Willi, and Joubert syndromes, muscular dystrophy (Harris, 2008), ASD (Serdarevic et al., 2017), cerebral palsy, and many other developmental disabilities. The increased risk for obesity in persons with developmental disabilities is well documented (Bandini et al., 2015). The prevalence of obstructive sleep apnea is seen in 1–4% of the general pediatric population but increases to 30–70% among individuals with Down syndrome and nearly 80% in children with Prader-Willi (Bassell, Phan, Leu, Kronk, & Visootsak, 2015; Rosen, 2011; Sedky et al., 2014). Based on a meta-analysis involving 1469 persons with Down syndrome, Sharanah et al. (2017) concluded that nearly 70% have OSA. In population-based, cross-sectional study of children with Down syndrome aged 7–17, 65% of parents reported a child with sleep disturbances and 46% with sleep-related breathing disorders (Hoffmire et al., 2014). The American Academy of Pediatrics Health Supervision for Children with Down Syndrome recommends that all children with Down syndrome receive a polysomnogram by age 4 (Bassell et al., 2015). Age-related differences in sleep apnea phenotype are present in individuals with Prader-Willi syndrome. Obstructive sleep apnea is the most common finding in individuals with Prader-Willi syndrome older than 2 years, whereas central sleep apnea predominates in infants and children with Prader-Willi syndrome younger than 2 (Gillett & Perez, 2016).

In central sleep apnea, the pontomedullary pacemaker in the brain fails to signal to inspiratory muscles leading to apneic episodes (Javaheri et al., 2017). While chronic opioid use is commonly associated with central sleep apnea, persons with brain stem lesions, spinal cord injury, or neuromuscular disorders are also at increased risk (Javaheri, 2010). Cases of central sleep apnea have been reported in children with

developmental disabilities including cerebral palsy, Down syndrome, Prader-Willi, Rett syndrome, and Vici syndrome (Angriman, Caravale, Novelli, Ferri, & Bruni, 2015; El-Kersh, Jungbluth, Gringras, & Senthilvel, 2015).

Sleep-related hypoventilation is characterized by slowed breathing resulting in decreased oxygen in the blood (Böing & Randerath, 2015). The DSM-5 identifies three types of sleep-related hypoventilation-idiopathic, congenital central alveolar, and comorbid sleep related due to a medical condition (APA, 2013), and the ICSD 3 identifies three additional types-obesity related, medication or substance related, and late-onset central hypoventilation dysfunction (AASM, 2014). Sleep-related hypoventilation is rare in the general population; however, a prospective cohort study of 46 children with progressive congenital neuromuscular disease identified approximately 15% who experienced nocturnal hypoventilation (Katz et al., 2010).

Circadian Rhythm Sleep-Wake Disorders

Disturbances of the neurochemical regulation of the circadian system may result in circadian rhythm sleep-wake disorders. The DSM-5 diagnosis of a circadian rhythm sleep-wake disorder requires

> a misalignment between the endogenous circadian rhythm and the sleep-wake schedule required by an individual's physical environment or social or professional schedule. (p. 390, APA, 2013)

The category of circadian rhythm sleep-wake disorders includes five types: delayed sleep phase, advanced sleep phase, irregular sleep-wake, non-24-hour sleep-wake, and shift work (APA, 2013). Sensitivity and exposure to light are the primary environmental influences to the circadian system.

Delayed sleep phase disorder is common in adolescents and young adults, ranging from 7% to 16% with genetics, environment, and behavior playing roles in delayed sleep phase disorder. Psychiatric comorbidity is high with reports noting depression, anxiety, and alcohol use (Abbott, Reid, & Zee, 2015; Saxvig, Pallesen, Wilhelmsen-Langeland, Molde, & Bjorvatn, 2012). Delayed sleep in children and adolescents with ASD is a common caregiver concern (Goldman, Richdale, Clemons, & Malow, 2012; Hodge, Carollo, Lewin, Hoffman, & Sweeney, 2014). Sleep logs, actigraphy, chronotype questionnaires, and analysis of the biomarker salivary dim light melatonin onset aid in the diagnosis of delayed sleep phase disorder (Abbott et al., 2015).

Individuals with advanced sleep phase disorder have normal sleep duration but go to sleep earlier and wake earlier than expected (Abbott et al., 2015). In a study of 9100 adults aged 20–59 years, 0.25–7.13% experienced advanced sleep phase disorder, the prevalence increasing with age (Paine, Fink, Gander, & Warman, 2014). Advanced sleep phase disorder is rare in children (Jan, Bax, Owens, Ipsiroglu, & Wasdell, 2012) but may occur as a familial trait. Genetic variations of circadian clock protein hPER2 have been implicated in advanced sleep phase disorder (Toh et al., 2001; Xu et al., 2005).

The random sleep-wake pattern with at least three sleep periods and without an episode of major sleep typifies irregular sleep-wake rhythm disorder (APA, 2013). Presenting complaints may include daytime sleepiness and evening insomnia. The lack of an established circadian rhythm in irregular sleep-wake rhythm disorder may be due to SCN central pacemaker dysfunction, abnormal signals to the SCN, and/or altered melatonin production (Abbott & Zee, 2015). Persons with brain injury, dementia, ocular blindness, and severe or profound neurodevelopmental delays, including those with Angelman or Smith-Magenis syndromes, are at greater risk of irregular sleep-wake rhythm disorder (Abbott & Zee, 2015; Jan et al., 2012). A genetic marker for irregular sleep-wake rhythm disorder has not been identified (Sack et al., 2007). Sleep logs and actigraphy may assist in the diagnosis of irregular sleep-wake rhythm disorder. The lighting, noise, and activity of institutional settings and the difficulty an individual may have interpreting environmental condition settings may promote irregular sleep-wake rhythm disorder (Abbott &

Zee, 2015). A program for improving and managing the environment (PRIME) of institutional settings with light, noise, and motion sensors to identify awakenings is under development (Zavrel et al., 2018).

Non-24-hour sleep-wake disorder is rare in the general population and can be difficult to diagnose if not suspected (Abbott et al., 2015; APA, 2013). Suspicion should be high in persons with blindness and with complaints of insomnia and daytime sleepiness with variable sleep cycles. Non-24-hour sleep-wake disorder has been reported in over 60% of persons with blindness without light perception (Flynn-Evans, Tabandeh, Skene, & Lockley, 2014). Persons with total ocular blindness (as opposed to cortical blindness) are at increased risk for non-24-hour sleep-wake disorder due to progressively delayed melatonin production caused by disruption of the retinal-hypothalamic connection to the SCN (Jan et al., 2012). Sleep logs, actigraphy, and multiple dim light melatonin onset analyses may be helpful in identifying non-24-hour sleep-wake disorder. Comorbidity with bipolar disorder and depression is common (Abbott et al., 2015).

Parasomnias

The DSM-5 includes three categories of parasomnia: NREM movement sleep arousal disorders, nightmare disorder, and REM sleep behavior disorder (APA, 2013). The parasomnias encompass a range of undesirable phenomena during sleep and are typically distressing (Thorpy, 2012). The boundaries between states may overlap during the neurochemical shifts that signal sleep state transitions. Immaturity of the boundary regulation is the hypothesized etiology of parasomnias (Bollu, Goyal, Thakkar, & Sahota, 2018; de Lecea et al., 1998). Parasomnias in the general population range between 4% and 67% (Dosier, Vaughn, & Fan, 2017) and are more common in children (Angriman et al., 2015). While parasomnias are typically transient, they may be chronic in persons with neurodevelopmental disabilities (Angriman et al., 2015). Parasomnias may be related to epilepsy, asthma, gastrointestinal disorders, febrile illness, mental disorders, medications, substance use, or sleep deprivation (Richdale & Baker, 2014; Stores, 2014).

NREM Movement Sleep Arousal Disorders

NREM movement sleep arousal disorders typically occur early in the sleep cycle (Bollu et al., 2018) and include sleep walking (somnambulism), sleep terrors (night terrors or pavor nocturnus), and sleep-related eating or sexual behaviors with amnesia for the event (APA, 2013). While sleep walking and sleep terrors often occur among family members, sleep walking and sleep terrors are reported more often in youth with neurofibromatosis type 1 than in unaffected siblings (Licis et al., 2013; Stores, 2014). When NREM movement sleep arousal disorders are identified in multiple family members, referral to a geneticist should be considered. For persons experiencing NREM movement sleep arousal disorders, frequency may increase during times of stress or illness (Stores, 2014).

Nightmare Disorder

Nightmare disorder is a REM parasomnia characterized by disturbing or dysphoric dreams which result in awakening. When awake, the nightmare is typically recalled. Nightmare disorder is diagnosed when significant distress or impairment is noted (APA, 2013; Manni, Toscano, & Terzaghi, 2018). It is more common in children, often ceasing by adolescence although in ASD nightmares may persist (Engelhardt, Mazurek, & Sohl, 2013). The AASM *Position Paper for the Treatment of Nightmare Disorder in Adults: An American Academy of Sleep Medicine Position Paper* notes that 4% of adults experience nightmare disorder which may be idiopathic, related to medications or substance use or to a comorbid mental disorder such as PTSD (Morgenthaler et al., 2018).

REM Sleep Behavior Disorder

REM sleep behavior disorders include vocalization or complex motor behaviors during sleep (APA, 2013). The vocalizations or complex

motor behaviors are considered the enactment of dreams which occur during REM sleep. REM sleep is typically associated with muscle atonia, which prevents movements. Disruption of this mechanism may be responsible for REM sleep behavior disorders which may include minor movements or complex and aggressive movements and/or vocalizations (Bollu et al., 2018). REM sleep behavior disorders may increase in response to certain medications or substances or may be comorbid with neurodegenerative disorders or other sleep disorders. An EEG during the sleep study may be necessary to rule out a seizure disorder (McCarter et al., 2013).

Restless Legs Syndrome

Restless legs syndrome (Willis-Ekbom disease) is a neurological condition creating an uncomfortable and difficult-to-describe sensation in the legs accompanied by an irresistible impulse to move the legs. Ohayon, O'Hara, and Vitiello (2012) conducted a review of epidemiological studies, reporting incidence in the general population from 1.9% to 4.6% with an increased incidence in women particularly during pregnancy. Familial incidence has been noted, but a specific genetic variant has not been identified. Large-scale whole-exome or whole-genome sequencing studies are needed to advance our understanding of restless legs syndrome (Winkelmann et al., 2017). Persons with ASD and fragile X experience restless legs syndrome more frequently than neurotypicals (Lane et al., 2015; Summers et al., 2014). Because restless legs syndrome interferes with sleep, insomnia and daytime sleepiness are common complaints. While it is often comorbid with medical and mental disorders, symptoms similar to restless legs syndrome may be caused by numerous medical conditions including iron deficiency anemia, renal disease or drug-induced akathisia (Trenkwalder, Allen, Högl, Paulus, & Winkelmann, 2016). The International Restless Legs Syndrome Study Group has published a comprehensive history and consensus criteria for the diagnosis of restless legs syndrome (Allen et al., 2014).

Substance/Medication-Induced Sleep Disorder

The DSM-5 includes the category of substance/medication-induced sleep disorder to capture insomnia, daytime sleepiness, and parasomnias caused by intoxication or discontinuation/withdrawal (APA, 2013). Because persons with intellectual disability are at increased risk for medical and mental disorders, treatment with medication is common. Sleep disturbance is associated with many medications including those for allergies, heart conditions, seizures, and mental disorders.

Screening for Sleep Disturbances

Professional organizations including those representing psychologists, physicians, and dentists recommend that clinicians routinely screen all patients for sleep disturbances (Addy et al., 2018; Meltzer, Phillips, & Mindell, 2009; Sorscher, 2011; Watson et al., 2015). Even with adequate screening, sleep disorders may go undiagnosed in persons with intellectual disability because of their complex presentation and the frequent comorbidity. Table 23.2 summarizes common sleep disorders and diagnostic considerations in persons with intellectual disability.

In a large practice-based network study screening waiting room patients, more than 90% reported sleep disturbances, but less than 20% reported discussing their symptoms with their primary care provider. Only 23% of the providers reported routine screening for sleep issues (Mold et al., 2011). Failure to screen for sleep disorders has been noted in reviews of patient records (Erichsen et al., 2012). Persons with intellectual disability may be at greater risk for having sleep disturbances underidentified due to communication difficulties, ageism, caregivers viewing sleep disturbances as behavioral problems, or diagnostic overshadowing. Diagnostic overshadowing refers to the tendency of clinicians to misattribute symptoms to the primary disorder or a behavioral problem, leading to a failure to properly diagnose. An Australian study of persons living in group homes for the

Table 23.2 Common sleep disorders and diagnostic considerations in persons with intellectual disability

Primary disorder	Common sleep disorders	Diagnostic considerations
ADHD The most common neurodevelopmental disorder in childhood, characterized by poor attention, impulsivity, and hyperactivity. [a]	Any sleep disorder 23–73%[b, c] OSA 25–30%[d] RLS/periodic limb movement up to 44%[e]	The AAP recommends screening for sleep disorders as part of the ADHD assessment[f] Medications used to treat ADHD may cause poor sleep or sedation[g]
Angelman syndrome A disorder with multiple genetic etiologies characterized by dysmorphic facial features[h]	Any sleep disorder 20–80%[i] Severe night waking 46%, increased risk for breathing disorders[j] ISWD risk increased[k, l]	The consensus guidelines for AS note abnormal sleep-wake cycles and diminished need for sleep as common characteristics[h]
Autism spectrum A neurodevelopmental disorder of impaired social communication and interaction with restricted or repetitive features[m]	Any sleep disorder 50–80%[n] Difficulties with sleep onset (30%) and sleep maintenance 43%[j] Reduced sleep time[o] 60% difficulty falling asleep[p, q] RLS increased[r, s]	The AAP has issued guidelines for ASD[t] Childhood insomnia may persist into adulthood[p]
Visual impairment Persons without light perception have greater sleep disturbance than those with cortical blindness	Any sleep disorder up to 60%[u, j, k] ISWRD 35%[u] non-24 SW disorder increased with ocular blindness (no light perception)[j, k]	With age, failing vision may contribute to sleep disturbance
Cerebral palsy A disorder caused by a nonprogressive brain injury occurring in the perinatal period[w]	Any sleep disorder 44.0% Disorders of initiation and maintenance of sleep, was found in 26.0%[v, w]	Up to 80% have comorbid health conditions, including pain, that contribute to sleep disturbances[v, w]
Down syndrome A genetic disorder characterized by a full or partial copy of chromosome 21[x]	Any sleep disorder 65%[y] OSA 30–63%[z] Up to 70% with OSA[aa] 21% with sleep-related movement disorder[y]	Over half of parents may not recognize their child has a sleep disorder[y] AAP recommends PSG by age 4[z]
Fetal alcohol spectrum A common cause of ID due to maternal alcohol consumption during fetal development[x]	Any sleep disturbance up to 80%[bb]	Increased prevalence of sleep disorders with comorbid ASD or ADHD[x] The AAP provides a (FASD) toolkit at https://www.aap.org/en-us/advocacy-and-policy/aap-health-initiatives/fetal-alcohol-spectrum-disorders-toolkit/Pages/default.aspx
Fragile X The most common genetic disorder associated with ID. It is caused by a mutation in the FMR1 gene[x]	Any sleep disturbance 27–50%[cc] Sleep initiation problems and frequent nighttime awakenings >60% Early awakening 49% Multiple sleep disturbances are common[dd]	Because pain thresholds are often high in persons with fragile X, the presence of medical conditions causing pain may present as sleep disturbance[cc]

(continued)

Table 23.2 (continued)

Primary disorder	Common sleep disorders	Diagnostic considerations
Neurofibromatosis type 1 (NF1) A genetic disorder caused by an abnormality of chromosome 17q11.2 which disrupts the protein neurofibromin causing tumor growth[ee]	Any sleep disorder >50% Sleep walking and sleep terrors[ff] Periodic limb movements 54% Features of obstructive sleep apnea 43% Confusion on waking >10%[gg]	If a seizure disorder is present, it may exacerbate sleep disturbances The American College of Medical Genetics and Genomics published a clinical practice resource for adults with NF1[ee]
Prader-Willi A genetic disorder caused by a chromosome 15 abnormality which disrupts hypothalamic function[x]	OSA in nearly 80%, narcolepsy 35.71%[hh] Excessive sleepiness 67% Hypersomnia disorder 43% Narcolepsy 35% Only 30% had normal sleep[ii]	The AAP has issued a clinical report on the health supervision of children with PWS advising screening for obstructive sleep apnea, restlessness, and excessive daytime sleepiness[jj]
Smith-Magenis syndrome A rare developmental disorder caused by a chromosome 17 p11.2 deletion[kk]	Any sleep disturbance 92%, night waking 81%, early morning waking 73%[j] ISWRD[k, l]	An altered melatonin profile contributes to the sleep disturbances of persons with SMS[kk] If a seizure disorder is present, it may exacerbate sleep disturbances Gastroesophageal reflux was associated with increased sleep-disordered breathing[j]
Williams syndrome A rare developmental disorder associated with ID caused the deletion of genes at chromosome 17 q11.23[x]	Any sleep disturbance 97% by parent report Daytime sleepiness 61%, snoring 36% Resisting bedtime 45%, sleep talking 15%[ll] Decreased sleep efficiency 48%[mm]	Breathing and heart problems may contribute to sleep disturbances[ll] Melatonin and cortisol abnormalities may play a role in sleep disturbances in WS[nn]

[a]Thomas, Sanders, Doust, Beller, and Glasziou (2015), [b]Reale et al. (2017), [c]Sung, Hiscock, Sciberras, and Efron (2008), [d]Youssef, Ege, Angly, Strauss, and Marx (2011), [e]Cortese et al. (2005), [f]"ADHD: Clinical Practice Guideline for the Diagnosis, Evaluation, and Treatment of Attention-Deficit/Hyperactivity Disorder in Children and Adolescents" (2011), [g]Spruyt and Gozal (2011), [h]Williams et al. (2006), [i]Dosier et al. (2017), [j]Trickett, Richards, Heald, and Oliver (2016), [k]Jan et al. (2012), [l]Abbott and Zee (2015), [m]APA (2013), [n]Kotagal and Broomall (2012), [o]Humphreys et al. (2014), [p]Mayes and Calhoun (2009), [q]Engelhardt et al. (2013), [r]Lane et al. (2015), [s]Summers et al. (2014), [t]Malow et al. (2012), [u]Tamura et al. (2016), [v]Horwood et al. (2018), [w]Lelis, Cardoso, and Hall (2016), [x]Stores (2014), [y]Hoffmire et al. (2014), [z]Bassell et al. (2015), [aa]Sharanah et al. (2017), [bb]Chen, Olson, Picciano, Starr, and Owens (2012), [cc]Kidd et al. (2014), [dd]Kronk et al. (2010), [ee]Stewart, Korf, Nathanson, Stevenson, and Yohay (2018), [ff]Licis et al. (2013), [gg]Leschziner, Golding, and Ferner (2013), [hh]Sedky et al. (2014), [ii]Ghergan et al. (2017), [jj]McCandless (2010), [kk]Spruyt, Braam, Smits, and Curfs (2016), [ll]Annaz, Hill, Ashworth, Holley, and Karmiloff-Smith (2011), [mm]Mason et al. (2011), [nn]Sniecinska-Cooper et al. (2014)

elderly with intellectual disabilities documented misattribution by care staff resulting in a failure to refer for sleep assessment. Misattributing sleep disorders to cognitive decline or dementia is particularly concerning as sleep disorders may contribute to cognitive decline and dementia (Mold et al., 2011). The need for clinician and caregiver training on sleep disorders has been widely advocated (Addy et al., 2018; Meltzer et al., 2009; Mindell et al., 2013; Sorscher, 2011).

Clinicians should screen all new patients for sleep disturbances and periodically rescreen (Fig. 23.2).

Screening should include direct questioning of the patient when possible. In addition, caregivers should be queried and may be asked to complete standardized screening instruments (See

Is there difficulty falling or staying asleep?

Are there snoring or breathing issues?

Is there daytime sleepiness?

Is there difficulty waking?

Are there other sleep behaviors of concern?

Fig. 23.2 Basic screening questions for sleep disturbances

Table 23.3). The need for rescreening may be signaled by changes in behavior including night wandering, excessive sleepiness, loss of adaptive skills, increased irritability, and/or snoring. Clinicians should keep in mind that more than one sleep disorder may be present (Abbott & Zee, 2015; Cortese, Ivanenko, Ramtekkar, & Angriman, 2014). While screening identifies the possibility of a sleep disorder, further assessment of sleep disturbances must be conducted to differentiate sleep problems and sleep disorders.

The Assessment of Sleep Disorders

The assessment of sleep disorders begins with obtaining a detailed sleep history (Table 23.4) to distinguish sleep disturbances representing a sleep problem from those indicating a sleep disorder. Common sleep problems include bedtime resistance, difficulty settling at bedtime, fears, difficulty staying in bed, restless sleep, difficulty returning to sleep after arousals, night awakenings, and night wandering (Cortese et al., 2014; Schutte-Rodin et al., 2008). Sleep problems are the most common complaints from caregivers. Other common complaints that may indicate a specific sleep disorder include somnambulism (sleep walking), somniloquy (sleep talking), night terrors, snoring or sleep-disordered breathing, unusual sleep movements, excessive sleepiness, and difficulty falling or staying asleep (Arbuckle et al., 2010; Guilleminault & Pelayo, 2000; Kovachy et al., 2013; Laberge, Tremblay, Vitaro, & Montplaisir, 2000; Montgomery-Downs, O'Brien, Holbrook, & Gozal, 2004; Szelenberger, Niemcewicz, & DĄbrowska, 2005). Of course, sleep problems and sleep disorders are often comorbid as are developmental disabilities, for example, fragile X, Smith-Magenis syndrome, and Rett syndrome may be comorbid with ASD (Mullegama et al., 2015). This adds to the diagnostic complexities of the assessment of sleep disorders in persons with developmental disabilities. The clinician should obtain a detailed sleep history to rule out a sleep disorder before diagnosing a sleep problem. Ruling out a sleep disorder often requires multidisciplinary expertise.

The comprehensive sleep history of persons with intellectual disability has been detailed elsewhere (Barnhill, 2006; Fletcher et al., 2016; Stores, 2014). In addition, the AASM publishes practice guidelines which may be accessed at https://aasm.org/clinical-resources/practice-standards/. The sleep needs of persons with specific neurodevelopmental disorders are addressed by the American Academy of Pediatrics *Health Supervision for Children with Down Syndrome* (Bull, 2011), *Management of Children with Autism Spectrum Disorders* (Myers & Johnson, 2007), and *Health Supervision for Children with Fragile X Syndrome* (Hersh & Saul, 2011). The National Down Syndrome Society has published *Aging and Down Syndrome: A Health & Well-being Guidebook* (http://www.ndss.org/wp-content/uploads/2017/11/Aging-and-Down-Syndrome.pdf) (Moran, 2017).

Clinicians should be aware that a bidirectional risk relationship between sleep disorders and psychiatric disorders has been identified (Barnhill et al., 2016; Schutte-Rodin et al., 2008). Persons with intellectual disability are at increased risk for mental disorders (Buckles, Luckasson, & Keefe, 2013; Munir, 2016) as are persons with ASD (Adams, Matson, Cervantes, & Goldin, 2014). Sleep disturbances are common in persons with most mental disorders including ADHD, mood disorders, anxiety disorders, psychotic disorders, and PTSD. A systematic review between the relationship of ADHD and OSA notes a bidirectional relationship with 95% of persons with OSA experiencing attention difficulties and

Table 23.3 Common sleep assessment tools

Sleep diaries	
AASM Sleep Diary Available at: http://yoursleep.aasmnet.org/pdf/sleepdiary.pdf	Adults, self-administered
Consensus Sleep Diaries Carney et al. (2012) Available at: http://drcolleencarney.com/wp-content/uploads/2013/10/Final-CSD-Morning-only-with-instructions.pdf	Adults, self-administered 3 versions available which were developed by experts in insomnia with the collaboration of potential users of the diaries
National Sleep Foundation Sleep Diary Available at: https://www.sleepfoundation.org/sites/default/files/SleepDiaryv6.pdf	Adults, self-administered
National Sleep Foundation Sleep Diary for Children Available at: http://www.sleepforkids.org/pdf/SleepDiary.pdf	Children, children will need assistance to complete this diary Includes graphics as prompts, sleep tips, and puzzles
Sleep questionnaires	
Athens Insomnia Scale (AIS) (Soldatos, Dikeos, & Paparrigopoulos, 2000) Available at: https://www.med.upenn.edu/cbti/assets/user-content/documents/Athens%20Insomnia%20Scale%20(AIS).pdf	Adults and adolescents, has been used in persons with ASD, ID 8- and 5-item scales Based on ICD-10 criteria for insomnia
BEARS Sleep Screening Algorithm (Owens & Dalzell, 2005) Available at: https://depts.washington.edu/dbpeds/Screening Tools/BEARSsleep.doc	Toddler/preschooler, school-aged children, and adolescents Completed by caregiver for younger children; questions asked directly to adolescents Screening tool for sleep disorders (acronym: *b*edtime problems; *e*xcessive daytime sleepiness; *a*wakenings; *r*egularity and duration of sleep; *s*noring) Used in children with neurodevelopmental disabilities (Jan et al., 2008)
Bedtime Routines Questionnaire (BRQ) (Henderson & Jordan, 2010) Available in article	Preschool and early school age, completed by caregiver 31 item assessment of bedtime routines Has been used in children with ASD (Henderson, Barry, Bader, & Jordan, 2011)
Behavioral Evaluation of Disorders of Sleep Scale (BEDS) (Schreck, Mulick, & Rojahn, 2003)	Children, completed by caregiver Includes 6 month history of sleep disturbance, problems with awakening, sleep environment and facilitators of sleep, apnea, bruxism Assists with identification of sleep disorders Has been studied in children with ASD (Taylor, Schreck, & Mulick, 2012), DS (Esbensen & Hoffman, 2017), and Angelman Syndrome (Schreck et al., 2003; Walz, Beebe, & Byars, 2005)
Bergen Insomnia Scale (BIS) (Pallesen et al., 2008)	Adults, self-administered Six items – sleep onset, maintenance, early morning awakening, sleep satisfaction, daytime impairment, and subjective impression of restfulness
Berlin Questionnaire (BQ) (Netzer, Stoohs, Netzer, Clark, & Strohl, 1999)	Adults, 11 questions Predicts high or low risk for OSA. Not recommended by the AASM for the diagnosis of OSA due to high number of false positives (Kapur et al., 2017)

(continued)

Table 23.3 (continued)

Calgary Sleep Apnea Quality of Life Index (SAQLI) (Ward Flemons & Reimer, 1998)	Adults, 35 questions, 30 minutes to complete Assesses daily functioning, social interactions, emotional functioning, and symptoms of sleep apnea
Children's Sleep Habits Questionnaire (Owens, Spirito, & McGuinn, 2000) Available at: njaap.org/.../uploads/2016/04/Childrens-Sleep-Habits-Questionnaire.pdf	Preschool, school-age children (ages 4–10) completed by caregiver Assesses major medical and behavioral sleep disorders of childhood Sleep behavior and problems observed including bedtime behavior, anxiety, onset and duration of sleep, daytime sleepiness, parasomnias, and sleep-disordered breathing; available as abbreviated version Has been studied in children with Williams syndrome (Annaz et al., 2011), ASD (Johnson et al., 2016), ID (Esbensen & Hoffman, 2017), CP (Wayte, McCaughey, Holley, Annaz, & Hill, 2012), and DS/ID (Esbensen & Hoffman, 2017)
Cleveland Adolescent Sleepiness Questionnaire (CASQ) (Spilsbury, Drotar, Rosen, & Redline, 2007) Available at: http://yoursleep.aasmnet.org/pdf/CASQ.pdf	Questions asked to adolescent Subjective sleepiness, alertness, and falling asleep at school and at home Used for children with neurodevelopmental disorders (Szuhay & Rotenberg, 2009)
Dysfunctional Beliefs and Attitudes about Sleep Questionnaire (DBAS) (Morin, 1994) *Brief Version* (DBAS-16) (Morin, Vallières, & Ivers, 2007)	Adults, self-administered 28-item assessment of sleep-disruptive cognitions Useful in the assessment of insomnia
Epworth Sleepiness Scale (ESS) (Johns, 1991) Available at: https://epworthsleepinessscale.com/	Adults, self-administered Measures daytime sleepiness in various situations in daily life to help identify sleep disorders Validated against the multiple sleep latency test and polysomnography Not recommended by the AASM for the diagnosis of OSA due to high number of false positives (Kapur et al., 2017)
Epworth Sleepiness Scale for Children and Adolescents (ESS-CHAD) (Janssen, Phillipson, O'Connor, & Johns, 2017) Available at: https://epworthsleepinessscale.com/	Children and adolescents completed by caregiver until age 9; may ask questions directly youth aged 9–18, measures daytime sleepiness Similar to adult ESS in content but questions modified to better reflect experiences and comprehension of this age group
Functional Outcomes of Sleep Questionnaire (FOSQ) (Weaver et al., 1997) *Functional Outcomes of Sleep Questionnaire* (FOSQ-10) (Chasens, Ratcliffe, & Weaver, 2009)	Adults, 30- and 10-item formats Evaluate the impact of excessive daytime sleepiness
Infant Sleep Questionnaire (ISQ) (Morrell, 1999) *Brief Infant Sleep Questionnaire* (BISQ) (Sadeh, 2004) Available in article	Infant to 3 years, completed by caregiver Includes duration, awakenings, sleep onset, position, and sleep location
Insomnia Severity Index (ISI) (Morin, 1993)	Adults, self-administered Evaluates sleep disturbances, quantifies severity of insomnia (criteria to determine "clinical insomnia"), and measures responses to intervention Validated by Bastien, Vallieres, and Morin (2001)
Multidimensional Fatigue Inventory or Fatigue Severity Scale (Smets, Garssen, Bonke, & De Haes, 1995)	Adults Measures five components of fatigue: general, mental, physical, and diminished motivation and activity level Used in a wide range of physical illnesses

(continued)

Table 23.3 (continued)

Multivariable Apnea Prediction Questionnaire (MAP) (Maislin et al., 1995)	Adults A mathematical formula using three OSA symptom questions, BMI, age and sex screens for OSA Not recommended for elderly with mild cognitive impairment (Wilson et al., 2014) Not recommended by the AASM for the diagnosis of OSA due to high number of false positives (Kapur et al., 2017)
Obstructive Sleep Disorders 6-Survey (OSD-6) (de Serres et al., 2000)	Young children through adolescence Completed by caregiver Physical issues, emotional distress, sleep disturbance, speech/swallowing problems, and concerns of caregiver
Parental Interactive Bedtime Behavior Scale (PIBBS) (Morrell & Cortina-Borja, 2002)	Infants – completed by caregiver Comforting of infant (active and passive physical comforting and social comforting), movement, and promotion of autonomy
Pediatric Daytime Sleepiness Scale (PDSS) (Drake et al., 2003) Available in article	School-age children (targets middle school), self-administered Includes patterns of sleep and sleepiness during the day Higher scores associated with lower achievement, more frequent illness, decreased enjoyment of school
Pediatric Sleep Questionnaire (PSQ) (Chervin, Hedger, Dillon, & Pituch, 2000) Available at multiple cites online	Children and adolescents, completed by caregiver Includes assessment of snoring, sleep apnea, restless sleep, difficulty waking up, unrefreshing sleep, sleepiness, and related behavior problems Has been used in children with CP (Sandella, O'Brien, Shank, & Warschausky, 2011)
Pittsburgh Sleep Quality Index (PSQI) (Buysse, Reynolds 3rd, Monk, Berman, & Kupfer, 1989) Available at: http://www.sleep.pitt.edu/research/instruments.html	Adults, self-rated questionnaire Measures sleep quality and sleep disturbance over the past month Subjective report of sleep duration, latency, quality, efficiency, disturbances, medications for insomnia, and daytime dysfunction Used as a measure of parental stress (Hoffman et al., 2008) and adults with ASD (Baker, Richdale, & Hazi, 2018)
Sleep Disorders Questionnaire (SDQ) (Douglass et al., 1994)	Adults 175-item screen for sleep apnea, psychiatric disorders, narcolepsy, and sleep-related movement disorders Has been used in persons with ID (Maas et al., 2010)
Sleep Disturbance Scale for Children (SDSC) (Bruni et al., 1996) http://www.midss.org/content/sleep-disturbance-scale-children-sdsc	Children and adolescents, completed by caregiver Screens for all major sleep disorders Has been studied in children with ASD (Miano et al., 2007) and DS (Esbensen & Hoffman, 2017)
Sleep Questionnaire (SQ-SP) (Simonds & Parraga, 1982)	Individuals with ID (Maas et al., 2011) Evaluates sleep disturbances and associated behavioral problems Includes snoring, daytime sleepiness, sleep-disordered breathing, anxiety, and other sleep-related complaints
Sleep and Settle Questionnaire (SSQ) (Matthey, 2001)	Infants, completed by caregiver Sleep patterns including time to settle, crying, temperament, and caregiver confidence on getting infant to sleep again
STOP-BANG Questionnaire (Chung et al., 2012) Available at: http://stopbang.ca/osa/screening.php	Adults Screening tool for obstructive sleep apnea Questions about snoring, apnea observed during sleep, tiredness. Includes information on risk factors of male gender, age, BMI, neck circumference, and elevated blood pressure Not recommended by the AASM for the diagnosis of OSA due to high number of false positives (Kapur et al., 2017)
Tayside Children's Sleep Questionnaire (McGreavey, Donnan, Pagliari, & Sullivan, 2005) Available at: jacqui.mcgreavey@tpct.scot.nhs.uk	Young children, completed by caregiver Problems with sleep onset or awakenings, includes caregiver interventions

Table 23.4 Sleep history

Primary insomnia complaint:	Characterization of complaint(s): Difficulty falling asleep Awakenings Poor or unrefreshing sleep Onset Duration Frequency Severity Course Perpetuating factors Past and current treatments and responses
Presleep conditions:	Pre-bedtime activities Bedroom environment Evening physical and mental status
Sleep-wake schedule (average, variability):	Bedtime: Time to fall asleep: Factors prolonging sleep onset Factors shortening sleep Awakenings: Number, characterization, duration Associated symptoms Associated behaviors Final awakening versus time out of bed Amount of sleep obtained
Nocturnal symptoms:	Respiratory Motor Other medical Behavioral and psychological
Daytime activities and function:	Identify sleepiness versus fatigue Napping Work Lifestyle Travel Daytime consequences: Quality of life Mood disturbance Cognitive dysfunction Exacerbation of comorbid conditions

Citation: Schutte-Rodin, Broch, Buysse, Dorsey, and Sateia (2008)

20–30% of persons with ADHD qualifying for an OSA diagnosis (Youssef et al., 2011). When assessing attention difficulties or hyperactivity, sleep disorders should be included in the differential diagnosis. When sleep disturbance is identified in a person with a mental disorder, care should be taken to rule out a comorbid sleep disorder. In addition, the assessment should include inquiry into the use of caffeine, nicotine, substance use, and prescribed medications. Psychiatric medications as well as medications prescribed for seizures, heart conditions, asthma, and allergies are known to interfere with sleep. Over-the-counter medications for headache and colds may cause sedation or difficulties falling asleep. The contribution of herbal products, such as St. John's wort (*Hypericum*) and coenzyme Q10, to sleep disturbance should also be considered. The National Center for Complementary and Integrative Health (NCCIH) provides up-to-date information on herbals and other alternative medications at https://nccih.nih.gov/health/herbsataglance.htm.

When a sleep disorder is suspected or interventions for sleep problems are unsuccessful, coordination with primary care or other specialists is indicted. Persons with intellectual disability often receive care from multiple health-care providers. Coordination of care between health-care providers and with caregivers is critical to the well-being of persons with intellectual disability.

Primary Care and Specialty Assessments

The evaluation for sleep disorders and refractory sleep problems should include referral to a primary care provider. A physical examination should be performed to identify risk factors for sleep disorders. For example, risk for breathing-related sleep disorders is associated with obesity, neck circumference, upper airway problems, and craniofacial abnormalities (Schutte-Rodin et al., 2008). Laboratory studies may be done to identify iron deficiency anemia or renal disease, which are associated with restless legs syndrome (Trenkwalder et al., 2016), and dim light melatonin onset may be assessed for sleep phase typing (Burgess, Wyatt, Park, & Fogg, 2015). A referral to a sleep specialist for a sleep study may be necessary to capture objective measures.

If disordered breathing is identified during a sleep study, a referral to an otolaryngologist (ENT) may be appropriate. Sleep-disordered breathing is more common in persons with Down syndrome and Prader-Willi syndrome as well as persons with neuromuscular disorders or craniofacial abnormalities (Cortese et al., 2014; Stores, 2014). These conditions are often associated with obesity. Persons with intellectual disability are at increased risk of obesity, a known risk factor for sleep-disordered breathing (Wakefield, Sanderson, & McPherson, 2018). The ENT may recommend an adeno-tonsillectomy, or in the case of complex craniofacial abnormalities, a multidisciplinary surgical team including ENT, an oral surgeon, and plastic surgeon may perform multiple surgeries (Dehlink & Tan, 2016). Should the ENT recommend a surgical intervention, the clinician may need to assist in preparing the patient for the medical procedure.

The evaluation and treatment of pain as an etiology of disturbed sleep can be challenging in persons who have impaired communication and cognition. Persons with intellectual disability are at risk for comorbid health conditions. Some developmental disabilities like cerebral palsy and neuromuscular disorders may predispose a person to muscle or joint pain (Havercamp & Scott, 2015). Persons with intellectual disability may have atypical pain thresholds, with increased or reduced sensitivity to painful stimuli. Even in the absence of obvious discomfort, medical conditions causing pain may be present. Some individuals, particularly the elderly, may remain afebrile despite infection. Pain may present as the sudden onset of regression with the new onset of behavioral challenges or loss of skills, self-harm, or impaired sleep in persons with intellectual disability. Regression may include the new onset of behavioral challenges or loss of skills. The primary care provider should investigate possible infections, dental issues, headaches, gastrointestinal disorders, and musculoskeletal disorders or injury as conditions that may cause discomfort and contribute to sleep disturbances (Richdale & Baker, 2014). Medications for pain or related conditions may cause sleep disturbance. The chronic use of opioids is of particular concern as this is associated with decreased respiration. In some cases, referrals to medical specialists, neurologist, gastroenterologists, orthopedists, and other professionals may contribute to the assessment, diagnosis, and treatment of pain.

Sleep problems like nocturnal enuresis or incontinence may require assessment to rule out a physical cause such as diabetes or urinary tract infection. Persons with ID are more likely to have nocturnal enuresis or incontinence (Bassell et al., 2015; Niemczyk et al., 2017; von Gontard & Equit, 2015; Wagner, Niemczyk, Equit, Curfs, & von Gontard, 2017). Nocturnal enuresis or incontinence may represent a urological or neurological disorder. Primary nocturnal enuresis in children over nine, new onset nocturnal enuresis, or incontinence should be referred to a urologist. Loss of bladder control may indicate new onset or a poorly controlled seizure disorder requiring neurological assessment.

The primary care provider may refer to a geneticist or order genetic testing to identify syndromes associated with sleep disturbance. The majority of individuals with severe or profound intellectual disability have genetic findings on whole-exome and whole-genome sequencing (Vissers, Gilissen, & Veltman, 2016). Persons with HLA DQ B1∗05:01 and HLA DQ B1∗04 variants often have associated NREM movement sleep arousal disorders (Bollu et al., 2018). A family history of sleep walking should raise suspicion of single-nucleotide polymorphisms at chromosome 20q12-q13.12 (Licis et al., 2011). Genetic disorders with associated sleep disturbances include fragile X, tuberous sclerosis, and syndromes including Down, Williams, Smith-Magenis, Rett, Prader-Willi, Angelman, Lesch-Nyhan, and others (Dosier et al., 2017; Stores, 2014). Approximately a third of persons with cerebral palsy (Fahey et al., 2017) and more than 15% of persons with autism and intellectual disability (Bourgeron, 2016) may have genetic vulnerabilities. Stores' book *Sleep and Its Disorders in Children and Adolescents with a Neurodevelopmental Disorder* (Stores, 2014) offers a comprehensive review of genetic disorders associated with sleep disturbances.

When behavioral and primary care interventions fail to resolve sleep issues or a sleep study is needed, referral to a sleep specialist is indicated (Pagel, 2009). While the most common sleep-related complaints are challenging sleep-related behaviors and insomnia is the most common disorder addressed clinically, the most common referrals to sleep specialists are to evaluate sleep-disordered breathing (Wong & Ng, 2015). The American Board of Sleep Medicine (https://www.behavioralsleep.org) certifies predoctoral or doctoral psychologists and physicians as behavioral sleep medicine specialists. According to the American Board of Medical Specialties (https://www.abms.org/member-boards/specialty-subspecialty-certificates/), sleep medicine subspecialties are offered by the boards of anesthesiology, family medicine, internal medicine, otolaryngology, pediatrics, and psychiatry. The American Academy of Dental Sleep Medicine represents dentists specializing in sleep disorders (https://aadsm.org). The AASM has established the American Alliance for Sleep Health as a patient support organization (http://www.sleepallies.org) with information, educational materials, information about financial assistance, and sleep disorder fact sheets.

When making a referral to a sleep specialist, a thorough description of the sleep concerns should be sent. In addition, the referral should include any interventions that have been attempted, a note listing medications, caffeine and alcohol use, copies of standardized sleep questionnaires that have been completed, and a sleep diary with at least 2 weeks of data (Schutte-Rodin et al., 2008). Common diaries and sleep assessment instruments are noted in Table 23.2.

Sleep diaries are necessary to augment the clinical sleep history and sleep questionnaires. Sleep diaries provide a record of sleep onset, wake time, and sleep duration along with other factors impacting sleep efficiency (Mouthon & Huber, 2015). A caregiver should be asked to complete 2 weeks of data collection using a sleep diary. Careful instructions should be given about the importance of recording data each morning and evening as accurate information greatly enhances diagnosis (Werner, Molinari, Guyer, & Jenni, 2008). The sleep diary may be modified with simple additions to capture illness, pain, seizure activity, screen time, medication administration times, and other patient-specific factors which main impact sleep. Sleep diary apps are now available and are comparable to paper versions for reliability (Ibáñez, Silva, & Cauli, 2018b). Because sleep diaries have been found to be a less accurate measure of sleep awakenings, actigraphy may be combined with a sleep diary to capture awakenings (Werner et al., 2008). Ibáñez, Silva, and Cauli (2018a) have compiled a comprehensive systematic review of sleep assessment questionnaires with data on uses, accuracy, and validation studies. The psychometric properties of questionnaires vary widely. While many have been used in studies involving persons with intellectual disability, few are fully validated (Spruyt & Gozal, 2011). Questionnaires are not standalone diagnostic instruments but are valuable to inform clinical judgment and allow comparison of patient and/or caregiver reports over time. For persons with neurodevelopmental disorders, there should be a low threshold for requesting objective studies.

In addition to subjective sleep questionnaires, sleep specialists use a variety of objective measures as part of a sleep study to further evaluate sleep and diagnose sleep disorders. Sleep studies are typically performed overnight in a bedroom-like setting for the assessment of sleep-disordered breathing, circadian rhythm disorders, sleep-related movement disorders, parasomnias, or insomnia. To assess symptoms of hypersomnia or narcolepsy, the nocturnal sleep study may be combined with a daytime multiple sleep latency test. Prior to a sleep study, patient-specific instruction should be obtained from the sleep lab. Typical instructions include a regular sleep schedule prior to the study and avoiding naps and vigorous exercise the day of the study. Because electrodes will be placed on the scalp, hair should be clean but free of any oils or other products. Caregivers should be informed which medications to take the day of the sleep study as stimulants or sleep-promoting medications may need to be held. Persons with intellectual disability may require additional preparation for the proce-

dure. This may include simulated medical procedures, exposure therapy, distraction, and counterconditioning (Paasch, Leibowitz, Accardo, & Slifer, 2016). The Kennedy Krieger Institute, the leading center for research and treatment of children with developmental disabilities, in association with the AASM and NIH has developed *The Sleep Study Story* to help prepare children for the sleep study experience. It can be accessed at https://www.kennedykrieger.org/sites/default/files/library/documents/patient-care/centers-and-programs/sleep-disorders-clinic-and-lab/sleep-story.pdf.

The polysomnogram is considered the "gold standard" of sleep assessment as it captures sleep architecture and a number of body functions including brain wave activity (EEG), eye movements (EOG), muscle activity (EMG), cardiac function (EKG and blood pressure), and a range of pulmonary functions (Deak & Epstein, 2009; Gregory & Sadeh, 2016). In addition to these measures, video recording captures movement including seizure activity as well as snoring, snorting, and gasping. If gastroesophageal issues such as reflux are suspected, additional parameters may be collected (Ibáñez et al., 2018b).

Actigraphs are small monitors that measure physical activity. They are typically worn of the wrist but may be placed on the ankle, waist, or chest. Complex algorithms analyze the data from the actigraph to provide information on sleep-wake patterns. While the actigraph does not capture sleep architecture or respiratory information, it has the advantage of allowing data collection in the home environment and can be worn for several sleep cycles making it useful in the assessment of insomnia and circadian rhythm sleep-wake disorders or for persons who cannot tolerate the sleep lab (Smith et al., 2018).

From the use of the EEG to study sleep in the 1930s to the polysomnography, sleep assessment technology has steadily progressed and become more advanced (Deak & Epstein, 2009). Medically prescribed home sleep apnea testing is now available; however, it is recommended only for adults. Furthermore, it should not be used for persons with cardiac, respiratory, or neuromuscular conditions (Kapur et al., 2017). Numerous apps and home devices are marketed directly to the consumer for the assessment of sleep. To date, this technology varies widely and has not achieved the efficacy of the polysomnograph in assessing sleep efficiency (Bhat et al., 2015; de Zambotti, Goldstone, Claudatos, Colrain, & Baker, 2018; Ong & Gillespie, 2016).

Sleep specialists may interpret the sleep study results and implement treatment strategies or send results to the referring primary care provider to coordinate follow-up. In addition to behavioral interventions, referrals may be made to a neurologist, psychiatrist, ENT, or gastroenterologist for targeted intervention. Mental health clinicians will need to make a specific request for the sleep study results. While the report will document the reasons for the study, sleep study technical parameters, and recommendations, the clinician will need to personalize the details to address individual patient needs (Shrivastava, Jung, Saadat, Sirohi, & Crewson, 2014). The clinician's knowledge of the patient's strengths and supports is critical to the successful treatment of sleep disturbances. On receiving the report, the clinician should review prereferral assessments and interventions. Elements with a potential impact on the sleep study results should each be reconsidered. For example, a complaint of difficulty falling asleep at home paired with a sleep study indicating a short sleep latency (falling asleep quickly) may direct attention to environmental or behavioral concerns. Excessive movement or shortened REM sleep during the study may necessitate reconsidering the contribution of medications to sleep disturbances. Over 25 medications may cause sleep-related movements (Stallman, Kohler, & White, 2018). Care should be taken to avoid diagnostic overshadowing. Just because poor sleep quality is more common in persons with intellectual disability, the etiologies of intellectual disability and poor sleep should not be conflated. Rigorous attention must be given to potential medical or mental health disorders as the cause of disordered sleep. In addition, the clinician will need to develop person-centered strategies to address sleep hygiene concerns including caffeine, nicotine or alcohol use, weight loss, exercise, and electronic use. Because

of the complexity of diagnosing and treating sleep disorders in persons with developmental disabilities, clinicians, primary care providers, medical specialists, and sleep specialists should coordinate their efforts with those of caregivers to address sleep disorders.

Future Directions

Sleep health is critical to mental health and general well-being. It is well documented that sleep disturbances, both sleep problems and sleep disorders, are common in persons with intellectual disability but are often underdiagnosed (Fletcher et al., 2016; Stores, 2016; Surtees et al., 2018). This highlights the necessity of conducting a sleep assessment as part of all mental health evaluations. In addition, a disproportionate impact from medical and mental health disorders is common among persons with developmental disabilities. Sleep disturbances are known to exacerbate health, mental health, and behavioral problems and must be identified and treated to improve quality of life. Sleep health should be an element of all person-centered planning.

The concept of sleep health for persons with intellectual and developmental disabilities is a concept that has not been fully explored. Buysse (2014) has defined the general concept of sleep health as:

> a multidimensional pattern of sleep-wakefulness, adapted to individual, social, and environmental demands, that promotes physical and mental well-being. Good sleep health is characterized by subjective satisfaction, appropriate timing, adequate duration, high efficiency, and sustained alertness during waking hours. (p. 12)

While Buysse's definition of sleep health was coined broadly, this definition is consistent with person-centered planning. Sleep parameters such as satisfaction, duration, and efficacy can be measured for an individual using clinical assessment, questionnaires, sleep diaries, polysomnography, and other methods detailed in this chapter. The clinician can then integrate sleep health needs into care plans. To further refine the sleep health concept, additional research is required regarding sleep in persons with intellectual disability and subpopulations with intellectual disability (Buckles et al., 2013; Richdale & Baker, 2014; Surtees et al., 2018). Sleep diaries and questionnaires should be standardized for subpopulations. Translating research to outcomes for individuals will require multidisciplinary training specifically geared toward patients, caregivers, and clinicians. Providing a thorough assessment of sleep and promoting sleep health should be standard care for all persons with intellectual disability.

References

Abbott, S. M., Reid, K. J., & Zee, P. C. (2015). Circadian rhythm sleep-wake disorders. *Psychiatric Clinics of North America, 38*(4), 805–823. https://doi.org/10.1016/j.psc.2015.07.012

AASM (2014). American Academy of Sleep Medicine International Classification of Sleep Disorders 3rd ed. Darien, IL.

Abbott, S. M., & Zee, P. C. (2015). Irregular sleep-wake rhythm disorder. *Sleep Medicine Clinics, 10*(4), 517–522. https://doi.org/10.1016/j.jsmc.2015.08.005

Adams, H. L., Matson, J. L., Cervantes, P. E., & Goldin, R. L. (2014). The relationship between autism symptom severity and sleep problems: Should bidirectionality be considered? *Research in Autism Spectrum Disorders, 8*(3), 193–199. https://doi.org/10.1016/j.rasd.2013.11.008

Addy, N., Bennett, K., Blanton, A. O., Dort, L., Levine, M., Postol, K., … Smith, H. (2018). Policy statement on a dentist's role in treating sleep-related breathing disorders. *Journal of Dental Sleep Medicine, 5*(1), 25–26.

ADHD: Clinical practice guideline for the diagnosis, evaluation, and treatment of attention-deficit/hyperactivity disorder in children and adolescents. (2011). *Pediatrics,* 128(5), 1007.

Allen, R. P., Picchietti, D. L., Garcia-Borreguero, D., Ondo, W. G., Walters, A. S., Winkelman, J. W., … Lee, H. B. (2014). Restless legs syndrome/Willis–Ekbom disease diagnostic criteria: Updated International Restless Legs Syndrome Study Group (IRLSSG) consensus criteria – history, rationale, description, and significance. *Sleep Medicine, 15*(8), 860–873. https://doi.org/10.1016/j.sleep.2014.03.025

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

Angriman, M., Caravale, B., Novelli, L., Ferri, R., & Bruni, O. (2015). Sleep in children with neurodevel-

opmental disabilities. *Neuropediatrics, 46*(3), 199–210. https://doi.org/10.1055/s-0035-1550151

Annaz, D., Hill, C. M., Ashworth, A., Holley, S., & Karmiloff-Smith, A. (2011). Characterisation of sleep problems in children with Williams syndrome. *Research in Develomental Disabilities, 32*(1), 164–169. https://doi.org/10.1016/j.ridd.2010.09.008

Arbuckle, R., Abetz, L., Durmer, J. S., Ivanenko, A., Owens, J. A., Croenlein, J., … Picchietti, D. L. (2010). Development of the Pediatric Restless Legs Syndrome Severity Scale (P-RLS-SS): A patient-reported outcome measure of pediatric RLS symptoms and impact. *Sleep Medicine, 11*(9), 897–906. https://doi.org/10.1016/j.sleep.2010.03.016

Arroll, B., Fernando, A., Falloon, K., Goodyear-Smith, F., Samaranayake, C., & Warman, G. (2012). Prevalence of causes of insomnia in primary care: A cross-sectional study. *British Journal of General Practice, 62*(595), e99.

Aserinsky, E., & Kleitman, N. (1953). Regularly occurring periods of eye motility, and concomitant phenomena, during Sleep. *Science, 118*(3062), 273–274. Retrieved from http://www.jstor.org/stable/1680525. website.

Baker, E. K., & Richdale, A. L. (2015). Sleep patterns in adults with a diagnosis of high-functioning autism spectrum disorder. *Sleep, 38*(11), 1765–1774. https://doi.org/10.5665/sleep.5160

Baker, E. K., Richdale, A. L., & Hazi, A. (2018). Employment status is related to sleep problems in adults with autism spectrum disorder and no comorbid intellectual impairment. *Autism*, 1362361317745857. https://doi.org/10.1177/1362361317745857

Bandini, L., Danielson, M., Esposito, L. E., Foley, J. T., Fox, M. H., Frey, G. C., … Humphries, K. (2015). Obesity in children with developmental and/or physical disabilities. *Disability and Health Journal, 8*(3), 309–316. https://doi.org/10.1016/j.dhjo.2015.04.005

Barnhill, J. (2006). The assessment and differential diagnosis of insomnia in people with developmental disabilities. *Mental Health Aspects of Developmental Disabilities, 9*(4), 109–118.

Barnhill, J., Soda, T., Poindexter, A., & Hollway, J. A. (2016). Sleep disorders. In R. Fletcher (Ed.), *Diagnostic manual-intellectual disability: A textbook of diagnosis of mental disorders in persons with intellectual disability*. Kingston, NY: NADD.

Bassell, J. L., Phan, H., Leu, R., Kronk, R., & Visootsak, J. (2015). Sleep profiles in children with Down syndrome. *American Journal of Med Genetics Part A, 167a*(8), 1830–1835. https://doi.org/10.1002/ajmg.a.37096

Bastien, C. H., Vallieres, A., & Morin, C. M. (2001). Validation of the Insomnia Severity Index as an outcome measure for insomnia research. *Sleep Medicine, 2*(4), 297–307.

Bhat, S., Ferraris, A., Gupta, D., Mozafarian, M., DeBari, V. A., Gushway-Henry, N., … Chokroverty, S. (2015). Is there a clinical role for smartphone sleep apps? Comparison of sleep cycle detection by a smartphone application to polysomnography. *Journal of Clinical Sleep Medicine: JCSM, 11*(7), 709–715. https://doi.org/10.5664/jcsm.4840

Billiard, M., & Sonka, K. (2016). Idiopathic hypersomnia. *Sleep Medicine Reviews, 29*, 23–33. https://doi.org/10.1016/j.smrv.2015.08.007

Blackmer, A. B., & Feinstein, J. A. (2016). Management of sleep disorders in children with neurodevelopmental disorders: A review. *Pharmacotherapy: The Journal of Human Pharmacology and Drug Therapy, 36*(1), 84–98. https://doi.org/10.1002/phar.1686

Böing, S., & Randerath, W. J. (2015). Chronic hypoventilation syndromes and sleep-related hypoventilation. *Journal of Thoracic Disease, 7*(8), 1273–1285. https://doi.org/10.3978/j.issn.2072-1439.2015.06.10

Bollu, P. C., Goyal, M. K., Thakkar, M. M., & Sahota, P. (2018). Sleep medicine: Parasomnias. *Missouri Medicine, 115*(2), 169–175.

Borbely, A. A., Daan, S., Wirz-Justice, A., & Deboer, T. (2016). The two-process model of sleep regulation: A reappraisal. *Journal of Sleep Research, 25*, 131–143.

Bourgeron, T. (2016). Current knowledge on the genetics of autism and propositions for future research. *Comptes Rendus Biologies, 339*(7), 300–307. https://doi.org/10.1016/j.crvi.2016.05.004

Brady, S. T., Siegel, G. J., Albers, R. W., & Price, D. L. (2012). *Basic neurochemistry principles of molecular, cellular, and medical neurobiology* (pp. xxiv, 1096 p). Retrieved from http://www.sciencedirect.com/science/book/9780123749475

Brown, R. E., Basheer, R., McKenna, J. T., Strecker, R. E., & McCarley, R. W. (2012). Control of sleep and wakefulness. *Physiological Reviews, 92*(3), 1087–1187. https://doi.org/10.1152/physrev.00032.2011

Bruni, O., Ottaviano, S., Guidetti, V., Romoli, M., Innocenzi, M., Cortesi, F., & Giannotti, F. (1996). The Sleep Disturbance Scale for Children (SDSC). Construction and validation of an instrument to evaluate sleep disturbances in childhood and adolescence. *Journal of Sleep Research, 5*(4), 251–261.

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. https://doi.org/10.1080/19315864.2011.651682

Bull, M. J. (2011). Health supervision for children with Down syndrome. *Pediatrics, 128*(2), 393–406. https://doi.org/10.1542/peds.2011-1605

Burger, H. (1929). Ueber das Elektroenkephalogramm des Menschen. *Archiv für Psychiatrie und Nervenkrankheiten, 87*, 527–570. https://doi.org/10.1007/FB01797193

Burgess, H. J., Wyatt, J. K., Park, M., & Fogg, L. F. (2015). Home circadian phase assessments with measures of compliance yield accurate dim light melatonin onsets. *Sleep, 38*(6), 889–897. https://doi.org/10.5665/sleep.4734

Buysse, D. J. (2014). Sleep health: Can we define it? Does it matter? *Sleep, 37*(1), 9–17. https://doi.org/10.5665/sleep.3298

Buysse, D. J., Reynolds, C. F., 3rd, Monk, T. H., Berman, S. R., & Kupfer, D. J. (1989). The Pittsburgh Sleep Quality Index: A new instrument for psychiatric practice and research. *Psychiatry Research, 28*(2), 193–213.

Calhoun, S. L., Fernandez-Mendoza, J., Vgontzas, A. N., Liao, D., & Bixler, E. O. (2014). Prevalence of insomnia symptoms in a general population sample of young children and preadolescents: Gender effects. *Sleep Medicine, 15*(1), 91–95. https://doi.org/10.1016/j.sleep.2013.08.787

Chasens, E. R., Ratcliffe, S. J., & Weaver, T. E. (2009). Development of the FOSQ-10: A short version of the functional outcomes of sleep questionnaire. *Sleep, 32*(7), 915–919. https://doi.org/10.1093/sleep/32.7.915

Chen, M. L., Olson, H. C., Picciano, J. F., Starr, J. R., & Owens, J. (2012). Sleep problems in children with fetal alcohol spectrum disorders. *Journal of Clinical Sleep Medicine: JCSM, 8*(4), 421–429. https://doi.org/10.5664/jcsm.2038

Chervin, R. D., Hedger, K., Dillon, J. E., & Pituch, K. J. (2000). Pediatric sleep questionnaire (PSQ): Validity and reliability of scales for sleep-disordered breathing, snoring, sleepiness, and behavioral problems. *Sleep Medicine, 1*(1), 21–32.

Chung, F., Subramanyam, R., Liao, P., Sasaki, E., Shapiro, C., & Sun, Y. (2012). High STOP-Bang score indicates a high probability of obstructive sleep apnoea. *British Journal of Anaesthesia, 108*(5), 768–775. https://doi.org/10.1093/bja/aes022

Cortelli, P. (2015). Chronomedicine: A necessary concept to manage human diseases. *Sleep Medicine Reviews, 21*, 1–2. https://doi.org/10.1016/j.smrv.2015.01.005

Cortese, S., Ivanenko, A., Ramtekkar, U., & Angriman, M. (2014). Sleep disorders in children and adolescents: A practical guide. In *IACAPAP International Association for Child and Adolescent Psychiatry and Allied Professions Textbook*. International Association for Child and Adolescent Psychiatry and Allied Professions.

Cortese, S., Konofal, E., Lecendreux, M., Arnulf, I., Mouren, M. C., Darra, F., & Dalla Bernardina, B. (2005). Restless legs syndrome and attention-deficit/hyperactivity disorder: A review of the literature. *Sleep, 28*(8), 1007–1013.

Cuthbert, B. N. (2014). The RDoC framework: Facilitating transition from ICD/DSM to dimensional approaches that integrate neuroscience and psychopathology. *World Psychiatry, 13*(1), 28–35. https://doi.org/10.1002/wps.20087

Dale, H. H., Feldberg, W., & Vogt. (1936). Release of acetylcholine at voluntary motor nerve endings. *The Journal of Physiology, 86*, 353–380.

Davis, H., Davis, P. A., Loomis, A. L., Harvey, E. N., & Hobart, G. (1937). Changes in human brain potentials during the onset of sleep. *Science, 86*(2237), 448–450. https://doi.org/10.1126/science.86.2237.448

de Lecea, L., Kilduff, T. S., Peyron, C., Gao, X., Foye, P. E., Danielson, P. E., … Sutcliffe, J. G. (1998). The hypocretins: Hypothalamus-specific peptides with neuroexcitatory activity. *Proceedings of the National Academy of Sciences of the United States of America, 95*(1), 322–327.

de Serres, L. M., Derkay, C., Astley, S., Deyo, R. A., Rosenfeld, R. M., & Gates, G. A. (2000). Measuring quality of life in children with obstructive sleep disorders. *Archives of Otolaryngology and Head and Neck Surgery, 126*(12), 1423–1429.

de Zambotti, M., Goldstone, A., Claudatos, S., Colrain, I. M., & Baker, F. C. (2018). A validation study of Fitbit Charge 2 compared with polysomnography in adults. *Chronobiology International, 35*(4), 465–476. https://doi.org/10.1080/07420528.2017.1413578

Deak, M., & Epstein, L. J. (2009). The history of polysomnography. *Sleep Medicine Clinics, 4*(3), 313–321. https://doi.org/10.1016/j.jsmc.2009.04.001

Dehlink, E., & Tan, H. L. (2016). Update on paediatric obstructive sleep apnoea. *Journal of Thoracic Disease, 8*(2), 224–235. https://doi.org/10.3978/j.issn.2072-1439.2015.12.04

Dement, W., & Kleitman, N. (1957). Cyclic variations in EEG during sleep and their relation to eye movements, body motility, and dreaming. *Electroencephalography and Clinical Neurophysiology, 9*(4), 673–690.

Dosier, L. B. M., Vaughn, B. V., & Fan, Z. (2017). Sleep disorders in childhood neurogenetic disorders. *Children (Basel, Switzerland), 4*(9), 82. https://doi.org/10.3390/children4090082

Douglass, A. B., Bomstein, R., Nino-Murcia, G., Keenan, S., Miles, L., Zarcone, J. V. P., … Dement, W. C. (1994). The sleep disorders questionnaire I: Creation and multivariate structure of SDQ. *Sleep, 17*(2), 160–167. https://doi.org/10.1093/sleep/17.2.160

Drake, C., Nickel, C., Burduvali, E., Roth, T., Jefferson, C., & Pietro, B. (2003). The pediatric daytime sleepiness scale (PDSS): sleep habits and school outcomes in middle-school children. *Sleep, 26*(4), 455–458.

Duffy, J., Zitting, K., & Chinoy, E. (2015). Aging and circadian rhythms. *Sleep Medicine Clinics, 10*, 423–434.

El-Kersh, K., Jungbluth, H., Gringras, P., & Senthilvel, E. (2015). Severe central sleep apnea in Vici syndrome. *Pediatrics, 136*(5), e1390.

Engelhardt, C. R., Mazurek, M. O., & Sohl, K. (2013). Media use and sleep among boys with autism spectrum disorder, ADHD, or typical development. *Pediatrics, 132*(6), 1081–1089. https://doi.org/10.1542/peds.2013-2066

Erichsen, D., Godoy, C., Granse, F., Axelsson, J., Rubin, D., & Gozal, D. (2012). Screening for sleep disorders in pediatric primary care: Are we there yet? *Clinical Pediatrics, 51*(12), 1125–1129. https://doi.org/10.1177/0009922812464548

Esbensen, A. J., & Hoffman, E. K. (2017). Reliability of parent report measures of sleep in children with Down

syndrome. *Journal of Intellectual Disability Research, 61*(3), 210–220. https://doi.org/10.1111/jir.12315

Esbensen, A. J., & Schwichtenberg, A. J. (2016). Sleep in neurodevelopmental disorders. *International Review of Research in Developmental Disabilities, 51*, 153–191. https://doi.org/10.1016/bs.irrdd.2016.07.005

Esposito, M., & Carotenuto, M. (2013). Intellectual disabilities and power spectra analysis during sleep: A new perspective on borderline intellectual functioning. *Journal of Intellectual Disability Research, 58*(5), 421–429. https://doi.org/10.1111/jir.12036

Fahey, M. C., Maclennan, A. H., Kretzschmar, D., Gecz, J., & Kruer, M. C. (2017). The genetic basis of cerebral palsy. *Developmental Medicine and Child Neurology, 59*(5), 462–469. https://doi.org/10.1111/dmcn.13363

Fletcher, R., Barnhill, J., & McCarthy, S.-A. (2016). *Diagnostic manual-intellectual disability (DM-ID): A clinical guide for diagnosis of mental disorders in persons with intellectual disability* (2nd ed.). Kingston, NY: NADD.

Flynn-Evans, E. E., Tabandeh, H., Skene, D. J., & Lockley, S. W. (2014). Circadian rhythm disorders and melatonin production in 127 blind women with and without light perception. *Journal of Biological Rhythms, 29*(3), 215–224. https://doi.org/10.1177/0748730414536852

Ford, E. S., Cunningham, T. J., Giles, W. H., & Croft, J. B. (2015). Trends in insomnia and excessive daytime sleepiness among U.S. adults from 2002 to 2012. *Sleep Medicine, 16*(3), 372–378. https://doi.org/10.1016/j.sleep.2014.12.008

Ghergan, A., Coupaye, M., Leu-Semenescu, S., Attali, V., Oppert, J. M., Arnulf, I., … Redolfi, S. (2017). Prevalence and phenotype of sleep disorders in 60 adults with Prader-Willi syndrome. *Sleep, 40*(12). https://doi.org/10.1093/sleep/zsx162

Gillett, E. S., & Perez, I. A. (2016). Disorders of sleep and ventilatory control in Prader-Willi syndrome. *Diseases (Basel, Switzerland), 4*(3), 23. https://doi.org/10.3390/diseases4030023

Goldman, S. E., Richdale, A. L., Clemons, T., & Malow, B. A. (2012). Parental sleep concerns in autism spectrum disorders: Variations from childhood to adolescence. *Journal of Autism and Developmental Disorders, 42*(4), 531–538. https://doi.org/10.1007/s10803-011-1270-5

Gregory, A. M., & Sadeh, A. (2016). Annual research review: Sleep problems in childhood psychiatric disorders--a review of the latest science. *The Journal of Child Psychology and Psychiatry, 57*(3), 296–317. https://doi.org/10.1111/jcpp.12469

Grigg-Damberger, M., & Rawls, F. (2013). Treatment strategies for complex behavioral insomnia in children with neurodevelopmental disorders. *Current Opinion in Pulmonary Medicine, 19*(6), 616–625. https://doi.org/10.1097/MCP.0b013e328365ab89

Guilleminault, C., & Pelayo, R. (2000). Narcolepsy in children: A practical guide to its diagnosis, treatment and follow-up. *Paediatic Drugs, 2*(1), 1–9. https://doi.org/10.2165/00148581-200002010-00001

Han, F., Lin, L., Warby, S. C., Faraco, J., Li, J., Dong, S. X., … Mignot, E. (2011). Narcolepsy onset is seasonal and increased following the 2009 H1N1 pandemic in China. *Annauls of Neurology, 70*(3), 410–417. https://doi.org/10.1002/ana.22587

Harris, S. R. (2008). Congenital hypotonia: Clinical and developmental assessment. *Developmental Medicine and Child Neurology, 50*(12), 889–892. https://doi.org/10.1111/j.1469-8749.2008.03097.x

Havercamp, S. M., & Scott, H. M. (2015). National health surveillance of adults with disabilities, adults with intellectual and developmental disabilities, and adults with no disabilities. *Disability Health Journal, 8*(2), 165–172. https://doi.org/10.1016/j.dhjo.2014.11.002

Henderson, J. A., Barry, T. D., Bader, S. H., & Jordan, S. S. (2011). The relation among sleep, routines, and externalizing behavior in children with an autism spectrum disorder. *Research in Autism Spectrum Disorders, 5*(2), 758–767. https://doi.org/10.1016/j.rasd.2010.09.003

Henderson, J. A., & Jordan, S. S. (2010). Development and preliminary evaluation of the bedtime routines questionnaire. *Journal of Psychopathology and Behavioral Assessment, 32*(2), 271–280. https://doi.org/10.1007/s10862-009-9143-3

Herice, C., Patel, A. A., & Sakata, S. (2018). Circuit mechanisms and computational models of REM sleep. *Neuroscience Research*. https://doi.org/10.1016/j.neures.2018.08.003

Hersh, J. H., & Saul, R. A. (2011). Health supervision for children with fragile X syndrome. *Pediatrics, 127*(5), 994–1006. https://doi.org/10.1542/peds.2010-3500

Hill, C. M., Evans, H. J., Elphick, H., Farquhar, M., Pickering, R. M., Kingshott, R., … Gringras, P. (2016). Prevalence and predictors of obstructive sleep apnoea in young children with Down syndrome. *Sleep Medicine, 27–28*, 99–106. https://doi.org/10.1016/j.sleep.2016.10.001

Hodge, D., Carollo, T. M., Lewin, M., Hoffman, C. D., & Sweeney, D. P. (2014). Sleep patterns in children with and without autism spectrum disorders: Developmental comparisons. *Research in Developmental Disability, 35*(7), 1631–1638. https://doi.org/10.1016/j.ridd.2014.03.037

Hoffman, C. D., Sweeney, D. P., Lopez-Wagner, M. C., Hodge, D., Nam, C. Y., & Botts, B. H. (2008). Children with autism: Sleep problems and mothers' stress. *Focus on Autism and Other Developmental Disabilities, 23*(3), 155–165. https://doi.org/10.1177/1088357608316271

Hoffmire, C. A., Magyar, C. I., Connolly, H. V., Fernandez, I. D., & van Wijngaarden, E. (2014). High prevalence of sleep disorders and associated comorbidities in a community sample of children with Down syndrome. *Journal of Clinical Sleep Medicine, 10*(4), 411–419. https://doi.org/10.5664/jcsm.3618

Holst, S. C., & Landolt, H.-P. (2018). Sleep-wake neurochemistry. *Sleep Medicine Clinics, 13*, 137–146. Retrieved from https://doi.org/10.1016/j.jsmc.2018.03.002 website.

Horwood, L., Mok, E., Li, P., Oskoui, M., Shevell, M., & Constantin, E. (2018). Prevalence of sleep problems and sleep-related characteristics in preschool- and school-aged children with cerebral palsy. *Sleep Medicine, 50*, 1–6. https://doi.org/10.1016/j.sleep.2018.05.008

Humphreys, J. S., Gringras, P., Blair, P. S., Scott, N., Henderson, J., Fleming, P. J., & Emond, A. M. (2014). Sleep patterns in children with autistic spectrum disorders: A prospective cohort study. *Archives of Disease in Childhood, 99*(2), 114–118. https://doi.org/10.1136/archdischild-2013-304083

Ibáñez, V., Silva, J., & Cauli, O. (2018a). A survey on sleep questionnaires and diaries. *Sleep Medicine, 42*, 90–96. https://doi.org/10.1016/j.sleep.2017.08.026

Ibáñez, V., Silva, J., & Cauli, O. (2018b). A survey on sleep assessment methods. *Peer J, 6*, e4849–e4849. https://doi.org/10.7717/peerj.4849

International Classification of Sleep Disorders. (2014). (3rd ed.). Darien, IL: American Academy of Sleep Medicine.

Jan, J. E., Bax, M. C. O., Owens, J. A., Ipsiroglu, O. S., & Wasdell, M. B. (2012). Neurophysiology of circadian rhythm sleep disorders of children with neurodevelopmental disabilities. *European Journal of Paediatric Neurology, 16*(5), 403–412. https://doi.org/10.1016/j.ejpn.2012.01.002

Jan, J. E., Owens, J. A., Weiss, M. D., Johnson, K. P., Wasdell, M. B., Freeman, R. D., & Ipsiroglu, O. S. (2008). Sleep hygiene for children with neurodevelopmental disabilities. *Pediatrics, 122*(6), 1343.

Janssen, K. C., Phillipson, S., O'Connor, J., & Johns, M. W. (2017). Validation of the Epworth sleepiness scale for children and adolescents using Rasch analysis. *Sleep Medicine, 33*, 30–35. https://doi.org/10.1016/j.sleep.2017.01.014

Javaheri, S. (2010). Central sleep apnea. *Clinics in Chest Medicine, 31*(2), 235–248. https://doi.org/10.1016/j.ccm.2010.02.013

Javaheri, S., Barbe, F., Campos-Rodriguez, F., Dempsey, J. A., Khayat, R., Javaheri, S., … Somers, V. K. (2017). Sleep apnea: Types, mechanisms, and clinical cardiovascular consequences. *Journal of the American College of Cardiology, 69*(7), 841–858. https://doi.org/10.1016/j.jacc.2016.11.069

Johns, M. W. (1991). A new method for measuring daytime sleepiness: The Epworth sleepiness scale. *Sleep, 14*(6), 540–545.

Johnson, C. R., DeMand, A., Lecavalier, L., Smith, T., Aman, M., Foldes, E., & Scahill, L. (2016). Psychometric properties of the children's sleep habits questionnaire in children with autism spectrum disorder. *Sleep Medicine, 20*, 5–11. https://doi.org/10.1016/j.sleep.2015.12.005

Kapur, V. K., Auckley, D. H., Chowdhuri, S., Kuhlmann, D. C., Mehra, R., Ramar, K., & Harrod, C. G. (2017). Clinical practice guideline for diagnostic testing for adult obstructive sleep apnea: An American Academy of Sleep Medicine clinical practice guideline. *Journal of Clinical Sleep Medicine, 13*(3), 479–504. https://doi.org/10.5664/jcsm.6506

Katz, S. L., Gaboury, I., Keilty, K., Banwell, B., Vajsar, J., Anderson, P., … Maclusky, I. (2010). Nocturnal hypoventilation: Predictors and outcomes in childhood progressive neuromuscular disease. *Archives of Disease in Childhood, 95*(12), 998–1003. https://doi.org/10.1136/adc.2010.182709

Kidd, S. A., Lachiewicz, A., Barbouth, D., Blitz, R. K., Delahunty, C., McBrien, D., … Berry-Kravis, E. (2014). Fragile X syndrome: A review of associated medical problems. *Pediatrics, 134*(5), 995–1005. https://doi.org/10.1542/peds.2013-4301

Köse, S., Yılmaz, H., Ocakoğlu, F. T., & Özbaran, N. B. (2017). Sleep problems in children with autism spectrum disorder and intellectual disability without autism spectrum disorder. *Sleep Medicine, 40*, 69–77. https://doi.org/10.1016/j.sleep.2017.09.021

Kotagal, S. (2018). Treatment of narcolepsy and other organic hypersomnias in children. *Paediatric Respiratory Reviews, 25*, 19–24. https://doi.org/10.1016/j.prrv.2017.06.012

Kotagal, S., & Broomall, E. (2012). Sleep in children with autism spectrum disorder. *Pediatric Neurololgy, 47*(4), 242–251. https://doi.org/10.1016/j.pediatrneurol.2012.05.007

Kovachy, B., O'Hara, R., Hawkins, N., Gershon, A., Primeau, M. M., Madej, J., & Carrion, V. (2013). Sleep disturbance in pediatric PTSD: Current findings and future directions. *Journal of Clinical Sleep Medicine: JCSM, 9*(5), 501–510. https://doi.org/10.5664/jcsm.2678

Kronk, R., Bishop, E. E., Raspa, M., Bickel, J. O., Mandel, D. A., & Bailey, D. B., Jr. (2010). Prevalence, nature, and correlates of sleep problems among children with fragile X syndrome based on a large scale parent survey. *Sleep, 33*(5), 679–687.

Kryger, M. H., Roth, T., & Dement, W. C. (2017). *Principles and practice of sleep medicine*. (pp. 1 online resource (li, 1678, 1674 p)). Retrieved from https://www.clinicalkey.com/dura/browse/bookChapter/3-s2.0-C20120035430

Kumar, S., & Sagili, H. (2014). Etiopathogenesis and neurobiology of narcolepsy: A review. *Journal of Clinical and Diagnostic Research : JCDR, 8*(2), 190–195. https://doi.org/10.7860/JCDR/2014/7295.4057

Laberge, L., Tremblay, R. E., Vitaro, F., & Montplaisir, J. (2000). Development of parasomnias from childhood to early adolescence. *Pediatrics, 106*(1 Pt 1), 67–74.

Lane, R., Kessler, R., Buckley, A. W., Rodriguez, A., Farmer, C., Thurm, A., … Felt, B. (2015). Evaluation of periodic limb movements in sleep and iron status in children with autism. *Pediatric Neurology, 53*(4), 343–349. https://doi.org/10.1016/j.pediatrneurol.2015.06.014

Lelis, A. L., Cardoso, M. V., & Hall, W. A. (2016). Sleep disorders in children with cerebral palsy: An integrative review. *Sleep Medicine Reviews, 30*, 63–71. https://doi.org/10.1016/j.smrv.2015.11.008

Leschziner, G. D., Golding, J. F., & Ferner, R. E. (2013). Sleep disturbance as part of the neurofibromatosis type 1 phenotype in adults. *American Journal of Medical Genetics Part A, 161a*(6), 1319–1322. https://doi.org/10.1002/ajmg.a.35915

Liblau, R. S., Vassalli, A., Seifinejad, A., & Tafti, M. (2015). Hypocretin (orexin) biology and the pathophysiology of narcolepsy with cataplexy. *The Lancet Neurology, 14*(3), 318–328. https://doi.org/10.1016/S1474-4422(14)70218-2

Licis, A. K., Desruisseau, D. M., Yamada, K. A., Duntley, S. P., & Gurnett, C. A. (2011). Novel genetic findings in an extended family pedigree with sleepwalking. *Neurology, 76*(1), 49–52. https://doi.org/10.1212/WNL.0b013e318203e964

Licis, A. K., Vallorani, A., Gao, F., Chen, C., Lenox, J., Yamada, K. A., … Gutmann, D. H. (2013). Prevalence of sleep disturbances in children with neurofibromatosis type 1. *Journal of Child Neurology, 28*(11), 1400–1405. https://doi.org/10.1177/0883073813500849

Maas, A. P., Didden, R., Korzilius, H., Braam, W., Collin, P., Smits, M. G., & Curfs, L. M. (2011). Psychometric properties of a sleep questionnaire for use in individuals with intellectual disabilities. *Research in Developmental Disability, 32*(6), 2467–2479. https://doi.org/10.1016/j.ridd.2011.07.013

Maas, A. P. H. M., Sinnema, M., Didden, R., Maaskant, M. A., Smits, M. G., Schrander-Stumpel, C. T. R. M., & Curfs, L. M. G. (2010). Sleep disturbances and behavioural problems in adults with Prader–Willi syndrome. *Journal of Intellectual Disability Research, 54*(10), 906–917. https://doi.org/10.1111/j.1365-2788.2010.01306.x

McCandless, S. E. (2010). Clinical report—health supervision for children with Prader-Willi syndrome. Pediatrics.

MacLean, J. E., Fitzgerald, D. A., & Waters, K. A. (2015). Developmental changes in sleep and breathing across infancy and childhood. *Paediatric Respiratory Reviews, 16*(4), 276–284. https://doi.org/10.1016/j.prrv.2015.08.002

Maislin, G., Pack, A. I., Kribbs, N. B., Smith, P. L., Schwartz, A. R., Kline, L. R., … Dinges, D. F. (1995). A survey screen for prediction of apnea. *Sleep, 18*(3), 158–166.

Malow, B. A., Byars, K., Johnson, K., Weiss, S., Bernal, P., Goldman, S. E., … Glaze, D. G. (2012). A practice pathway for the identification, evaluation, and management of insomnia in children and adolescents with autism spectrum disorders. *Pediatrics, 130*, S106–S124. https://doi.org/10.1542/peds.2012-0900I

Mander, B. A., Winer, J. R., & Walker, M. P. (2017). Sleep and human aging. *Neuron, 94*(1), 19–36. https://doi.org/10.1016/j.neuron.2017.02.004

Manni, R., Toscano, G., & Terzaghi, M. (2018). Therapeutic symptomatic strategies in the Parasomnias. *Current Treatment Options in Neurology, 20*(7), 26. https://doi.org/10.1007/s11940-018-0508-3

Mason, T. B. A., Arens, R., Sharman, J., Bintliff-Janisak, B., Schultz, B., Walters, A. S., … Pack, A. I. (2011). Sleep in children with Williams syndrome. *Sleep Medicine, 12*(9), 892–897. https://doi.org/10.1016/j.sleep.2011.05.003

Matthey, S. (2001). The sleep and settle questionnaire for parents of infants: Psychometric properties. *Journal of Paediatric Child Health, 37*(5), 470–475.

Mayes, S. D., & Calhoun, S. L. (2009). Variables related to sleep problems in children with autism. *Research in Autism Spectrum Disorders, 3*(4), 931–941. https://doi.org/10.1016/j.rasd.2009.04.002

Mazurek, M. O., & Sohl, K. (2016). Sleep and behavioral problems in children with autism spectrum disorder. *Journal of Autism and Developmental Disorders, 46*(6), 1906–1915. https://doi.org/10.1007/s10803-016-2723-7

McCarter, S. J., Boswell, C. L., St. Louis, E. K., Dueffert, L. G., Slocumb, N., Boeve, B. F., … Tippmann-Peikert, M. (2013). Treatment outcomes in REM sleep behavior disorder. *Sleep Medicine, 14*(3), 237–242. https://doi.org/10.1016/j.sleep.2012.09.018

McGreavey, J. A., Donnan, P. T., Pagliari, H. C., & Sullivan, F. M. (2005). The Tayside children's sleep questionnaire: A simple tool to evaluate sleep problems in young children. *Child: Care, Health and Development, 31*(5), 539–544. https://doi.org/10.1111/j.1365-2214.2005.00548.x

McKenna, H., & Reiss, I. K. M. (2018). The case for a chronobiological approach to neonatal care. *Early Human Development*. https://doi.org/10.1016/j.earlhumdev.2018.08.012

Meltzer, L. J., Phillips, C., & Mindell, J. A. (2009). Clinical psychology training in sleep and sleep disorders. *Journal of Clinical Psychology, 65*(3), 305–318. https://doi.org/10.1002/jclp.20545

Miano, S., Bruni, O., Elia, M., Trovato, A., Smerieri, A., Verrillo, E., … Ferri, R. (2007). Sleep in children with autistic spectrum disorder: A questionnaire and polysomnographic study. *Sleep Medicine, 9*(1), 64–70. https://doi.org/10.1016/j.sleep.2007.01.014

Mindell, J. A., Bartle, A., Ahn, Y., Ramamurthy, M. B., Huong, H. T. D., Kohyama, J., … Goh, D. Y. T. (2013). Sleep education in pediatric residency programs: A cross-cultural look. *BMC Research Notes, 6*, 130–130. https://doi.org/10.1186/1756-0500-6-130

Ming, X., Gordon, E., Kang, N., & Wagner, G. C. (2008). Use of clonidine in children with autism spectrum disorders. *Brain and Development, 30*(7), 454–460. https://doi.org/10.1016/j.braindev.2007.12.007

Mold, J. W., Quattlebaum, C., Schinnerer, E., Boeckman, L., Orr, W., & Hollabaugh, K. (2011). Identification by primary care clinicians of patients with obstructive sleep apnea: A practice-based research network (PBRN) study. *Journal of the American Board of Family Medicine, 24*(2), 138–145. https://doi.org/10.3122/jabfm.2011.02.100095

Montgomery-Downs, H. E., O'Brien, L. M., Holbrook, C. R., & Gozal, D. (2004). Snoring and sleep-

disordered breathing in young children: Subjective and objective correlates. *Sleep, 27*(1), 87–94.

Monti, J. M., & Monti, D. (2007). The involvement of dopamine in the modulation of sleep and waking. *Sleep Medicine Reviews, 11*, 113–133.

Moran, J. (2017). *Aging and down syndrome: A health and well-being guidebook* (p. 40). New York, NY: NDSS.

Morgenthaler, T. I., Auerbach, S., Casey, K. R., Kristo, D., Maganti, R., Ramar, K., … Kartje, R. (2018). Position paper for the treatment of nightmare disorder in adults: An American Academy of Sleep Medicine position paper. *Journal of Clinical Sleep Medicine, 14*(6), 1041–1055. https://doi.org/10.5664/jcsm.7178

Morin, C. (1994, Summer). Dysfunctional beliefs and attitudes about sleep: Preliminary scale development and description. *The Behavior Therapist, 17*, 163–164.

Morin, C. M. (1993). *Insomnia: Psychological assessment and management*. New York, NY: Guilford Press.

Morin, C. M., Vallières, A., & Ivers, H. (2007). Dysfunctional beliefs and attitudes about sleep (DBAS): Validation of a brief version (DBAS-16). *Sleep, 30*(11), 1547–1554.

Morphy, H., Dunn, K. M., Lewis, M., Boardman, H. F., & Croft, P. R. (2007). Epidemiology of insomnia: A longitudinal study in a UK population. *Sleep, 30*(3), 274–280.

Morrell, J., & Cortina-Borja, M. (2002). The developmental change in strategies parents employ to settle young children to sleep, and their relationship to infant sleeping problems, as assessed by a new questionnaire: the Parental Interactive Bedtime Behaviour Scale. *Infant and Child Development, 11*(1), 17–41. https://doi.org/10.1002/icd.251

Morrell, J. M. B. (1999). The role of maternal cognitions in infant sleep problems as assessed by a new instrument, the maternal cognitions about infant sleep questionnaire. *The Journal of Child Psychology and Psychiatry and Allied Disciplines, 40*(2), 247–258.

Morrison, A. R. (2014). Henri Piéron and Nathaniel Kleitman: Two major figures of 20(th) century sleep research. *Sleep, 37*(3), 621–621. https://doi.org/10.5665/sleep.3512

Mouthon, A. L., & Huber, R. (2015). Methods in pediatric sleep research and sleep medicine. *Neuropediatrics, 46*(3), 159–170. https://doi.org/10.1055/s-0035-1550232

Mullegama, S. V., Pugliesi, L., Burns, B., Shah, Z., Tahir, R., Gu, Y., … Elsea, S. H. (2015). MBD5 haploinsufficiency is associated with sleep disturbance and disrupts circadian pathways common to Smith-Magenis and fragile X syndromes. *European Journal of Human Genetics, 23*(6), 781–789. https://doi.org/10.1038/ejhg.2014.200

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), 95–102. https://doi.org/10.1097/YCO.0000000000000236

Myers, S. M., & Johnson, C. P. (2007). Management of children with autism spectrum disorders. *Pediatrics, 120*(5), 1162–1182. https://doi.org/10.1542/peds.2007-2362

Netzer, N. C., Stoohs, R. A., Netzer, C. M., Clark, K., & Strohl, K. P. (1999). Using the berlin questionnaire to identify patients at risk for the sleep apnea syndrome. *Annals of Internal Medicine, 131*(7), 485–491. https://doi.org/10.7326/0003-4819-131-7-199910050-00002

Nevsimalova, S. (2014). The diagnosis and treatment of pediatric narcolepsy. *Current Neurology Neuroscience Reports, 14*(8), 469. https://doi.org/10.1007/s11910-014-0469-1

Niemczyk, J., von Gontard, A., Equit, M., Medoff, D., Wagner, C., & Curfs, L. (2017). Incontinence in persons with Down syndrome. *Neurourology Urodynamics, 36*(6), 1550–1556. https://doi.org/10.1002/nau.23146

Ohayon, M. M., Carskadon, M. A., Guilleminault, C., & Vitiello, M. V. (2004). Meta-analysis of quantitative sleep parameters from childhood to old age in healthy individuals: Developing normative sleep values across the human lifespan. *Sleep, 27*(7), 1255–1273.

Ohayon, M. M., O'Hara, R., & Vitiello, M. V. (2012). Epidemiology of restless legs syndrome: A synthesis of the literature. *Sleep Medicine Reviews, 16*(4), 283–295. https://doi.org/10.1016/j.smrv.2011.05.002

Ong, A. A., & Gillespie, M. B. (2016). Overview of smartphone applications for sleep analysis. *World Journal of Otorhinolaryngology – Head and Neck Surgery, 2*(1), 45–49. https://doi.org/10.1016/j.wjorl.2016.02.001

Owens, J. A., & Dalzell, V. (2005). Use of the 'BEARS' sleep screening tool in a pediatric residents' continuity clinic: A pilot study. *Sleep Medicine, 6*(1), 63–69. https://doi.org/10.1016/j.sleep.2004.07.015

Owens, J. A., Spirito, A., & McGuinn, M. (2000). The Children's Sleep Habits Questionnaire (CSHQ): Psychometric properties of a survey instrument for school-aged children. *Sleep, 23*(8), 1043–1051.

Paasch, V., Leibowitz, L., Accardo, J., & Slifer, K. (2016). Preparing children with autism spectrum disorders for overnight sleep studies: A case series. *Clinical Practice in Pediatric Psychology, 4*(2), 153–163.

Pagel, J. F. (2009). Excessive daytime sleepiness. *American Family Physician, 79*(5), 391–396.

Paine, S. J., Fink, J., Gander, P. H., & Warman, G. R. (2014). Identifying advanced and delayed sleep phase disorders in the general population: A national survey of New Zealand adults. *Chronobiology International, 31*(5), 627–636. https://doi.org/10.3109/07420528.2014.885036

Paksarian, D., Rudolph, K. E., He, J. P., & Merikangas, K. R. (2015). School start time and adolescent sleep patterns: Results from the U.S. National Comorbidity Survey--Adolescent Supplement. *American Journal of Public Health, 105*(7), 1351–1357. https://doi.org/10.2105/ajph.2015.302619

Pallesen, S., Bjorvatn, B., Nordhus, I. H., Sivertsen, B., Hjørnevik, M., & Morin, C. M. (2008). A new scale for measuring insomnia: The Bergen insomnia scale. *Perceptual and Motor Skills, 107*(3), 691–706. https://doi.org/10.2466/pms.107.3.691-706

Paruthi, S., Brooks, L. J., D'Ambrosio, C., Hall, W. A., Kotagal, S., Lloyd, R. M., … Wise, M. S. (2016). Recommended amount of sleep for pediatric populations: A consensus statement of the American Academy of Sleep Medicine. *Journal of Clinical Sleep Medicine, 12*(6), 785–786. https://doi.org/10.5664/jcsm.5866

Peever, J., & Fuller, P. M. (2017). The biology of REM sleep. *Current Biology, 27*(22), R1237–r1248. https://doi.org/10.1016/j.cub.2017.10.026

Pellegrino, R., Kavakli, I. H., Goel, N., Cardinale, C. J., Dinges, D. F., Kuna, S. T., … Pack, A. I. (2014). A novel BHLHE41 variant is associated with short sleep and resistance to sleep deprivation in humans. *Sleep, 37*(8), 1327–1336. https://doi.org/10.5665/sleep.3924

Plazzi, G., Pizza, F., Palaia, V., Franceschini, C., Poli, F., Moghadam, K. K., … Bhatia, K. P. (2011). Complex movement disorders at disease onset in childhood narcolepsy with cataplexy. *Brain, 134*(12), 3480–3492. https://doi.org/10.1093/brain/awr244

Prihodova, I., Dudova, I., Mohaplova, M., Hrdlicka, M., & Nevsimalova, S. (2018). Childhood narcolepsy and autism spectrum disorders: Four case reports. *Sleep Medicine, 51*, 167–170. https://doi.org/10.1016/j.sleep.2018.07.017

Reale, L., Bartoli, B., Cartabia, M., Zanetti, M., Costantino, M. A., Canevini, M. P., … on behalf of Lombardy ADHD Group. (2017). Comorbidity prevalence and treatment outcome in children and adolescents with ADHD. *European Child & Adolescent Psychiatry, 26*(12), 1443–1457. https://doi.org/10.1007/s00787-017-1005-z

Reijnders, M. R., Leventer, R. J., Lee, B. H., Baralle, D., Selber, P., Paciorkowski, A. R., & Hunt, D. (2017). PURA-related neurodevelopmental disorders. *GeneReviews® [Internet].* Retrieved from https://www.ncbi.nlm.nih.gov/books/NBK426063/

Richdale, A. L., & Baker, E. K. (2014). Sleep in individuals with an intellectual or developmental disability: Recent research reports. *Current Developmental Disorder Reports, 1*, 74–85. https://doi.org/10.1007/s40474-014-0010-x

Rocca, F. L., Pizza, F., Ricci, E., & Plazzi, G. (2015). Narcolepsy during childhood: An update. *Neuropediatrics, 46*(3), 181–198. https://doi.org/10.1055/s-0035-1550152

Rosen, D. (2011). Management of obstructive sleep apnea associated with Down syndrome and other craniofacial dysmorphologies. *Current Opinion in Pulmonary Medicine, 17*(6), 431–436. https://doi.org/10.1097/MCP.0b013e32834ba9c0

Rosenwasser, A. M., & Turek, F. W. (2015). Neurobiology of circadian rhythm regulation. *Sleep Medicine Clinics, 10*(December), 403–412.

Ruoff, C., & Rye, D. (2016). The ICSD-3 and DSM-5 guidelines for diagnosing narcolepsy: Clinical relevance and practicality. *Current Medical Research and Opinion, 32*(10), 1611–1622. https://doi.org/10.1080/03007995.2016.1208643

Sack, R. L., Auckley, D., Auger, R. R., Carskadon, M. A., Wright, J. K. P., Vitiello, M. V., & Zhdanova, I. V. (2007). Circadian rhythm sleep disorders: Part I, basic principles, shift work and jet lag disorders. *Sleep, 30*(11), 1460–1483. https://doi.org/10.1093/sleep/30.11.1460

Sadeh, A. (2004). A brief screening questionnaire for infant sleep problems: Validation and findings for an Internet sample. *Pediatrics, 113*(6), e570–e577. https://doi.org/10.1542/peds.113.6.e570

Sandella, D. E., O'Brien, L. M., Shank, L. K., & Warschausky, S. A. (2011). Sleep and quality of life in children with cerebral palsy. *Sleep Medicine, 12*(3), 252–256. https://doi.org/10.1016/j.sleep.2010.07.019

Saper, C. B., & Fuller, P. M. (2017). Wake-sleep circuitry: An overview. *Current Opinion in Neurobiology, 44*, 186–192.

Saxvig, I. W., Pallesen, S., Wilhelmsen-Langeland, A., Molde, H., & Bjorvatn, B. (2012). Prevalence and correlates of delayed sleep phase in high school students. *Sleep Medicine, 13*(2), 193–199. https://doi.org/10.1016/j.sleep.2011.10.024

Schreck, K. A., Mulick, J. A., & Rojahn, J. (2003). Development of the behavioral evaluation of disorders of sleep scale. *Journal of Child and Family Studies, 12*(3), 349–359.

Schutte-Rodin, S., Broch, L., Buysse, D., Dorsey, C., & Sateia, M. (2008). Clinical guideline for the evaluation and management of chronic insomnia in adults. *Journal of Clinical Sleep Medicine: JCSM, 4*(5), 487–504.

Schwartz, M. D., & Kilduff, T. S. (2015). The neurobiology of sleep and wakefulness. *Psychiatric Clinics of North America, 38*, 615–644. https://doi.org/10.1016/j.psc.2015.07.002

Sedky, K., Bennett, D. S., & Pumariega, A. (2014). Prader Willi syndrome and obstructive sleep apnea: Co-occurrence in the pediatric population. *Journal of Clinical Sleep Medicine: JCSM, 10*(4), 403–409. https://doi.org/10.5664/jcsm.3616

Serdarevic, F., Ghassabian, A., van Batenburg-Eddes, T., White, T., Blanken, L. M. E., Jaddoe, V. W. V., … Tiemeier, H. (2017). Infant muscle tone and childhood autistic traits: A longitudinal study in the general population. *Autism Research, 10*(5), 757–768. https://doi.org/10.1002/aur.1739

Sharanah, R., Swarnalata, D., Racha, N., David, S. B., & Karim, S. (2017). Obstructive sleep apnea in individuals with down syndrome: A meta-analytic literature review. *Journal of Sleep and Sleep Disorder Research, 1*(2), 1–15. https://doi.org/10.14302/issn.2574-4518.jsdr-17-1754

Shrivastava, D., Jung, S., Saadat, M., Sirohi, R., & Crewson, K. (2014). How to interpret the results of a sleep study. *Journal of Community Hospital Internal Medicine Perspectives, 4*(5), 24983. https://doi.org/10.3402/jchimp.v4.24983

Simonds, J. F., & Parraga, H. (1982). Prevalence of sleep disorders and sleep behaviors in children and adoles-

cents. *Journal of the American Academy of Child and Adolescent Psychiatry, 21*(4), 383–388.

Skeldon, A. C., Derks, G., & Dijk, D. J. (2016). Modelling changes in sleep timing and duration across the lifespan: Changes in circadian rhythmicity or sleep homeostasis? *Sleep Medicine Reviews, 28*, 96–107. https://doi.org/10.1016/j.smrv.2015.05.011

Smets, E. M. A., Garssen, B., Bonke, B., & De Haes, J. C. J. M. (1995). The multidimensional fatigue inventory (MFI) psychometric qualities of an instrument to assess fatigue. *Journal of Psychosomatic Research, 39*(3), 315–325. https://doi.org/10.1016/0022-3999(94)00125-O

Smith, M. T., McCrae, C. S., Cheung, J., Martin, J. L., Harrod, C. G., Heald, J. L., & Carden, K. A. (2018). Use of actigraphy for the evaluation of sleep disorders and circadian rhythm sleep-wake disorders: An American Academy of Sleep Medicine systematic review, meta-analysis, and GRADE assessment. *Journal of Clinical Sleep Medicine: JCSM, 14*(7), 1209–1230. https://doi.org/10.5664/jcsm.7228

Sniecinska-Cooper, A., Iles, R., Butler, D. S., Jones, H., Bayford, R., & Dimitriou, D. (2014). Abnormal secretion of melatonin and cortisol in relation to sleep disturbances in children with Williams syndrome. *Sleep Medicine, 16*(1), 94–100.

Soldatos, C. R., Dikeos, D. G., & Paparrigopoulos, T. J. (2000). Athens insomnia scale: Validation of an instrument based on ICD-10 criteria. *Journal of Psychosomatic Research, 48*(6), 555–560. https://doi.org/10.1016/S0022-3999(00)00095-7

Sollars, P. J., & Pickard, G. E. (2015). The neurobiology of circadian rhythms. *Psychiatric Clinics, 38*(4), 645–665. https://doi.org/10.1016/j.psc.2015.07.003

Sorscher, A. J. (2011). Sleep and the family doctor: Time to lead. *Journal of the American Board of Family Medicine, 24*(2), 133–135. https://doi.org/10.3122/jabfm.2011.02.110014

Sowa, N. A. (2016). Idiopathic hypersomnia and hypersomnolence disorder: A systematic review of the literature. *Psychosomatics, 57*(2), 152–164. https://doi.org/10.1016/j.psym.2015.12.006

Spilsbury, J. C., Drotar, D., Rosen, C. L., & Redline, S. (2007). The Cleveland adolescent sleepiness questionnaire: A new measure to assess excessive daytime sleepiness in adolescents. *Journal of Clinical Sleep Medicine, 3*(6), 603–612.

Spruyt, K., Braam, W., Smits, M., & Curfs, L. M. (2016). Sleep complaints and the 24-h melatonin level in individuals with Smith-Magenis syndrome: Assessment for effective intervention. *CNS Neuroscience and Therapeutics, 22*(11), 928–935. https://doi.org/10.1111/cns.12653

Spruyt, K., & Gozal, D. (2011). Pediatric sleep questionnaires as diagnostic or epidemiological tools: A review of currently available instruments. *Sleep Medicine Reviews, 15*(1), 19–32. https://doi.org/10.1016/j.smrv.2010.07.005

Stahl, S. M. (2013). *Stahl's essential psychopharmacology* (4th ed.). Cambridge, UK: Cambridge University Press.

Stallman, H. M., Kohler, M., & White, J. (2018). Medication induced sleepwalking: A systematic review. *Sleep Medicine Reviews, 37*, 105–113. https://doi.org/10.1016/j.smrv.2017.01.005

Staton, S. L., Smith, S. S., Hurst, C., Pattinson, C. L., & Thorpe, K. J. (2017). Mandatory nap times and group napping patterns in child care: An observational study. *Behavioral Sleep Medicine, 15*(2), 129–143. https://doi.org/10.1080/15402002.2015.1120199

Stewart, D. R., Korf, B. R., Nathanson, K. L., Stevenson, D. A., & Yohay, K. (2018). Care of adults with neurofibromatosis type 1: A clinical practice resource of the American College of Medical Genetics and Genomics (ACMG). *Genetics in Medicine, 20*(7), 671–682. https://doi.org/10.1038/gim.2018.28

Stores, G. (2014). *Sleep and its disorders in children and adolescents with a neurodevelopmental disorder: A review and clinical guide*. Cambridge, UK: Cambridge University Press.

Stores, G. (2016). Multifactorial influences, including comorbidities, contributing to sleep disturbance in children with a neurodevelopmental disorder. *CNS Neuroscience & Therapeutics, 22*(11), 875–879. https://doi.org/10.1111/cns.12574

Strecker, R. E., Morairty, S., Thakkar, M. M., Porkka-Heiskanen, T., Basheer, R., Dauphin, L. J., … McCarley, R. W. (2000). Adenosinergic modulation of basal forebrain and preoptic/anterior hypothalamic neuronal activity in the control of behavioral state. *Behavioural Brain Research, 115*(2), 183–204.

Summers, S. M., Cogswell, J., Goodrich, J. E., Mu, Y., Nguyen, D. V., Brass, S. D., & Hagerman, R. J. (2014). Prevalence of restless legs syndrome and sleep quality in carriers of the fragile X premutation. *Clinical Genetics, 86*(2), 181–184.

Sung, V., Hiscock, H., Sciberras, E., & Efron, D. (2008). Sleep problems in children with attention-deficit/hyperactivity disorder: Prevalence and the effect on the child and family. *Archives of Pediatric and Adolescent Medicine, 162*(4), 336–342. https://doi.org/10.1001/archpedi.162.4.336

Surtees, A. D. R., Oliver, C., Jones, C. A., Evans, D. L., & Richards, C. (2018). Sleep duration and sleep quality in people with and without intellectual disability: A meta-analysis. *Sleep Medicine Reviews, 40*, 135–150. https://doi.org/10.1016/j.smrv.2017.11.003

Szelenberger, W., Niemcewicz, S., & DĄbrowska, A. J. (2005). Sleepwalking and night terrors: Psychopathological and psychophysiological correlates. *International Review of Psychiatry, 17*(4), 263–270. https://doi.org/10.1080/09540260500104573

Szuhay, G., & Rotenberg, J. (2009). Sleep apnea in pediatric neurological conditions. *Current Neurology and Neuroscience Reports, 9*(2), 145–152. https://doi.org/10.1007/s11910-009-0023-8

Tamura, N., Sasai-Sakuma, T., Morita, Y., Okawa, M., Inoue, S., & Inoue, Y. (2016). A nationwide cross-sectional survey of sleep-related problems in Japanese visually impaired patients: Prevalence and association with health-related quality of life. *Journal of Clinical Sleep Medicine, 12*(12), 1659–1667. https://doi.org/10.5664/jcsm.6354

Taylor, M. A., Schreck, K. A., & Mulick, J. A. (2012). Sleep disruption as a correlate to cognitive and adaptive behavior problems in autism spectrum disorders. *Research in Developmental Disability, 33*(5), 1408–1417. https://doi.org/10.1016/j.ridd.2012.03.013

Thomas, R., Sanders, S., Doust, J., Beller, E., & Glasziou, P. (2015). Prevalence of attention-deficit/hyperactivity disorder: A systematic review and meta-analysis. *Pediatrics, 135*(4), e994.

Thorpy, M. J. (2012). Classification of sleep disorders. *Neurotherapeutics, 9*(4), 687–701. https://doi.org/10.1007/s13311-012-0145-6

Toh, K. L., Jones, C. R., He, Y., Eide, E. J., Hinz, W. A., Virshup, D. M., … Fu, Y. H. (2001). An hPer2 phosphorylation site mutation in familial advanced sleep phase syndrome. *Science, 291*(5506), 1040–1043.

Tosini, G., Ferguson, I., & Tsubota, K. (2016). Effects of blue light on the circadian system and eye physiology. *Molecular Vision, 22*, 61–72.

Trickett, J., Richards, C., Heald, M., & Oliver, C. (2016). Describing sleep quality in children with Angelman syndrome and Smith-Magenis syndrome. *Journal of Intellectual Disability Research, 60*(7–8).

Trenkwalder, C., Allen, R., Högl, B., Paulus, W., & Winkelmann, J. (2016). Restless legs syndrome associated with major diseases: A systematic review and new concept. *Neurology, 86*(14), 1336–1343. https://doi.org/10.1212/WNL.0000000000002542

Trotti, L. M. (2017). Idiopathic hypersomnia. *Sleep Medicine Clinics, 12*(3), 331–344. https://doi.org/10.1016/j.jsmc.2017.03.009

Vissers, L. E., Gilissen, C., & Veltman, J. A. (2016). Genetic studies in intellectual disability and related disorders. *National Review of Genetics, 17*(1), 9–18. https://doi.org/10.1038/nrg3999

von Gontard, A., & Equit, M. (2015). Comorbidity of ADHD and incontinence in children. *European Child and Adolescent Psychiatry, 24*(2), 127–140. https://doi.org/10.1007/s00787-014-0577-0

Wagner, C., Niemczyk, J., Equit, M., Curfs, L., & von Gontard, A. (2017). Incontinence in persons with Angelman syndrome. *European Journal of Pediatrics, 176*(2), 225–232. https://doi.org/10.1007/s00431-016-2828-1

Wakefield, S. M., Sanderson, J., & McPherson, P. (2018). Assessment of obesity. In J. L. Matson (Ed.), *Handbook of childhood psychopathology and developmental disabilities assessment* (pp. 433–452). Cham, Switzerland: Springer International Publishing.

Walz, N. C., Beebe, D., & Byars, K. (2005). Sleep in individuals with Angelman syndrome: Parent perceptions of patterns and problems. *American Journal of Mental Retardation, 110*(4), 243–252. https://doi.org/10.1352/0895-8017(2005)110[243:siiwas]2.0.co;2

Wamsley, E., Donjacour, C. E. H. M., Scammell, T. E., Lammers, G. J., & Stickgold, R. (2014). Delusional confusion of dreaming and reality in narcolepsy. *Sleep, 37*(2), 419–422. https://doi.org/10.5665/sleep.3428

Ward Flemons, W., & Reimer, M. A. (1998). Development of a disease-specific health-related quality of life questionnaire for sleep apnea. *American Journal of Respiratory and Critical Care Medicine, 158*(2), 494–503. https://doi.org/10.1164/ajrccm.158.2.9712036

Watson, N. F., Badr, M. S., Belenky, G., Bliwise, D. L., Buxton, O. M., Buysse, D., … Tasali, E. (2015). Recommended amount of sleep for a healthy adult: A joint consensus statement of the American Academy of Sleep Medicine and Sleep Research Society. *Sleep, 38*(6), 843–844. https://doi.org/10.5665/sleep.4716

Wayte, S., McCaughey, E., Holley, S., Annaz, D., & Hill, C. M. (2012). Sleep problems in children with cerebral palsy and their relationship with maternal sleep and depression. *Acta Paediatrica, 101*(6), 618–623. https://doi.org/10.1111/j.1651-2227.2012.02603.x

Weaver, M. D., Barger, L. K., Malone, S. K., Anderson, L. S., & Klerman, E. B. (2018). Dose-dependent associations between sleep duration and unsafe behaviors among US high school students. *JAMA Pediatrics*. https://doi.org/10.1001/jamapediatrics.2018.2777

Weaver, T. E., Laizner, A. M., Evans, L. K., Maislin, G., Chugh, D. K., Lyon, K., … Dinges, D. E. (1997). An instrument to measure functional status outcomes for disorders of excessive sleepiness. *Sleep, 20*(10), 835–843. https://doi.org/10.1093/sleep/20.10.835

Weber, F., & Dan, Y. (2016). Circuit-based interrogation of sleep control. *Nature, 538*(7623), 51–59. https://doi.org/10.1038/nature19773

Werner, H., Molinari, L., Guyer, C., & Jenni, O. G. (2008). Agreement rates between actigraphy, diary, and questionnaire for children's sleep patterns. *Archives of Pediatrics & Adolescent Medicine, 162*(4), 350–358. https://doi.org/10.1001/archpedi.162.4.350

Williams, C. A., Beaudet, A. L., Clayton-Smith, J., Knoll, J. H., Kyllerman, M., Laan, L. A., … Wagstaff, J. (2006). Angelman syndrome 2005: Updated consensus for diagnostic criteria. *American Journal of Medical Genetics Part A, 140*(5), 413–418. https://doi.org/10.1002/ajmg.a.31074

Wilson, G., Terpening, Z., Wong, K., Grunstein, R., Norrie, L., Lewis, S. J. G., & Naismith, S. L. (2014). Screening for sleep apnoea in mild cognitive impairment: The utility of the multivariable apnoea prediction index. *Sleep Disorders, 2014*, 945287. https://doi.org/10.1155/2014/945287

Winkelmann, J., Schormair, B., Xiong, L., Dion, P. A., Rye, D. B., & Rouleau, G. A. (2017). Genetics of restless legs syndrome. *Sleep Medicine, 31*, 18–22. https://doi.org/10.1016/j.sleep.2016.10.012

Wong, S.-H., & Ng, B.-Y. (2015). Review of sleep studies of patients with chronic insomnia at a sleep disorder unit. *Singapore Medical Journal, 56*(6), 317–323. https://doi.org/10.11622/smedj.2015089

World Health Organization. (2007). *International statistical classification of diseases and related health problems* (10th Revision ed.). Geneva, Switzerland: World Health Organization.

Xu, Y., Padiath, Q. S., Shapiro, R. E., Jones, C. R., Wu, S. C., Saigoh, N., … Fu, Y.-H. (2005). Functional consequences of a CKIδ mutation causing familial advanced sleep phase syndrome. *Nature, 434*, 640. https://doi.org/10.1038/nature03453. https://www.nature.com/articles/nature03453#supplementary-information

Youssef, N. A., Ege, M., Angly, S. S., Strauss, J. L., & Marx, C. E. (2011). Is obstructive sleep apnea associated with ADHD? *Annals of Clinical Psychiatry, 23*(3), 213–224.

Zavrel, E., Zadeh, S., Eshelman, R., Chen, P., Davaji, B., Lal, A., … Krieger, A. (2018). *Clinical validation of the program for improving and managing the environment sleep monitoring system*. Paper presented at the Biomedical Engineering Society Annual Conference, Atlanta, GA.

Zero to Three (2016). Diagnostic classification of mental health and developmental disorders of infancy and early childhood: Revised edition DC: 0-5. ZERO TO THREE Press, Washington, DC.

Noncompliance in Dual Disorders

24

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The Merriam-Webster online dictionary defines noncompliance as "failure or refusal to comply with something (such as a rule or regulation): a state of not being in compliance" (https://www.merriam-webster.com/dictionary/noncompliance#other-words). A more clinical definition is "Non-compliance is used to describe when an individual does not or refuses to follow the directions, rules or wishes of someone else. Non-compliance can be passive, such as not following a direction, or active, such as whining/crying, or becoming aggressive or self-injurious. It is helpful to remember that non-compliance can be purposeful, but at times can also result from lack of understanding, lack of motivation, fatigue, or poor organizational or motor planning issues" (Autism Speaks, 2012, p. 9). Specific to children, it has also been defined as doing anything other than what has been requested by a parent or other adult authority figure within a specific time frame (Kalb & Loeber, 2003). Noncompliance is an important behavior on which to focus as it is related to several psychiatric disorders (Kalb & Loeber, 2003), parents rate it as a primary concern in referrals (McMahon & Forehand, 2003), it is related to poor academic performance (Wehby & Lane, 2009), and it makes learning other skills more difficult (Lipschultz & Wilder, 2017).

Schoen (1983) proposed that noncompliance in response to an instruction could take one of the following: "(a) no response is forthcoming, (b) no response is initiated within a prespecified period of time, or (c) some other, non-requested behavior is performed" (p. 483). Similarly, Walker, Ramsey, and Gresham (2004) identified four forms of noncompliance: (a) passive noncompliance where the individual does not perform the requested behavior but engages in non-overt behaviors; (b) simple refusal where the individual acknowledges the directive but indicates via words or gestures that he/she will not comply; (c) direct defiance where the individual displays hostility, anger, or overt resistance; and (d) negotiation where the individual attempts to bargain, compromise, or propose alternative behavior. It has been argued, however, that in any case of noncompliance, the individual is engaging in some other, possibly aberrant, behavior (Walker, 1993). Therefore, while an operational definition of the target behavior is an essential component of any intervention, it is even more important in a behavior such as noncompliance which can take multiple forms.

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Incidence

Noncompliance has been identified as among the most common childhood behavior problems (Majdalany, Wilder, Allgood, & Sturkie, 2017) with Walker (1993) noting that it is common in individuals with intellectual disabilities. Smith and Lerman (1999) reported that results of several studies suggest that noncompliance is the primary reason that parents of children with developmental disabilities request behavioral services. Breiner and Forehand (1982) found that developmentally delayed children evidenced noncompliance at a greater rate than nondisabled children. Fidura, Lindsey, and Walker (1987) identified that 87% of clients in a residential setting who were referred for behavior problems had noncompliance identified as one of the referring problems.

Lowe et al. (2007) examined the prevalence of challenging behaviors within the learning disability population in South Wales. Results indicated that noncompliance was identified as the most prevalent form of difficult/disruptive behavior with over 80% of the sample reporting noncompliance. Kalb and Loeber (2003) estimated the prevalence of noncompliance in children and adolescents to be between 25% and 65%. Sukhodolsky, Cardona, and Martin (2005) evaluated aggression and noncompliance in child psychiatric inpatients. Moderate-to-high correlations were observed between four types of aggression (verbal and physical against self, others, or objects) and noncompliant behavior. In addition, intellectual disability significantly predicted aggression and noncompliance. Noncompliant behavior was associated with length of hospitalization and number of psychiatric medications at the time of discharge.

As was previously mentioned, noncompliance can take many forms and can occur any time an individual/child is doing anything other than what has been requested by a parent or other adult authority figure within a specific time frame (Kalb & Loeber, 2003). Another interesting way to categorize noncompliance is if the adult command is a "do" or "don't" directive. Most of the literature focuses on "do" commands in which case noncompliance is a failure to follow a command to engage in some specified behavior. Walker (1993) also brought in the idea that disruptive behavior could be considered a form of noncompliance. The rest of this chapter will, however, focus on noncompliance and facilitating compliance of "do" commands.

Assessment of Noncompliance

For the most part, norm-referenced behavior rating scales (e.g., BASC-3, CBCL, Conners CBRS) do not have specific measures for noncompliance. Behavioral assessment techniques are therefore the focus of assessment and intervention planning for individuals exhibiting noncompliant-related behaviors. As was previously mentioned, noncompliance can take multiple forms; therefore, an operational definition of the target behavior is the initial step in any assessment. An operational definition defines behavior in such a way as to remove confusion and ambiguity with regard to the occurrence of a behavior. It contains an agreed-upon description of the motor behavior involved in observable and measurable terms (Alberto & Troutman, 2013). For example, noncompliance could be operationally defined as (a) any occurrence of saying "no," "I don't want to," or "I won't do it" to any adult directive or (b) any response that does not match the delivered instruction within a certain period of time (e.g., 5 seconds) from the time the directive was delivered. If after observation of the behavior and/or interviews with relevant adults, the definition of noncompliance could be specific to the form it is taking or specific to the target individual (e.g., passive noncompliance, simple refusal, overt resistance).

The definition of noncompliance should be an accurate, complete, and concise description of the behavior (Cooper, Heron, & Heward, 2007). Morris (1985) offered three questions to insure the accuracy and completeness of the definition: (a) Can you quantify the behavior (e.g., What is the frequency of the behavior in a specified period of time)? (b) Will a stranger know if the child is engaging in the behavior by reading your

definition? (c) Can you break down the target behavior into smaller behavioral components? The answer to the first two questions should be yes and the answer to the third question should be no. Cooper et al. suggested a function-based definition in which the behavior is defined according to its effect on the environment. For example, does the noncompliance serve a reinforcing or escape function?

It may appear that the primary function of noncompliance is task avoidance (escape). However, several recent studies on FBA have found that noncompliance may be maintained by both positive and negative reinforcements (McKerchar & Abby, 2012; Rodriguez, Thompson, & Baynham, 2010). Rodriguez et al. (2010) examined the effects of attention (positive reinforcement) and escape (negative reinforcement) on noncompliance for three children, one with ID. Results indicated that the function of noncompliance was mostly attention for the two students without ID with both escape and attention being functions for the child with ID. Therefore, it is recommended to assess the function of specific behavior of each individual prior to implementing an intervention (Wadsworth, Hansen, & Wills, 2015).

Functional behavior assessment (FBA) is a "systematic assessment for obtaining information about the purpose (functions) a problem behavior serves for a person" Cooper et al., 2007, p. 696). Functional analysis (FA) is a subset of FBA where antecedents and consequences of a behavior are examined experimentally so their separate effects on the problem behavior can be observed. It is important to note that a functional analysis of problematic behaviors is not always necessary when completing an FBA. There are times when indirect and descriptive assessments will yield enough information to address the functions of the behaviors. In addition, FA has several disadvantages. Specific to noncompliance, these include the following: (a) The FA procedures may temporarily increase the noncompliance. (b) Arranging conditions to reinforce noncompliance may seem counterintuitive to relevant adults. (c) Analysis of the behavior in a controlled environment may not match the environmental conditions in the natural environment. (d) The time, effort, and expertise required to conduct the FA may not be justified. Therefore, the focus of this chapter is functional behavior assessment without hypothesis testing and both indirect and direct observational methods.

FBA is a multistep process that may include some or all of the following: (a) record review, (b) parent and staff interviews, (c) child interviews if appropriate, (d) completion of rating scales such as the Motivation Assessment Scale (MAS) (Durand & Crimmins, 1992) or the Questions About Behavior Function (QABF) (Paclawskyj, Matson, Rush, Smalls, & Vollmer, 2000), and (e) direct observation of the student in the problem activity/setting.

The purpose of the behavioral interview is to gather as much information as possible about the topography of the problem behavior and the environmental conditions associated with the behavior. In addition to the topography of the behavior, the interviewer wants to gain information on the times of day the behavior occurs, during what activities the behavior is most likely to occur, the settings in which the behavior is most likely to occur, any specific materials in which the child is engaged when noncompliance occurs, what people are present, what often happens before the noncompliance (antecedent), what does the child or others do right after the noncompliance (consequences), and any efforts that have already been made to reduce the noncompliance.

Direct observation is an important component in understanding the topography and identifying the function of noncompliance in children as they are more reliable than informant reports (Alberto & Troutman, 2013). Observation helps provide an accurate description of the context, antecedents, and consequences of the behavior. It is recommended that the initial observation sessions consist of a narrative (anecdotal) recording. In a narrative recording, the observer keeps detailed narrative account of behavior and surrounding environmental events in a sequential manner as it happens. Information from this recording can then be transferred into a structured format which includes instances of the target behavior as well as antecedents and consequences. This should aid

the observer in determining the function of the behavior.

A scatterplot procedure can also be used to collect data over a longer period of time than possible by the behavioral professional (e.g., psychologist, behavior analyst). Data for a scatterplot analysis are usually collected by a teacher, paraprofessional, or parent. A common format for a scatterplot is the creation of a grid in which days are listed horizontally along the top and times during the day are listed vertically at the left of the grid. Time is divided into hour, half hour, quarter hour, etc. intervals. Each cell contains a designation of either the rate of noncompliance (e.g., high, medium, or low rate) or the actual number of instances of noncompliance during that interval.

A final observational strategy is to do an ABC analysis. Using this approach, the observer records antecedents and consequences associated with each observation of the target behavior. As hypotheses regarding the function of the behavior have usually been developed prior to ABC observations, it may be helpful to create a coding system with codes for possible antecedents and consequences. Alberto and Troutman (2013), Cooper et al. (2007), and Zirpoli (2012) provide examples of ABC recording sheets.

Behavior rating scales specific to eliciting information about the function of the behavior are also a useful addition to the assessment process. Examples of these instruments include the Motivation Assessment Scale (MAS) (Durand & Crimmins, 1992) and the Questions About Behavior Function (QABF) (Paclawskyj et al., 2000). The MAS consists of 16 items each assessing the target behavior on the four possible functions of behavior (sensory reinforcement, escape, attention, tangible). The QABF is comprised of 25 items rating the target behavior on five potential maintaining functions (attention, escape, nonsocial, physical, tangible).

It has been argued, however, that FBA is not necessarily the "tool of choice" for many, if not all, referrals in psychology and education (Noell & Gansle, 2009). Noell and Gansel argue that for many referral concerns, including possibly noncompliance, interventions with established efficacy have already been developed and when matched to the referral concern can be effective for many individuals. Such interventions are discussed in the next section.

Interventions

Walker (1993) reviewed literature pertaining to the noncompliant behavior of people with intellectual disabilities. Noncompliant behavior was considered in terms of antecedents (characteristics of instructions), behaviors (characteristics of tasks being refused), and consequences (environmental results of noncompliance). Conclusions drawn from this review indicated the following:

- Noncompliance is more likely when instructions are vague and/or interrupted with other instructions.
- A series of high-probability requests can enhance compliance to a subsequent low-probability request.
- Noncompliance is more likely when task demands are too difficult.
- Either positive reinforcement (attention, tangibles) or negative reinforcement (escape) may maintain noncompliant behavior.
- Oppositional children respond poorly to verbal reward alone and somewhat better to tangible rewards and to programs utilizing time-out. By contrast, noncompliance in children with ID respond well to programs utilizing combinations of social and tangible rewards, but not necessarily time-out.

Majdalany, Wilder, Allgood, and Sturkie (2017) recognized that noncompliance can be conceptualized in one of two ways, antecedent or consequent variables. With regard to antecedent variables (Radley & Dart, 2016), noncompliance may result from a skill deficit. That is, the child does not comply because he/she either cannot understand the command or lacks the necessary capability to comply. Consequent variables may play a more important role for other children. In these cases, the child has the skills to comply but support for compliance may be weak. There may

be insufficient reinforcement to comply, or the noncompliance may be maintained by another source of reinforcement which may be greater than the reinforcement for compliance. They point out that identification of antecedent and consequent variables responsible for the occurrence of noncompliance is therefore important in intervention planning. They provide a method, such as FBA, for the assessment and treatment of noncompliance that includes an examination of the extent to which noncompliance is due to a skill deficit before intervening. This study illustrates the importance of using an FBA to examine antecedent and consequent conditions prior to intervention planning.

Wadsworth et al. (2015) examined noncompliance in three elementary-age students with intellectual disabilities. Initial FBA indicated functional behavioral escape as the primary function of the behavior in all three students, with access to tangible items identified as a secondary function in one student. Their results suggest that compliance can be successfully maintained when teacher monitoring and self-monitoring procedures are used. They suggest that the link between self-monitoring and the children's rewards was directly based on the identified function of their behavior. Teacher monitoring preceded self-monitoring and served as a form of modeling and instruction for self-monitoring. Kamps, Wendland, and Culpepper (2006) and Lane et al. (2007) also conducted research which supports self-monitoring as an intervention to improve compliance. They also found that the combination of both self-monitoring and function-based interventions can be implemented effectively and efficiently by teachers.

Belfiore, Basile, and Lee (2008) used the concept of behavioral momentum (Nevin, 1996) to explain their results which demonstrated an increase in compliance to low-probability classroom commands for a 7-year-old student with moderate ID. High-probability command sequence (HPCS) has been shown to increase task compliance across a wide variety of behaviors, individuals, and settings. Belfiore et al. preceded each low-probability command (LP) with three to five high-probability commands (HP), randomly drawn from the preliminary list of commands. Results indicated an increase in compliance to low-probability classroom commands suggesting this as an effective antecedent intervention to increase compliance. Others (e.g., Ardoin, Martens, and Wolfe (1999), Austin and Agar (2005)), Mace and Belfiore (1990), Mace et al. (1988)), Wehby and Hollanhan (2000))) have also found support for the efficacy of HPCS.

Bryce and Jahromi (2013) examined children's compliance and noncompliance in relation to parental control strategies in 20 children with high-functioning autism (HFA) and 20 matched typically developing children. Results indicated that typically developing children demonstrated significantly more compliance, and significantly less noncompliance, than children with HFA when parents used indirect commands. Indirect commands are those commands that can be interpreted as optional or implied or stated in question form.

Ducharme and DiAdamo (2005) used an errorless approach to reduce severe noncompliance with two 5-year-old girls with Down syndrome in a special education classroom. In errorless compliance training, noncompliance is regarded as an "error." Errors are minimized through the delivery of commands that are easy to follow and which are likely to be complied. More demanding commands are introduced gradually with reinforcement for compliance. As a result of the graduated nature of intervention, noncompliance is minimized throughout treatment, rendering reductive consequences unnecessary. Results indicated substantial improvements in the classroom compliance in both participants. Ducharme, Sanjuan, and Drain (2007) used a similar errorless compliance training program to improve compliance behavior in children with Asperger syndrome.

Mindfulness can be defined as "keeping one's consciousness alive to the present reality" (Nhat Hanh, 1976, P. 11). It has been operationalized and examined in the United States through the work of Jon Kabat-Zinn and others. Kabot-Zinn (2006), who developed mindfulness-based stress reduction, defines mindfulness as the awareness that arises through paying attention, on purpose, in the present moment, without judgment. Dumas (2005) provides a model of mindfulness-based parent training that incorporates the following assump-

tions: (a) Much of human behavior is automatized, that is, it is a specific way of coping which behaviors are performed with little conscious awareness, and these behaviors are stable and highly resistant to change. (b) Conflict in families with disruptive children reflects ineffective automatized behaviors that are maintained by strong negative emotions. (c) "Intervention relies on mindful practices to teach parents to consider their own and their child's behavior nonjudgmentally, to distance themselves from negative emotions, and to develop parenting goals that are accompanied by motivated action plans" (p. 780), (d) Effective ways of coping that become automatized with practice facilitate maintenance and generalization of intervention gains. The program itself encourages to share their experiences and concerns and attend to their immediate thoughts and feelings nonjudgmentally, to distance themselves from their existing coping mechanisms and the resulting negative emotional states, and to choose effective goals for themselves and their children and establish a plan to reach these goals.

Mindfulness-based interventions have been found to be effective with a variety of groups and presenting conditions. These include level of anxiety in youth with anxiety disorders (Borquist-Conlon, Maynard, Brendel, & Farina, 2019), increasing psychological well-being and decreasing psychological distress in teachers (Klingbeil & Renshaw, 2018), decreasing symptoms of depression (Wang et al., 2018), reducing negative affectivity (Schumer, Lindsay, & Creswell, 2018), disruptive behavior in adolescents (Klingbeil et al., 2017), improving cognitive performance and resilience to stress with children in schools (Zenner, Herrnleben-Kurz, and Walach (2014), problem behaviors of children with ASD (Bluth, Roberson, Billen, & Sams, 2013), improvement in task performance and a decrease in task avoidance behaviors in children with ID (Kim & Kwan, 2018), and compliance in children with ADHD (Singh et al., 2010).

Kim and Kwon (2018) examined the effects of a mindfulness-based intervention to decrease task avoidance behavior and to increase on-task behavior in three children diagnosed with mild ID. The mindfulness intervention was implemented individually with each of the three participants and consisted of 25 biweekly sessions at 45 minutes per session. Results indicated that all participants exhibited improvement in task performance including an increase in on-task behavior, improvement in completion accuracy in arithmetic, and a decrease in time taken to task completion. In addition, there was a decrease in task avoidance behaviors with all participants and participants' mothers all reported children's daily life behaviors as distinctively improved as a result of the intervention. While the results of mindfulness-based intervention with children with ID are still preliminary, they do appear to be a promising intervention for children with ID.

Effective instruction delivery alone has been found to increase compliance in children (Roberts, Tingstrom, Olmi, & Bellipanni, 2008). Roberts et al. found that effective instruction giving by parents increased compliance to greater than 80% of all ($n = 4$) child participants and that the addition of time-in and contingent praise further increased compliance to three of the four participants.

Recommended Approach to Intervention

Positive Behavior Supports In schools, whenever possible, use positive behavior supports (PBS)). Within PBS, there are three tiers of support with corresponding goals and activities (Simonsen & Sugai, 2019). Using effective compliance strategies can facilitate compliance at all three tiers of PBS, especially at Tiers 1 and 2.

- Tier 1 – Prevent academic and behavior problems: school-wide academic and behavior interventions.
- Tier 2 – Prevent the development of more serious problems and improve problem behavior: target interventions for students not responding to Tier 1
- Tier 3 – Decrease impact of antisocial behavior on a student's daily functioning: develop individualized intervention to meet the unique needs of student

Give Effective Commands Walker et al. (2004) offer the following effective command giving strategies:

- Only give as many commands as needed (decreased compliance occurs with increases in the number of commands given).
- Obtain student attention and eye contact.
- Use more "initiating" (or "start") commands versus "terminating" (or "stop") commands.
- Deliver one directive or command at a time – for tasks with multiple steps, give a separate command for each step.
- Use clear, concise, and specific language ("alpha" commands).
- Allow time for student to comply.
- Only give the command two times – if not followed after second time, provide consequence for noncompliance.
- Give direction from a distance of approximately 3 feet.
- Use a matter-of-fact and nonemotional tone of voice (do not yell, plead, or threaten).
- Reinforce compliance!

Use Precision Requests Precision request is a method for delivering teacher/parent directions to prompt compliance and consistently follow up noncompliance (Jenson & Reavis, 1997). Consideration should be given to using precision requests in combination with other strategies as part of a multicomponent intervention (e.g., Kehle, Bray, Theodore, & Jenson, 2000). Steps include:

1. First request for compliance, use "please" and characteristics of effective commands (see above).
2. Wait 5 seconds – If there is compliance, *reinforce*!
3. Noncompliance: Repeat request using signal words – "You need to…."
4. Compliance: *Reinforce*!
5. Noncompliance: Mild preplanned negative consequence (e.g., loss of opportunity to earn token for that time period).

Offer Choices Offering a child two or more options and allowing him/her to independently select an option can increase compliance as choice can provide the individual an opportunity to have control over his/her environments. Choice can be used to encourage and support appropriate behaviors and academic growth in a variety of ways for individuals with or without disabilities including those with severe disabilities. Research supporting choice as an intervention include the following: choice of routine activity and steps within activity (Dibley & Lim, 1999), choice of academic task (Dunlap et al., 1994), choice of task sequence (Wehby & Lane, 2019), choice of math intervention for general education students (Carson & Eckert, 2003), and choice of task and reinforcement for students with severe disabilities (Cosden, Gannon, & Haring, 1995).

Use High-Probability Request Sequence A high-probability request sequence is the presentation of a series of directions that an individual is likely to perform (i.e., high-probability command) delivered immediately before a request that a student is less likely to perform (i.e., low-probability command) (Wehby & Lane, 2009). Using a series of high-probability requests builds behavioral momentum in order to increase the probability of compliance with the low-probability request. Davis (1995) provided suggestions for implementing a high-probability request sequence: (a) deliver a series of three to five high-p commands at a rapid pace, (b) provide praise for each performance of the high-probability command, (c) deliver a low-probability command, and (d) provide praise for the performance of the low-probability request.

Functional Behavior Assessment As was previously discussed, FBA is a "systematic assessment for obtaining information about the purpose (functions) a problem behavior serves for a person" Cooper et al., 2007, p. 696) and is thought as a fundamental procedure in behavior analysis. However, it is not recommended in all cases of noncompliance as the first choice as an assessment and intervention strategy. As Noell and Gansle (2009) point out, there are established intervention procedures that have years of empirical support that can be matched to most individuals with compliance concerns starting with improved command giving by parents and

teachers. Other intervention strategies discussed above can also be considered in lieu of an FBA. In addition, in those cases where an FBA is warranted, an informal approach is recommended as the first choice (Pindiprolu, 2009).

Summary

Noncompliance, i.e., not following or refusing to follow the directions, rules, or wishes of someone else, can be passive, such as not following a direction, or active, such as whining/crying or becoming aggressive or self-injurious. It is also helpful to remember that noncompliance can be purposeful, but not at all times. Noncompliance is an important behavior on which to focus as it is related to several psychiatric disorders, parents rate it as a primary concern in referrals, it is related to poor academic performance, and it makes learning other skills more difficult. Noncompliance has been identified as among the most common childhood behavior problems and is common in individuals with intellectual disabilities (Walker, 1993) with Smith and Lerman (1999) reporting that noncompliance is the primary reason that parents of children with developmental disabilities request behavioral services.

Noncompliance can take many forms across individuals and environmental conditions. Therefore, an important first step in any assessment is developing a clear operational definition of the behavior. Functional behavior assessment should be considered in the assessment of noncompliance, but more informal techniques are probably sufficient, and hypothesis testing should only be considered when standard interventions have been unsuccessful. Intervention can be as simple as training caregivers in effective instruction giving, but other techniques such as providing choice or using a high-probability request sequence are usually all that is needed. If, however, these interventions are not successful in ameliorating the noncompliance, an FBA and function-based intervention may be required.

References

Alberto, P. A., & Troutman, A. C. (2013). *Applied behavior analysis for teachers* (9th ed.). Upper Saddle River, NJ: Pearson.

Ardoin, S. P., Martens, B. K., & Wolfe, L. A. (1999). Using high probability instructional sequences with fading to increase student compliance during transitions. *Journal of Applied Behavior Analysis, 32*, 339–351. https://doi.org/10.1901/jaba.1999.32-339

Austin, J. L., & Agar, G. (2005). Helping young children follow their teachers' directions: The utility of high probability command sequences in pre-k and kindergarten classrooms. *Education and Treatment of Children, 28*, 222–236.

Autism Speaks. (2012). *Aggressive and Challenging Behaviors Tool Kit*. Retrieved from https://www.autismspeaks.org/sites/.../Challenging%20Behaviors%20Tool%20Kit.pdf

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*, 160–171. https://doi.org/10.1007/s10864-007-9054-x

Bluth, K., Roberson, P. N. E., Billen, R. M., & Sams, J. M. (2013). Cultivating mind: Mindfulness interventions for children with autism spectrum disorder and problem behaviours, and their mothers. *Journal of Child and Family Studies, 24*, 3093–3106. https://doi.org/10.1007/s10826-015-0114-x

Borquist-Conlon, D. S., Maynard, B. R., Brendel, K. E., & Farina, A. S. J. (2019). Mindfulness-based interventions for youth with anxiety: A systematic review and meta-analysis. *Research on Social Work Practice, 29*, 195–205. https://doi.org/10.1177/1049731516684961

Breiner, J., & Forehand, R. (1982). Mother-child interactions: A comparison of a clinic-referred developmentally delayed group and two nondelayed groups. *Applied Research in Mental Retardation, 3*, 175–183.

Bryce, C. I., & Jahromi, L. B. (2013). Brief report: Compliance and noncompliance to parental control strategies in children with high-functioning autism and their typical peers. *Journal of Autism and Developmental Disorders, 43*, 236–243. https://doi.org/10.1007/s10803-012-1564-2

Carson, P. M., & Eckert, T. L. (2003). An experimental analysis of mathematics instructional components: Examining the effects of student- selected versus empirically selected interventions. *Journal of Behavioral Education, 12*, 35–54. https://doi.org/10.1023/A:1022370305486

Cooper, J. O., Heron, T. E., & Heward, W. L. (2007). *Applied behavior analysis* (2nd ed.). Upper Saddle River, NJ: Pearson.

Cosden, M., Gannon, C., & Haring, T. G. (1995). Teacher-control versus student-control over choice of task and reinforcement for students with severe behavior problems. *Journal of Behavioral Education, 5*, 11–27. https://doi.org/10.1007/BF02110212

Davis, C. A. (1995). Peer as behavior change agents for preschoolers with behavioral disorders. *Preventing School Failure*, *39*, 4–9. https://doi.org/10.1080/1045988X.1995.9944635

Dibley, S., & Lim, L. (1999). Providing choice making opportunities within and between daily school routines. *Journal of Behavioral Education, 9*, 117–132. https://doi.org/10.1023/A:1022888917128

Ducharme, J. M., & DiAdamo, C. (2005). An errorless approach to management of child noncompliance in a special education setting. *School Psychology Review, 34*, 107–115.

Ducharme, J. M., Sanjuan, E., & Drain, T. (2007). Errorless compliance training: Success-focused behavioral treatment of children with Asperger syndrome. *Behavior Modification, 31*, 329–344. https://doi.org/10.1177/0145445506295050

Dumas, J. E. (2005). Mindfulness-based parent training: Strategies to lessen the grip of automaticity in families with disruptive children. *Journal of Clinical Child and Adolescent Psychology, 34*, 779–791. https://doi.org/10.1207/s15374424jccp3404_20

Dunlap, G., DePerezel, M., Clarke, S., Wilson, D., Wright, S., White, R., & Gomez, A. (1994). Choice making to promote adaptive behavior for students with emotional and behavioral disorders. *Journal of Applied Behavior Analysis, 27*, 505–518. https://doi.org/10.1901/jaba.1994.27-505

Durand, V. M., & Crimmins, D. (1992). *Motivation assessment scale*. Topeka, KS: Monaco & Associates.

Fidura, J. G., Lindsey, E. R., & Walker, G. R. (1987). A special behavior unit for treatment of behavior problems of persons who are mentally retarded. *Mental Retardation., 25*, 107–111.

Jenson, W. R., & Reavis, H. K. (1997). Contracting to enhance motivation. In W. R. Jenson, D. P. Morgan, S. J. Kukic, & H. K. Reavis (Eds.), *Best practices: Behavioral and educational strategies for teachers* (pp. 65–71). Longmont, CA: Sopris West.

Kabot-Zinn, J. (2006). *Mindfulness for beginners: Reclaiming the present moment and your life*. Louisville, CO: Sounds True.

Kalb, L. M., & Loeber, R. (2003). Child disobedience and noncompliance: A review. *Pediatrics, 111*, 641–652. https://doi.org/10.1542/peds.111.3.641

Kamps, D. M., Wendland, M., & Culpepper, M. (2006). Active teacher participation in functional behavior assessment for students with emotional and behavioral disorder risks in general education classrooms. *Behavioral Disorders, 31*, 128–146. https://doi.org/10.1177/019874290603100203

Kehle, T. M., Bray, M. A., Theodore, L., & Jenson, W. R. (2000). A multi-component intervention designed to reduce disruptive classroom behavior. *Psychology in the Schools, 37*, 474–481. https://doi.org/10.1002/1520-6807(200009)37:5<475::AID-PITS7>3.0.CO;2-P

Kim, J., & Kwon, M. (2018). Effects of mindfulness-based intervention to improve task performance for children with intellectual disabilities. *Journal of Applied Research in Intellectual Disabilities, 31*, 87–97. https://doi.org/10.1111/jar.12333

Klingbeil, D. A., Fischer, A. J., Renshaw, T. L., Bloomfield, B. S., Polakoff, B., Willenbrink, J. B., … Chan, K. T. (2017). Effects of mindfulness-based interventions on disruptive behavior: A meta-analysis of single-case research. *Psychology in the Schools, 54*, 70–87. https://doi.org/10.1002/pits.21982

Klingbeil, D. A., & Renshaw, T. L. (2018). Mindfulness-based interventions for teachers: A meta-analysis of the emerging evidence base. *School Psychology Quarterly, 33*, 501–511. https://doi.org/10.1037/spq0000291.supp

Lane, K. L., Rogers, L. A., Parks, R. J., Weisenbach, J. L., Mau, A. C., Merwin, M. T., & Bergman, W. A. (2007). Function-based interventions for students who are nonresponsive to primary and secondary prevention efforts: Illustrations at the elementary and middle school levels. *Journal of Emotional and Behavioral Disorders, 15*, 169–183. https://doi.org/10.1177/10634266070150030401

Lipschultz, J. L., & Wilder, D. A. (2017). Behavioral assessment and treatment of noncompliance: A review of the literature. *Education and Treatment of Children, 40*, 253–298. 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*, 625–636. https://doi.org/10.1111/j.1365-2788.2006.00948.x

Mace, F. C., & Belfiore, P. J. (1990). Behavioral momentum in the treatment of escape-motivated stereotypy. *Journal of Applied Behavioral Analysis, 23*, 507–514. https://doi.org/10.1901/jaba.1990.23-507

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*, 123–141. https://doi.org/10.1901/jaba.1988.21-123

McKerchar, P. M., & Abby, L. (2012). Systematic evaluation of variables that contribute to noncompliance: A replication and extension. *Journal of Applied Behavior Analysis, 45*, 607–611. https://doi.org/10.1901/jaba.2012.45-607

McMahon, R. J., & Forehand, R. L. (2003). *Helping the noncompliant child: Family-based treatment for oppositional behavior* (2nd ed.). New York, NY: Guilford Press.

Majdalany, L. M., Wilder, D. A., Allgood, J., & Sturkie, L. (2017). Evaluation of a preliminary method to examine antecedent and consequent contributions to noncompliance. Journal of Applied Behavior Analysis, 50, 146–158. https://doi.org/10.1002/jaba.353

Morris, R. J. (1985). *Behavior modification with exceptional children: Principles and practices*. Glenview, IL: Scott, Foresman.

Nevin, J. A. (1996). The momentum of compliance. *Journal of Applied Behavior Analysis, 29*, 535–547. https://doi.org/10.1901/jaba.1996.29-535

Nhat Hanh, T. (1976). *The miracle of mindfulness*. Boston, MA: Beacon Press.

Noell, G. H., & Gansle, K. A. (2009). Introduction to functional behavior assessment. In S. G. Little, A. Akin-Little, M. A. Bray, & T. J. Kehle (Eds.), *Behavioral interventions in schools: Evidence-based positive strategies* (1st ed., pp. 43–58). Washington, DC: APA Books.

Paclawskyj, T., Matson, J., Rush, K., Smalls, Y., & Vollmer, T. (2000). Questions about behavioral function (QABF): Behavioral checklist for functional assessment of aberrant behavior. *Research in Developmental Disabilities, 21*, 223–229. https://doi.org/10.1016/S0891-4222(00)00036-6

Pindiprolu, S. (2009). Functional assessment-based intervention in the general education setting. *Journal of Evidence-Based Practices for Schools, 10*, 48–65.

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*, 39–54. https://doi.org/10.1007/s10567-015-0197-3

Roberts, D. S., Tingstrom, D. H., Olmi, D. J., & Bellipanni, K. D. (2008). Positive antecedent and consequent components in child compliance training. *Behavior Modification, 32*, 21–38. https://doi.org/10.1177/0145445507303838

Rodriguez, N. M., Thompson, R. H., & Baynham, T. Y. (2010). Assessment of the relative effects of attention and escape on noncompliance. *Journal of Applied Behavior Analysis, 43*, 143–147. https://doi.org/10.1901/jaba.2010.43-143

Schoen, S. F. (1983). The status of compliance technology: Implications for programming. *Journal of Special Education, 17*, 483–496. https://doi.org/10.1177/002246698301700410

Schumer, M. C., Lindsay, E. K., & Creswell, J. D. (2018). Brief mindfulness training for negative affectivity: A systematic review and meta-analysis. *Journal of Consulting and Clinical Psychology, 86*, 569–583. https://doi.org/10.1037/ccp0000324.supp

Simonsen, B., & Sugai, G. (2019). School-wide positive behavioral interventions and supports: A systems level application of behavioral principles. In S. G. Little & A. Akin-Little (Eds.), *Behavioral interventions in schools: Evidence-based positive strategies* (2nd ed.). Washington, DC: APA Books.

Singh, N. N., Singh, A. N., Lancioni, G. E., Singh, J., Winton, A. S. W., & Adkins, A. D. (2010). Mindfulness training for parents and their children with ADHD increases the children's compliance. *Journal of Child and Family Studies, 19*, 157–166. https://doi.org/10.1007/s10826-009-9272-z

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*, 183–195. https://doi.org/10.1016/S0891-4222(99)00002-5

Sukhodolsky, D. G., Cardona, L., & Martin, A. (2005). Characterizing aggressive and noncompliant behaviors in a children's psychiatric inpatient setting. *Child Psychiatry and Human Development, 36*, 177–193. https://doi.org/10.1007/s10578-005-3494-0

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. https://doi.org/10.1177/0741932514554102

Walker, G. R. (1993). Noncompliant behavior of people with mental retardation. *Research in Developmental Disabilities, 14*, 87–105. https://doi.org/10.1016/0891-4222(93)90014-B

Walker, H. M., Ramsey, E., & Gresham, F. M. (2004). *Antisocial behavior in school: Evidenced-based practices* (2nd ed.). Wadsworth/Thomson Learning: Belmont, CA.

Wang, Y., Li, X., Zheng, W., Xu, Z., Ng, C. H., Ungvari, G. S., … Xiang, Y. (2018). Mindfulness-based interventions for major depressive disorder: A comprehensive meta-analysis of randomized controlled trials. *Journal of Affective Disorders, 229*, 429–436. https://doi.org/10.1016/j.jad.2017.12.093

Wehby, J. H., & Hollanhan, M. S. (2000). Effects of high probability requests on the latency to initiate academic tasks. *Journal of Applied Behavioral Analysis, 33*, 259–262. https://doi.org/10.1901/jaba.2000.33-259

Wehby, J. H., & Lane, K. L. (2009). Proactive instructional strategies for classroom management. In S. G. Little, A. Akin-Little, M. A. Bray, & T. J. Kehle (Eds.), *Behavioral interventions in schools: Evidence-based positive strategies* (pp. 141–156). Washington, DC: APA Books.

Wehby, J. H., & Lane, K. L. (2019). Classroom management. In S. G. Little & A. Akin-Little (Eds.), *Behavioral interventions in schools: Evidence-based positive strategies* (2nd ed.). Washington, DC: APA Books.

Zenner, C., Herrnleben-Kurz, S., & Walach, H. (2014). Mindfulness-based interventions in schools – A systematic review and meta-analysis. *Frontiers in Psychology, 5*, ArtID: 603.

Zirpoli, T. J. (2012). *Behavior management: Positive applications for teachers* (6th ed.). Upper Saddle River, NJ: Pearson.

25

Social Behavior for Individuals with Intellectual Disabilities and Dual Diagnosis: Common Deficits and Assessment Tools

Justin B. Leaf, Julia L. Ferguson, Christine Milne, and Joseph H. Cihon

An intellectual disability describes intellectual and adaptive limitations with deficits across three major domains of adaptive skills including conceptual, social, and practical adaptive skills (American Psychiatric Association, 2013). The *Diagnostic and Statistical Manual of Mental Disorders* (DSM-V; American Psychiatric Association, 2013) further clarifies that for an individual to receive a diagnosis of intellectual disability they must fail to meet developmental and sociocultural standards for personal independence and social responsibility and these deficits in adaptive behaviors limit functioning in daily life (e.g., social participation) in multiple environments (e.g., home, school, and community; American Psychiatric Association, 2013). Although professionals and nonprofessionals commonly associate intellectual disability as primarily a cognitive deficit, it is clear that a major defining characteristic of intellectual disability is deficits in social and adaptive behavior that impede the individuals' functioning and overall quality of life (Griffiths, Condillac, & Legree, 2014; Raymond & Matson, 1989; Sparrow, Cicchetti, & Saulnier, 2016).

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There has yet to be a universally accepted definition of what constitutes social behavior; however, some commonly agreed-upon features include, but are not limited to, engaging with others, learning through observation, learning from contextual cues, engaging in behaviors that increase the likelihood of future interactions with others, and engaging in behavior required to access items or activities that are unattainable alone. Ultimately, there are a myriad of different social skills that individuals potentially use on a daily basis, and many individuals diagnosed with intellectual disabilities have deficits with a few or many of these needed skills. These social skills could include joint attention (Charman & Campbell, 1997; Mundy & Newell, 2007; Paparella & Kasari, 2004; Summers & Impey, 2011; Zampini, Salvi, & D'Odorico, 2015), observational learning (Foti et al., 2015), social communication (Belva, Matson, Sipes, & Bamburg, 2012), perspective taking (Benson, Abbeduto, Short, Nuccio, & Maas, 1993), theory of mind (Abbeduto, Short-Meyerson, Benson, & Dolish, 2004; Jervis & Baker, 2004; Fiasse & Nader-Grosbois, 2012; Thirion-Marissiaux & Nader-Grosbois, 2008; Yirmiya, Erel, Shaked, & Solomonica-Levi, 1998;), and/or emotional regulation (McClure, Halpern, Wolper, & Donahue, 2009).

A well-developed social skills repertoire is critical for individuals diagnosed with intellectual disabilities for multiple reasons. First, many

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social skills are behavioral cusps (Rosales-Ruiz & Baer, 1997) which permit the individual to access new contingencies and environments that, in turn, lead to the development of more social skills. For instance, by acquiring social skills such as observational learning and joint attention, there is higher likelihood for more rapid acquisition of language, problem solving, and independence with certain activities (Kotera, Kiyokawa, Ashikaga, & Ueda, 2011; Mundy, Sigman, & Kasari, 1990; Tomasello & Farrar, 1986).

Second, appropriate social behavior may be critical in forming and maintaining meaningful relationships and friendships (Whitehouse, Chamberlain, & O'Brien, 2001). When individuals do not brush their teeth (Belva et al., 2012), communicate with their friends, show good sportsmanship, or regulate emotions appropriately, it may negatively affect interactions with peers. This may reduce the likelihood of individuals forming relationships (e.g., acquaintances, friendships, romantic relationships) which could impede their overall quality of life (Schalock, 2004; Whitehouse et al., 2001).

Third, increases in social skills may be correlated with improvements in social communication and overall social competence (Gresham & MacMillan, 1997). Deficits in social behavior, like those commonly seen with individuals diagnosed with intellectual disabilities, may inhibit peers from initiating interaction. However, with a well-developed social skills repertoire, peers may be more likely to initiate and respond to initiations from individuals diagnosed with intellectual disabilities. With increased interactions, individuals diagnosed with intellectual disability will have more opportunities to engage in, and practice, appropriate social skills, which may lead to more effective and natural communication with peers.

Fourth, another important reason why social behavior is so critical is the direct benefit that it has on school performance and/or job performance (Ellenkamp, Brouwers, Embregts, Joosen, & van Weeghel, 2016). Researchers have demonstrated that when individuals have appropriate social behaviors and positive social relationships, they are more likely to attend school (e.g., Ashburner et al., 2018). Researchers have also shown that when individuals have positive social relationships with peers, they perform better in school (e.g., Ladd, Birch, & Buhs, 1999). Furthermore, researchers have found correlations between employment and an individual's social life, autonomy, and overall quality of life. That is, when individuals have appropriate social behaviors and positive social relationships, they are more likely to stay employed and do well in their jobs, and vice versa (e.g., Jahoda, Kemp, Riddell, & Banks, 2008).

Fifth, failing to develop appropriate social behavior can result in other long-term undesirable outcomes. These outcomes include, but are not limited to, loneliness (Gilmore & Cuskelly, 2014), depression (Hartley & Birgenheir, 2009), being bullied (Ashburner et al., 2018; Christensen, Fraynt, Neece, & Baker, 2012), incarceration (Hayes, 1994), or even suicide ideation (Ludi et al., 2012). Research has shown these outcomes are more likely for individuals diagnosed with intellectual disabilities compared to typically developing individuals (Austin, Hunter, Gallagher, & Campbell, 2018; Gilmore & Cuskelly, 2014). One potential reason for an increased likelihood for these outcomes with this population is a lack of appropriate social behaviors leading to less friendships.

Finally, and arguably the most substantial reason why it is imperative for individuals diagnosed with intellectual disabilities to develop desired social skills is to improve their quality of life (Schalock, 2004). One of the main outcomes desired by parents for their children, or professionals for their clients, is to live a meaningful and high-quality life. Many individuals diagnosed with intellectual disabilities can have good paying jobs, engage in enjoyable hobbies, maintain reciprocal friendships, and have romantic partners, all of which can lead to a high quality of life (Jahoda et al., 2008). However, without appropriate social behavior, these outcomes are less likely to be achieved.

In order for parents and professionals to effectively develop and employ interventions addressing social skills with individuals diagnosed with intellectual disabilities, they must: (a) know com-

mon deficits displayed by this population; (b) identify social skill assessments; and (c) implement effective, evidence-based procedures. While the range of deficits can vary based on the severity of the deficit, the purpose of this chapter is to outline common social deficits and provide an overview of common standardized assessments which can be used to identify social strengths and deficits for individuals diagnosed with intellectual disabilities.

Common Social Skill Deficits

Joint Attention

Joint attention refers to when individuals "coordinate attention with a social partner in relation to some object or event" (Naber et al., 2008, p. 143). Joint attention is commonly divided into two types: responding to joint attention bids and initiating bids for joint attention (Mundy & Newell, 2007; Summers & Impey, 2011). Responding to joint attention bids is when an individual follows the gaze or point of another person to an event. For example, if a mother was at a zoo and saw a hippopotamus, the mother might say, "Look Alexander, there is a hippopotamus," while looking at the hippopotamus. Then Alexander and the mother both look at the hippopotamus. Initiating bids for joint attention occurs when the individual sees the hippopotamus, gains the attention of another person, and informs the person (e.g., saying, "Look") of what they are seeing.

Joint attention usually develops prior to one year of age with the development of eye gaze to object and stimuli (Mundy, 2018). Joint attention has been identified as an essential skill for appropriate communication development (Tomasello & Farrar, 1986), social development (Mundy & Willoughby, 1998), and cognitive development (Mundy, 2018). Researchers have identified deficits in joint attention for individuals diagnosed with autism spectrum disorder (ASD) and/or intellectual disabilities (e.g., Bruinsma, Koegel, & Koegel, 2004; Naber et al., 2008). For example, Kasari, Freeman, Mundy, and Sigman (1995) found that children with Down syndrome scored worse on shifting their attention between an object and a caregiver than typically developing children.

Summers and Impey (2011) evaluated responding to joint attention bids and initiations of joint attention bids with four individuals diagnosed with Angelman syndrome. The results showed that the individuals were less impaired when responding to joint attention bids than when initiating joint attention bids; however, the joint attention behaviors were displayed less frequently than observed with typically developing children. These results differ from some research on joint attention with individuals diagnosed with Down syndrome, in which participants were more likely to initiate joint attention bids than follow joint attention bids (Landry & Chapieski, 1989). Overall, joint attention is a commonly observed deficit for individuals diagnosed with intellectual disabilities.

Observational Learning

Observational learning involves watching others' actions and the outcomes of those actions, which increases the likelihood that the observer engages/does not engage in similar actions to obtain or avoid similar outcomes in similar situations in the future (Bandura, 1971). Observational learning permits acquiring new behavior without direct intervention (Nadel, 2002). That is, observational learning can lead to the acquisition of more complex skills such as communication (Charlop, Schreibman, & Tyron, 1983), play skills (Collozi, Ward, & Crotty, 2008), and social skills (Wilson, 2013) without direct intervention. In addition, teaching procedures based upon observational learning can be used to teach a variety of skills including first aid skills (Ozkan, 2013), eliminating inappropriate sexual behavior (Dowrick & Ward, 1997), increasing reading skills (Rehfeldt, Latimore, & Stromer, 2003), and changing preference for play items (Leaf et al., 2012).

The development of observational learning and imitative repertoires begins at birth (Nadel, 2002). Esseily, Nadel, and Fagard (2010) noted that, depending on motor movement, observation

learning skills are effective around 12 months while Nadel (2002) described how some neonates (i.e., newborn babies) begin to imitate facial movements of others as early as 35 min old. Additionally, researchers have found that babies at 16 months of age begin imitating the use of tools through observational learning (Somogyi & Esseily, 2014).

Unfortunately, many individuals diagnosed with intellectual disabilities have deficits in observational learning. How these deficits manifest varies with the severity of the intellectual disability. For example, Foti et al. (2015) found that those diagnosed with Prader-Willi syndrome had major deficits in observational learning, compared to typically developing individuals, which interfered with their ability to correctly engage in a sequencing task, but those with Williams syndrome did not have these same deficits. Taylor and DeQuiznio (2012) noted that individuals diagnosed with ASD may have deficits in the prerequisite skills required to learn from observation such as attending, imitation, and discriminating contingencies. DeQuiznio and Taylor (2015) addressed one of these deficits by successfully teaching four children diagnosed with ASD to discriminate contingencies of others and use the information based on those contingencies when acquiring new expressive labels. Biederman, Stepanuik, Davey, Raven, and Ahn (1999) evaluated the observational learning skills of individuals diagnosed with Down syndrome. They found that children with Down syndrome learned through observation only when the video model was substantially slowed down. Therefore, there may be a range of types and level of severity of deficits for observation learning when it comes to individuals diagnosed with an intellectual disability.

Adaptive Behavior and Daily Living

One cluster of behaviors which are strongly related to social behaviors are adaptive (e.g., bathing, toileting, feeding) and daily living skills (e.g., setting the table, doing the dishes, doing the laundry). Although these skills are not inherently social, they do correlate with social behaviors and one's ability to make and sustain meaningful relationships. For instance, if an individual does not maintain appropriate hygiene, this could affect how people respond to the individual. Even if an individual has an established relationship, friendship or romantic, they must maintain a certain level of appropriate hygiene in order to maintain those relationships. Poor hygienic practice could deter others from spending long periods of time together, visiting the individual, interacting with the individual, or inviting them to their home.

Daily living skills, such as grocery shopping, require many skills necessary for a successful trip, such as making a list, finding and retrieving items on the list, and paying. Equally important are social skills that may be necessary when visiting the grocery store. If items are missing, or an individual is having trouble finding a specific item, the individual may need to engage in problem solving skills that require interactions with others. The individual must be able to recognize a problem, find the most appropriate person to ask for help, and ask appropriately. In addition, they must understand social etiquette such as waiting in line to check out, waiting to grab an item, or saying "excuse me" if someone is blocking a desired item.

Researchers have demonstrated that individuals diagnosed with intellectual disabilities have significant deficits in adaptive behavior and daily living skills (Belva & Matson, 2013). Belva and Matson (2013) conducted a comprehensive review of daily living skills as they relate to individuals diagnosed with profound intellectual disabilities. The authors used the Vineland Adaptive Behavior Scales (Sparrow, Cicchetti, & Balla, 2005; Sparrow et al., 2016) to evaluate the daily living skills of the 204 participants. The results showed low scores on many of the behaviors in the daily living skills domain. For example, only 5.39% of participants responded to caring for their hair without being reminded, 4.90% looked after their own health, and only 1.47% initiated telephone calls with others. Researchers have also shown that the acquisition of adaptive behaviors with individuals diagnosed with intel-

lectual disabilities may be slower than typically developing children (van Duijn, Dijkxhoorn, Scholte, & van Berckelaer-Onnes, 2010). Given this gap and the skill deficits that may be present for those diagnosed with an intellectual disability, interventions are needed to develop these skills.

Employment

A survey of individuals who were diagnosed with an intellectual disability between ages 21 and 64 revealed that approximately 34% were employed (Siperstein, Parker, & Drascher, 2013). The range of jobs held by those diagnosed with an intellectual disability varies due to the range of severity levels. There are a range of skills necessary to maintain a job including the skills needed to execute their job, as well as social skills to appropriately interact with fellow employees, potential customers, and clients. Belva and Matson (2013) reported that only 3.92% of participants diagnosed with intellectual disabilities held a full-time job. Additionally, very few participants diagnosed with intellectual disabilities could notify supervisors when they were absent due to an illness or let them know when they would be arriving late. These skills are critical to maintain a full-time job.

Emotional Regulation

Emotional regulation is comprised of a constellation of social behaviors, ranging from recognizing others' emotions in pictures to calming oneself down when angry, upset, or sad. Researchers have shown that individuals diagnosed with an intellectual disability can recognize basic emotions (e.g., happy, sad) but have difficulty with more complex emotions or when a picture displays a neutral face (Moore, 2001; Owen, Browning, & Jones, 2001). Fortunately, individuals diagnosed with intellectual disabilities can learn to receptively and/or expressively label emotions quickly (e.g., Garcia-Villamisar & Dattilo, 2018). For more advanced emotional regulation behaviors, such as using coping strategies when emotionally aroused or irritated, individuals diagnosed with intellectual disabilities commonly display deficits (Benson & Fuchs, 1999).

Theory of Mind

Another imperative social skill is commonly referred to as theory of mind (see Baron-Cohen, 2001 for a review). Theory of mind has been defined as "the ability to reason and infer about another's mental states such as beliefs, desires, intentions…" (Jervis & Baker, 2004 p. 49). Theory of mind begins to develop in children as early as four years of age (Astington, 1993). There are multiple assessments to test for theory of mind, and one of the most commonly used is the false belief test (Baron-Cohen, Leslie, & Frith, 1985). A common example of the false belief test is having a person or character place an object somewhere (e.g., placing a toy under the table) and then leave the room. When the person or character leaves the room, another person or character moves the object (e.g., takes the toy and places under the bed). The examiner would then ask where the first person would look for the object. These types of false belief tests have been highly predictive of measures of theory of mind (Astington, 1993) and can help to distinguish between individuals diagnosed with ASD and those diagnosed with an intellectual disability (Frith & Corcoran, 1996).

In fact, there has been some discussion if theory of mind is a deficit for individuals diagnosed with intellectual disabilities who are not diagnosed with ASD. In one of the more seminal works, Baron-Cohen et al. (1985) showed that individuals diagnosed with intellectual disabilities did not differ from typically developing children in terms of theory of mind, but both groups differed from individuals diagnosed with ASD. As such, researchers have typically treated individuals diagnosed with an intellectual disability as control participants (e.g., Adrien, Rossignol, Barthélémy, Jose, & Sauvage, 1995;

Blijd-Hoogewys, van Geert, Serra, & Minderaa, 2008).

There is not, however, a universal consensus on the deficits in theory of mind for individuals diagnosed with an intellectual disability (Abbeduto et al., 2004; Yirmiya et al., 1998). For example, Charman and Campbell (1997) found that only 39% of individuals diagnosed with and without Down syndrome were able to pass the false belief tasks. Ashcroft, Jervis, and Roberts (1999) found that even fewer (i.e., 13%) adults diagnosed with intellectual disabilities passed theory of mind tasks. These discrepant findings may be due to certain contextual variables. For example, one difference may be due to the age of an individual. Jervis and Baker (2004) compared the performance of 20 adults (28 to 45 years of age) to 20 children (9 to 13 years of age) on theory of mind tasks (i.e., deceptive box test with photographic cue, false-belief task, deceptive box test, and belief-desire reasoning task). The authors found that the children performed significantly better than the adults on these tasks. Another possible variable that may impact responding on theory of mind tasks may have to do with an individual's language capabilities. For example, Abbeduto et al. (2004) showed that individuals with more severe language impairments performed worse on false belief tasks. Therefore, some individuals diagnosed with intellectual disabilities may have deficits in theory of mind while others may not.

Friendship

Perhaps one of the most important outcomes of a well-developed social behavior repertoire is friendships. That is, a failure to develop many of the aforementioned skills can ultimately affect the development of friendships, which is why teaching basic, intermediate, and advanced social behaviors are so important to individuals diagnosed with intellectual disabilities. Although the definition of friendship changes across the life span, there are some universal characteristics of friendships including: (a) an emotional bond between the individuals; (b) mutual interests; (c) mutual enjoyment; (d) opportunities to interact with each other; and (e) that the interactions are reciprocal (Howes, 1983; Sigstad, 2016). Researchers have suggested that the development of friendships is critical for emotional and physical wellbeing (Berndt, 2002). When individuals have friendships, they perform better in school and at work (Hartup & Stevens, 1999), are less lonely (Gilmore & Cuskelly, 2014), and have less risk for depression and/or suicide (Hartley & Birgenheir, 2009).

Unfortunately, researchers have identified that individuals diagnosed with intellectual disabilities have fewer friendships and lower quality friendships compared to typically developing children (Fulford & Cobigo, 2018). For example, Bigby, Webber, Bowers, and McKenzie-Green (2008) evaluated 24 individuals diagnosed with an intellectual disability living in an institution in Australia. Within this study, 50% of the participants reported not having any friendships other than staff members. In a more recent study, Friedman and Rizzolo (2017) surveyed 1341 individuals diagnosed with developmental disabilities. While the results showed that 84% of the responders reported having friendships, the majority (i.e., 56%) indicated they were not satisfied with the number of friends and nearly half (i.e., 47%) were not satisfied with the amount of contact between friends.

Many individuals diagnosed with intellectual disabilities report that staff members are their friends (van Asselt-Goverts, Embregts, & Hendriks, 2015; Pottie & Sumarah, 2004). Bigby et al. (2008) reported that 83% of respondents identified a staff member as a friend. This may be problematic for a variety of reasons. First, there is a high rate of turnover amongst staff in residential placements (Hewitt & Larson, 2007). This high rate of turnover could result in perceived friendships quickly dissolving. Second, a true friendship (see Taubman, Rafuse, Leaf, & Leaf, 2011 for a discussion) must be reciprocal, and

friendships resulting in one party being paid to "hang out" or be a "friend" are not reciprocal. Third, a paid staff member as a friend might prevent the development of new friendships within the community. Finally, and unfortunately, having staff members as "friends" could result in an unbalance of power and might result in the staff member taking advantage of the individual diagnosed with an intellectual disability.

In addition to a less-developed social behavior repertoire, limited opportunities to interact with others may contribute to a lack of friendships within this population (Pottie & Sumarah, 2004). Bigby et al. (2008) showed that the average network size for an individual diagnosed with an intellectual disability was 1.92 (range 0–6 people). Bigby et al. further stated, "Four Residents (16%) had a *non-existent* network, with no contact with either family or friends outside their home…" (Bigby et al., 2008, p. 151). Friedman and Rizzolo (2017) found that 41.7% of the responders indicated that the organization in which the individual resides did not have proper support for enhancing, developing, or maintaining friendships. These limited network sizes could result in limited opportunities to interact with people outside of the home and may contribute to a lack of friendships.

Common Standardized Social Skill Assessments

Assessment is an important part of evaluating current social functioning, determining goals, and tracking ongoing progress. When assessing social skills for individuals diagnosed with intellectual disabilities, it is critical that there is ongoing assessment and evaluation, informal and formal, of their social skills development. Although standardized assessments play an important role in diagnosis, this section will focus on using assessments for intervention planning, determining goals, and assessing progress for individuals diagnosed with intellectual disabilities.

Informal Assessments

Observations One type of informal assessment is observation of an individual in naturally occurring social situations. This could be in a classroom, in the community, at home with family and relatives, or in workplace settings. All of these environments set the occasion for social behavior and opportunities to initiate and respond to social interactions. Assessing social behavior through naturalistic observations allows one to identify social deficits present within commonly encountered environments. This also allows one to record the potential antecedents and consequences that precede and follow wanted and unwanted social behaviors (Gresham, 1981). Observing an individual with an intellectual disability in their relevant environments could also set the occasion to observe typically developing individuals in those same environments to see what common social behaviors are occurring within that environment. This allows one to see what typical social norms and behaviors are present in that environment, the topographies of the social behaviors, the antecedents that set the occasion for the social behavior, and the consequences that maintain the social behavior in that environment. By observing the topography of common social behaviors displayed by typically developing individuals, an interventionist would be able to create a task analysis of what the social behavior should look like and the prerequisite skills necessary to engage in the social skill. Observing the antecedents that set the occasion for a social skill also allows an interventionist to teach the social cues that signal the opportunity to engage in the social skill. Responding to social cues is often a deficit for individuals diagnosed with intellectual disabilities (American Psychiatric Association, 2013). By observing the common social cues present within regularly visited environments, an interventionist would be able to teach the relevant social cues and how one should respond to those social cues in a way that is appropriately consequated by persons present in those environments. Observations in natural

environments also allows one to track progress of the target social behaviors more frequently than some standardized assessments and allows an interventionist to assess what skill deficits may still be present after targeting a specific social skill and adjust programming as necessary.

Interviews Another type of informal assessment is the use of an interview with people that are familiar with the individual diagnosed with an intellectual disability. When using an interview as an assessment technique, there are several factors to consider including (a) who to interview, (b) the qualifications of the person conducting the interview, and (c) the types of questions asked during the interview.

The person being interviewed should have frequent interactions with the individual diagnosed with intellectual disabilities and know them well. This could be an individual's classroom teacher, a parent or caregiver, or paraprofessional aides. The person being interviewed should have frequent interactions with the individual and a good understanding of the person's social skill strengths and deficits. Interviewing someone who does not interact with the individual frequently (e.g., principal of school, aunt or uncle from out of town) may not provide accurate information. This could also be true of people that do interact with the individual frequently. As such, it is recommended to include multiple respondents of the same interview questions to provide a better picture of the individual's social skill deficits across different environments.

It is also important to consider the qualifications of the person conducting the interview, especially if the interview is conducted in-person. Interviewing a caregiver about their child takes clinical sensitivity and clinical judgment (Taylor, LeBlanc, & Nosik, 2018). Questions about an individual's social communication deficits can be a sensitive subject for many caregivers. The person conducting the interview should show compassion and understanding during the interview process while also collecting the relevant information about the individual's social behavior.

The types of questions asked during the interview are also important to consider. Questions should be informed by the goal of the interview. Potential goals of the interview could be to (a) determine new social skill targets, (b) ask about ongoing progress of social skill targets, and (c) decide what intervention should be used to target certain skills. Knowing the goal of the interview helps inform the types of questions to ask during the interview and what follow-up questions are necessary. Once you determine the goal of the interview, one should then plan out the questions that will be asked. Open-ended questions, instead of yes-or-no questions, allows the respondent to provide more information on certain topics, but asking questions that are too broad may not allow for specific-enough answers or provide relevant information. The interviewer must also be cautious to ensure they are not leading the respondent to respond in a particular way when asking clarification questions. Overall, the interview format allows a person to validate what social goals are important to the relevant people in an individual's life and the types of interventions that would be acceptable to implement from people that know the individual best.

Standardized Assessments

Vineland-3 Adaptive Behavior Scales A commonly used standardized assessment with individuals diagnosed with intellectual disabilities is the Vineland-3 Adaptive Behavior Scales (Sparrow et al., 2016). Measuring overall adaptive behavior is useful for getting a complete picture of an individual's social-communicative skill level compared to other same-aged peers (Bielecki & Swender, 2004). Edgar Doll and Sara Sparrow developed the first iteration of the Vineland to evaluate adaptive behavior for individuals diagnosed with intellectual disabilities in 1965 (Sparrow et al., 2016), which contained, and still contains, several unique features. First, the Vineland assessment was one of the first to consider the relationship between mental deficits and social competence. Doll even stated, "No

mental diagnosis is complete if it does not begin with a sound estimate of social competence and end with a prediction of social competence following prognosis or treatment" (Sparrow et al., 2016, p. 11). The early emphasis on social competency and social behavior as part of adaptive functioning makes the Vineland assessment unique compared to other assessments available for individuals diagnosed with an intellectual disability that tend to focus on their intellectual capabilities. The creators of the Vineland also considered adaptive behavior as multifaceted, meaning adaptive behavior is not just about one skill set, but comprised of many. Currently the Vineland-3 assesses several domains including: communication, daily living skills, socialization, motor skills, and maladaptive behavior. Although some domains assessed may not seem necessarily social (e.g., daily living skills), as previously noted, skills within each of these domains impact overall social competence.

The Vineland-3 compares adaptive behavior of an individual to a normative population, and scores from each domain are compared to other individuals of the same age. This allows one to see a comparative score of social-adaptive behavior to other individuals of the same age. The Vineland-3 is scored on a Likert scale from 0–2, and respondents can be a parent/caregiver or a teacher. The Adaptive Behavior Composite score is comprised of three main domains: communication, daily living skills, and socialization. Additional domains on the Vineland-3 that are not included in the Adaptive Behavior Composite score are the motor skills domain and maladaptive behavior domain.

Perhaps the most relevant domain of the Vineland-3 with respect to the purpose of this chapter is the socialization domain. The socialization domain is broken down into three subdomains: interpersonal relationships, play and leisure, and coping skills. The interpersonal relationships subdomain focuses on how an individual responds and relates to others and asks questions about beginning social behavior, emotional development, friendships, conversational skills, interpersonal appropriateness, and caring toward others. The play and leisure subdomain focuses on how an individual engages in play and activities with others. Questions on the play and leisure subdomain include topics such as learning to play skills, responding to social cues, playing games and sports, and socializing with peers. The coping skills subdomain focuses on how well an individual demonstrates behavior and emotional control in different situations with others. Questions within this subdomain pertain to how an individual controls their emotions, is considerate to others, adapts to different situations, and manages social risks.

Although the socialization domain on the Vineland-3 provides the most relevant information about an individual's social behavior, other domains on the Vineland-3 also impact social competency and behavior. The communication domain and daily living skills domain greatly influence an individual's social behavior, and many questions on the Vineland under these domains should be considered when assessing an individual's social behavior. The communication domain involves questions pertaining to an individual's receptive and expressive communication skills and written communication skills. More specifically, many questions about an individual's receptive and expressive communication abilities relate to social behavior. For example, items under the communication domain such as looking at you when they hear your voice, looking when someone calls their name, understanding gestures, responding to the tone of your words, understanding the meaning of facial expressions on others, and understanding what people mean when they are being sarcastic all relate directly to social competency.

Items within the daily living skills domain do not directly relate to how an individual behaves socially, but many impact how others perceive social behavior and, without these skills, would impact an individual's ability to have meaningful social interactions and relationships. Items such as appropriate toileting behavior, wiping or cleaning face when eating something messy, brushing teeth, bathing/showering, washing hair, respecting people's right to privacy, and traveling independently all relate to social receptiveness.

Similar to the daily living skills domain, the maladaptive behavior domain on the Vineland-3 also provides information about challenging behaviors that could greatly impact an individual's social receptiveness. Challenging behaviors such as tantrums, bullying, breaking rules, being aggressive, or destroying other's possessions would all greatly impact an individual's social competency and should be taken into consideration as behaviors to decrease when looking at an individual's overall social behavior.

Ultimately, the Vineland-3 can provide an overall picture of an individual's social competency which can then help with intervention planning and choosing important social skills to address. After scoring a Vineland-3, the comprehensive score report provides intervention guidance divided by domain and content areas which can greatly help with selecting relevant social behavior goals for an individual diagnosed with intellectual disability.

Social Skills Improvement System The social skills improvement system (SSiS) is a multirater standardized assessment of social behaviors that affect teacher–student relationships, parent–child relationships, peer relationships, and academic performance at school (Gresham & Elliott, 2008). The SSiS can be filled out by a parent/caregiver, a teacher, or the individual themselves and uses a Likert scale to rate each item on the assessment. The SSiS is standardized and norm-referenced for preschool children aged 3 to 5 years, elementary school children aged 6 to 12 years, and teenagers aged 13 to 18 years. Using the SSiS as an assessment tool can help determine specific social skill deficits for individuals diagnosed with intellectual disabilities, what social skills are most important to the rater, and help guide intervention planning. The social skills domain on the SSiS includes seven subdomains: communication, cooperation, assertion, responsibility, empathy, engagement, and self-control. The problem behavior domain includes five subdomains: externalizing, bullying, hyperactivity/inattention, internalizing, and autism spectrum disorder. The teacher forms (i.e., to be completed by the teacher) of the SSiS include the academic competence (i.e., reading achievement, math achievement, motivation to learn) domain due to the correlation between social behavior and academic performance.

Although each domain impacts overall social competency, the questions within the social skills domain will heavily influence what social skills should be targeted for intervention, or track ongoing progress of skills already targeted. Questions pertaining to the communication subdomain query how an individual takes turns and makes eye contact during conversations, their voice tone and gestures, and common manners such as saying please and thank you. Questions on the cooperation subdomain determine how an individual shares and helps others, and complies with others' rules and directions. Questions for the assertion subdomain inquire how an individual initiates to others such as asking for information, introducing themselves, and responding to the actions of others. The responsibility subdomain includes questions about how an individual displays regards for the property or work of others and their ability to communicate with adults. The empathy subscale pertains to how an individual shows concern and respect for others' feelings and viewpoints. The engagement subscale includes questions relating to how the individual joins activities already in progress, invites other to join, initiates conversation, and makes friends. The final social skills subscale, self-control, asks questions about responding appropriately during a conflict, and non-conflict situations (e.g., taking turns and compromising).

Overall, the SSiS is a great standardized assessment tool that aides in intervention planning and tracking. The SSiS is also unique in that it has a built-in social validity measure that asks how important (i.e., not important, important, or critical) each social skill is to the person filling out the form. This is especially relevant when it comes to selecting what social skills to target for an individual diagnosed with an intellectual disability. The corresponding *Social Skills Intervention Guide* (Elliott & Gresham, 2008) also provides sample lessons, examples, and activities for how to target the corresponding social skills found in the SSiS assessment.

Social Responsiveness Scale The Social Responsiveness Scale (SRS-2; Constantino & Gruber, 2012) is a social assessment meant for individuals aged 2.5 years through adulthood. It is comprised of 65 questions and uses a Likert scale to answer each question. Although this assessment is typically used to measure social symptoms associated with a diagnosis of ASD, it can also be a useful tool to assess the social responsiveness for individuals diagnosed with an intellectual disability. The SRS-2 should not be used as a diagnostic tool for individuals with an intellectual disability, but can be useful for tracking goal progress and intervention planning. The 65 questions that comprise the SRS-2 create an overall social responsiveness T-score that corresponds to the level of severity or support that individual requires socially. The SRS-2 also provides T-scores for several subdomains including social awareness, social cognition, social communication, social motivation, and restricted interests and repetitive behavior. The corresponding T-scores fall into the categories of within normal limits, mild range, moderate range, and severe range for each subdomain. Each item on the SRS-2 has the respondent rate how true an item is for that individual (i.e., not true, sometimes true, often true, almost always true) based on their behavior from the past six months. Similar to other standardized assessments, the respondent should know the individual in question well to provide accurate ratings on the assessment. Once the assessment is scored, the T-scores can provide information about the social domains that need to be developed further through systematic intervention.

School Social Behavior Scales and Home and Community Social Behavior Scales The School Social Behavior Scales (SSBS-2; Merrell, 2002b) and the Home and Community Social Behavior Scales (HCSBS; Merrell, 2002a) are two social competency assessments developed to be used in two different settings. The SSBS-2 is an assessment meant for individuals in school settings and would be filled out by the student's teacher or school personnel that have frequent interactions with the student. The SSBS-2 was created out of the need for identifying students in classrooms with social deficits. The creators of the SSBS-2 developed the assessment as a screening tool for early identification of students at risk behaviorally, an assessment for classification and determination for special program eligibility, to aide in intervention plans, provide information relevant to conducting a functional behavior assessment, and as a tool for monitoring social behavior change after intervening on specific social behaviors (Crowley & Merrerll, 2003).

The SSBS-2 has two scales, social competence and antisocial behavior, and is intended for students in kindergarten through Grade 12. The social competence scale is further broken down into three subscales. The peer relations subscale focuses on how frequently a student engages in social skills that are necessary to establish positive relationships and gain social acceptance from their peers. The self-management/compliance subscale includes items related to social skills involving self-restraint, cooperation, and compliance with instructions from the teacher and school staff. The final subscale, academic behavior, consists of items relating to a student's engagement and performance on academic tasks. The antisocial behavior scale is further broken down into three subscales: hostile/irritable, antisocial/aggressive, and defiant/disruptive. The hostile/irritable subscale asks questions relating to student behaviors that would be considered annoying and self-centered and are likely to lead to rejection from their peers. The antisocial/aggressive subscale asks questions about how frequently a student violates school rules and harming others. The final antisocial subscale, defiant/disruptive, has items that ask how likely a student is to disrupt ongoing activities at school and place inappropriate demands on peers or teachers.

The Home and Community Social Behavior Scales (HCSBS) is very similar to the SSBS-2 but differs in its intent to assess social behavior of an individual in home and/or community settings instead of within a school setting. The HCSBS is

comprised of two scales, social competence and antisocial behavior. Unlike the SSBS-2, the HCSBS is only comprised of two subscales. The social competence scale consists of the peer relations subscale and the self-management/compliance subscale and the antisocial behavior scale consists of the defiant/disruptive and antisocial/aggressive subscales. The HCSBS can be filled out by a parent, guardian, or supervisor of the individual in question and is meant to be used for individuals aged 5–18 years old.

Each question on the SSBS-2 and HCSBS assessments has the respondent rate each item using a 5-point Likert scale ranging from 1 (never) to 5 (frequently). The rating the respondent provides should be based on observations from the past 3 months. Unlike the SRS-2 or the Vineland-3 (i.e., assessments that can be used through adulthood), this assessment is similar to the SSiS in that it can only be used with individuals diagnosed with an intellectual disability up to the age of 18 years. Although these assessments are limited to use with children and adolescents, they do provide important information for individuals diagnosed with intellectual disabilities within this age range. The antisocial behavior scale provides critical information about behaviors that should be targeted to decrease, and in return the social competence scale provides information about what social skill replacement behavior should be taught as a replacement to antisocial behaviors.

The Social Communication Questionnaire The Social Communication Questionnaire (SCQ; Rutter, Bailey, & Lord, 2003) was originally developed as a screening tool for individuals diagnosed with ASD and corresponded closely to the Autism Diagnostic Interview-Revised (ADI-R). Although the SCQ was originally intended for use with individuals diagnosed with or suspected of a diagnosis of ASD, it has recently been utilized as an assessment tool for adults diagnosed with an intellectual disability (e.g., Brooks & Benson, 2013; Derks et al., 2017; Sappok, Diefenbacher, Gaul, & Bölte, 2015; Sappok, Brooks, Heinrich, McCarthy, & Underwood, 2017). Although the SCQ comes in two forms (i.e., lifetime and current version), when using the SCQ with adults diagnosed with intellectual disabilities, the current version should be used for screening (Sappok et al., 2015). The SCQ is a 40-item assessment that should be filled out by a caregiver that is familiar with the developmental history and the current social/communication behavior of the individual. The SCQ uses a yes/no format for each item on the questionnaire instead of having the respondent rate each item using a Likert scale like many other social assessments (e.g., SSiS, SRS, Vineland-3). The SCQ can be used with individuals of all ages as long as the individual has a mental age of at least 2 years. The lifetime version of this form is strongly associated with diagnosis, but the current version of the SCQ can help aide one in social intervention planning, goal selection, and for tracking progress over time for an individual with an intellectual disability.

Matson Evaluation of Social Skills with Youngsters and Matson Evaluation of Social Skills for Individuals with Severe Retardation The Matson Evaluation of Social Skills with Youngsters (MESSY; Matson, 1988) is a social behavior assessment for children between the ages of 2 and 18 years. The MESSY was initially intended and designed to assess social behavior in typically developing children but has been researched and used with children diagnosed with intellectual disabilities, children with hearing and visual impairments, children diagnosed with anxiety, and children diagnosed with ASD (Matson, Horovitz, Mahan, & Fodstad, 2013). Similar to other social skill assessments for individuals diagnosed with intellectual disabilities, the MESSY is comprised of scales assessing appropriate and inappropriate social behaviors (Matson, 1988). The MESSY has 64 questions and includes a self-rating scale and a parent/teacher rating scale. Each item is scored on a Likert scale ranging from 1 (not at all) to 5 (very much). Items on the MESSY relating to appropriate social behavior include items such as smiling at others, making others laugh, asking to help others, friendly to new people, and working

well on a team. Items relating to inappropriate social behavior include threatening others, being bossy, complaining often, getting upset when they have to wait, and picking on others. Scores on the MESSY range from 64 to 340. A lower total score suggests higher social competency, and higher total score suggests lower social competency and a higher rate of inappropriate social behaviors. Having scales corresponding to appropriate and inappropriate social behavior will help others decide what social behaviors are necessary to target to increase and teach systematically, as well as other aberrant behaviors that should be targeted to decrease and replace with more appropriate social behavior.

Unlike other social behavior assessments, The Matson Evaluation of Social Skills for Individuals with Severe Retardation (MESSIER; Matson, 1995) was specifically designed to measure social behavior strengths and weaknesses for adults diagnosed with intellectual disabilities that fall in the severe-to-profound range. This makes the MESSIER unique compared to other social behavior assessments that may focus on social behaviors too complex for this population. The MESSIER includes 85 items that fall into six behavior categories: positive verbal, positive nonverbal, positive general, negative verbal, negative nonverbal, and negative general. Items on the MESSIER are rated on a Likert scale ranging from 0 (i.e., never) to 3 (i.e., almost always). Items on the MESSIER should be rated by a parent, caregiver, or staff member that has frequent interactions with the individual, knows them well, and has known them for at least 6 months. The MESSIER is also typically conducted in a semi-structured interview format in which an individual trained in the test administration conducts the interview with the parent, caregiver, or staff member that knows the individual well. Examples of items on the MESSIER that are prosocial positive behaviors include (a) turning head in the direction of caregiver, (b) looking at the face of caregiver when spoken to, (c) smiling in response to positive statements, and (d) saying "please" when asking for something (Matson, 1995). Examples of items on the MESSIER that would fall within the negative verbal, nonverbal, and general categories are (a) disturbing others, (b) preferring to be alone, (c) crying at inappropriate times, and (d) avoiding eye contact (Matson, 1995). Similar to the MESSY assessment tool, the MESSIER is also an assessment tool that can provide valuable information for identifying social behavior goals to increase and identifying aberrant behaviors to decrease and replace them with more appropriate social behaviors.

Conclusion

Knowing common deficits of individuals with intellectual disabilities as well appropriate assessments to evaluate social behavior is critical for treatment planning. It can help professionals and parents design intervention programs which can effectively improve important social behaviors. This should result in practitioners implementing interventions which have empirical support and scientific evidence to support their use (e.g., video modeling, behavioral skills training, social skill groups, and the teaching interaction procedure) and avoiding interventions which have weak empirical evidence (e.g., Social Stories™; Leaf et al., 2015), are not evidence based (e.g., Social Thinking®; Leaf et al., 2016), or have the hallmarks of pseudoscience or antiscience (e.g., Social Thinking® or Floortime; Leaf et al., 2016). Doing so will improve the quality of life of individuals diagnosed with intellectual disabilities so they can live meaningful and happy lives (Schalock, 2004).

References

Abbeduto, L., Short-Meyerson, K., Benson, G., & Dolish, J. (2004). Relationship between theory of mind and language ability in children and adolescents with intellectual disability. *Journal of Intellectual Disability Research, 48*, 150–159.

Adrien, J. L., Rossignol, C., Barthélémy, C., Jose, C., & Sauvage, D. (1995). Développment et fonctionnent de la "théorie de l'espirt"c chez l'enfant autiste et chez

l'enfant normal. *Approche Neuropsychologique des Apprentissages chez l'enfant, 35*, 188–196.

American Psychiatric Association. (2013). *Diagnostic and statistical manual of mental disorders* (5th ed.). Arlington, VA: Amercan Psychiatric Publishing.

Ashburner, J., Saggers, B., Campbell, M. A., Dillon, W. J. A., Hwang, Y., Carrington, S., & Bobir, N. (2018). How are students on the autism spectrum affected by bullying? Perspectives of students and parents. *Journal of Research in Special Educational Needs. Advanced Online Publication*. https://doi.org/10.1111.1471–3802. 12421

Ashcroft, A., Jervis, N., & Roberts, C. (1999). A theory of mind (ToM) and people with learning disabilities: The effects of a training package. *Journal of Applied Research in Intellectual Disabilities, 12*, 58–68.

Astington, J. W. (1993). *The developing child: The child's discovery of the mind*. Cambridge, MA: Harvard University Press.

Austin, K. L., Hunter, M., Gallagher, E., & Campbell, L. E. (2018). Depression and anxiety symptoms during the transition to early adulthood for people with intellectual disabilities. *Journal of Intellectual Disability Research, 62*, 407–421.

Bandura, A. (1971). *Social learning theory*. New York, NY: General Learning Press.

Baron-Cohen, S. (2001). Theory of mind and autism: A review. *International Review of Research in Mental Retardation, 23*(169), 169–184.

Baron-Cohen, S., Leslie, A. M., & Frith, U. (1985). Does the autistic child have a "theory of mind"? *Cognition, 21*, 37–46.

Belva, B. C., & Matson, J. L. (2013). An examination of specific daily living skills deficits in adults with profound intellectual disabilities. *Research in Developmental Disabilities, 34*, 596–604.

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*, 525–529.

Benson, B. A., & Fuchs, C. (1999). Anger-arousing situations and coping responses of aggressive adults with intellectual disability. *Journal of Intellectual and Developmental Disability, 24*, 207–214.

Benson, G., Abbeduto, L., Short, K., Nuccio, J. B., & Maas, F. (1993). Development of a theory of mind in individuals with mental retardation. *American Journal on Mental Retardation, 98*, 427–433.

Berndt, T. J. (2002). Friendship quality and social development. *Current Directions in Psychological Science, 11*, 7–10.

Biederman, G., Stepaniuk, S., Davey, V., Raven, K., & Ahn, D. (1999). Observational learning in children with down syndrome and developmental delays: The effect of presentation speed in videotaped modelling. *Down Syndrome Research and Practice, 6*, 12–18.

Bielecki, J., & Swender, S. L. (2004). The assessment of social functioning in individuals with mental retardation. *Behavior Modification, 28*, 694–708.

Bigby, C., Webber, R., Bowers, B., & McKenzie-Green, B. (2008). A survey of people with intellectual disabilities living in residential aged care facilities in Victoria. *Journal of Intellectual Disability Research, 52*, 404–414.

Blijd-Hoogewys, E. M. A., van Geert, P. L. C., Serra, M., & Minderaa, R. B. (2008). Measuring theory of mind in children: Psychometric properties of the ToM storybooks. *Journal of Autism and Developmental Disorders, 38*, 1907–1930.

Brooks, W. T., & Benson, B. A. (2013). The validity of the social communication questionnaire in adults with intellectual disability. *Research in Autism Spectrum Disorders, 7*, 247–255.

Bruinsma, Y., Koegel, R. L., & Koegel, L. (2004). Joint attention and children with autism: A review of the literature. *Mental Retardation and Developmental Disability Research Review, 10*, 169–175.

Charlop, M. H., Schreibman, L., & Tyron, A. S. (1983). Learning through observation: The effects of peer modeling on acquisition and generalization in autistic children. *Journal of Abnormal Child Psychology, 11*, 355–366.

Charman, T., & Campbell, A. (1997). Reliability of theory of mind task performance by individuals with a learning disability: A research note. *Journal of Child Psychology and Psychiatry, 38*, 725–730.

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*, 49–65.

Collozi, G. A., Ward, L. W., & Crotty, K. W. (2008). Comparison of simultaneous prompting procedure in 1:1 and small group instruction to teach play skills to preschool students with pervasive developmental disorder and developmental disabilities. *Education and Training in Developmental Disabilities, 43*, 226–248.

Constantino, J. N., & Gruber, C. P. (2012). *Social responsiveness scale* (2nd ed.). Torrance, CA: Western Psychological Services.

Crowley, S. L., & Merrerll, K. W. (2003). The structure of the school social behavior scales: A confirmatory factor analysis. *Assessment for Effective Intervention, 28*, 41–53.

DeQuinzio, J. A., & Taylor, B. A. (2015). Teaching children with autism to discriminate the reinforced and nonreinforced responses of others: Implications for observational learning. *Journal of Applied Behavior Analysis, 48*, 38–51.

Derks, O., Heinrich, M., Brooks, W., Sterkenburg, P., McCarthy, J., Underwood, L., & Sappok, T. (2017). The social communication questionnaire for adults with intellectual disability: SCQ-AID. *Autism Research, 10*, 1481–1490.

Dowrick, P. W., & Ward, K. M. (1997). Video feedforward in the support of a man with intellectual disability and inappropriate sexual behaviour. *Journal of Intellectual and Developmental Disability, 22*, 147–160.

Ellenkamp, J. J., Brouwers, E. P., Embregts, P. J., Joosen, M. C., & van Weeghel, J. (2016). Work environment-

related factors in obtaining and maintaining work in a competitive employment setting for employees with intellectual disabilities: A systematic review. *Journal of Occupational Rehabilitation, 26*, 56–69.

Elliott, S. N., & Gresham, F. M. (2008). *Social skills improvement system: Intervention guide*. Minneapolis, MN: NCS Pearson.

Esseily, R., Nadel, J., & Fagard, J. (2010). Object retrieval through observational learning in 8-to 18-month-old infants. *Infant Behavior and Development, 33*, 695–699.

Fiasse, C., & Nader-Grosbois, N. (2012). Perceived social acceptance, theory of mind and social adjustment in children with intellectual disabilities. *Reserach in Developmental Disabilities, 33*, 1871–1880.

Foti, F., Menghini, D., Orlandi, E., Rufini, C., Crinò, A., Spera, S., … Mandolesi, L. (2015). Learning by observation and learning by doing in Prader-Willi syndrome. *Journal of Neurodevelopmental Disorders, 7*(6), 1–12. https://doi.org/10.1186/s11689-015-9102-0

Friedman, C., & Rizzolo, M. (2017). "Get us real jobs:" supported employment services for people with intellectual and developmental disabilities in Medicaid home and community based services waivers. *Journal of Vocational Rehabilitation, 46*, 107–116.

Frith, C. D., & Corcoran, R. (1996). Exploring 'theory of mind' in people with schizophrenia. *Psychological Medicine, 26*, 521–530.

Fulford, C., & Cobigo, V. (2018). Friendships and intimate relationships among people with intellectual disabilities: A thematic synthesis. *Journal of Applied Research in Intellectual Disabilities, 31*(1), e18–e35.

Garcia-Villamisar, D., & Dattilo, J. (2018). Effects of computer-facilitated emotion recognition training for adutls with autism spectrum disorders and intellectual disabilities. *Global Journal of Intellectual and Developmental Disabilities, 5*(2), 1–8.

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*, 192–199.

Gresham, F. M. (1981). Social skills training with handicapped children: A review. *Review of Educational Research, 51*, 139–176.

Gresham, F. M., & Elliott, S. N. (2008). *Social skills improvement system: Rating scales manual*. Minneapolis, MN: NCS Pearson.

Gresham, F. M., & MacMillan, D. L. (1997). Social competence and affective characteristics of students with mild disabilities. *Review of Educational Research, 67*, 337–415.

Griffiths, D., Condillac, R. A., & Legree, M. (2014). *Genetic syndromes and applied behavior analysis: A handbook for ABA practitioners*. London, UK: Jessica Kingsley Publishers.

Hartley, S. L., & Birgenheir, D. G. (2009). Nonverbal social skills of adults with mild intellectual disability diagnosed with depression. *Journal of Mental Health Research in Intellectual Disabilities, 2*, 11–28.

Hartup, W. W., & Stevens, N. (1999). Friendships and adaptations across the life span. *Current Directions in Psychological Science, 8*(3), 76–79.

Hayes, S. (1994). The criminal law and the person with intellectual disability. *Australia & New Zealand Journal of Developmental Disabilities, 19*, 287–292.

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.

Howes, C. (1983). Patterns of friendship. *Child Development, 54*, 1041–1053.

Jahoda, A., Kemp, J., Riddell, S., & Banks, P. (2008). Feelings about work: A review of the socio-emotional impact of supported employment on people with intellectual disabilities. *Journal of Applied Research in Intellectual Disabilities, 21*, 1–18.

Jervis, N., & Baker, M. (2004). Clinical and research implications of an investigation into theory of mind (ToM) task performance in children and adults with non-specific intellectual disabilities. *Journal of Applied Research in Intellectual Disabilities, 17*, 49–57.

Kasari, C., Freeman, S., Mundy, P., & Sigman, M. D. (1995). Attention regulation by children with down syndrome: Coordinated joint attention and social referencing looks. *American Journal of Mental Retardation, 100*, 128–136.

Kotera, A., Kiyokawa, S., Ashikaga, J., & Ueda, K. (2011). The role of observation in collaborative problem solving: Attributing the observed actions to self or other influences insight problem solving. *Cognitive Studies: Bulletin of the Japanese Cognitive Science Society, 18*, 114–126.

Ladd, G. W., Birch, S. H., & Buhs, E. S. (1999). Children's social and scholastic lives in kindergarten: Related spheres of influence? *Child Development, 70*, 1373–1400.

Landry, S. H., & Chapieski, M. L. (1989). Joint attention and infant toy exploration: Effects of down syndrome and prematurity. *Child Development, 60*, 103–118.

Leaf, J. B., Kassardjian, A., Oppenheim-Leaf, M. L., Cihon, J. H., Taubman, M., Leaf, R., & McEachin, J. (2016). Social thinking®: Science, pseudoscience, or antiscience? *Behavior Analysis in Practice, 9*, 152–157.

Leaf, J. B., Oppenheim-Leaf, M. L., Leaf, R., Courtemanche, A. B., Taubman, M., McEachin, J., … Sherman, J. A. (2012). Observational effects on the preferences of children with autism. *Journal of Applied Behavior Analysis, 45*, 473–483.

Leaf, J. B., Oppenheim-Leaf, M. L., Leaf, R. B., Taubman, M., McEachin, J., Parker, T., … Mountjoy, T. (2015). What is the proof? A methodological review of studies that have utilized social stories. *Education and Training in Autism and Developmental Disabilities, 50*, 127–141.

Ludi, E., Ballard, E. D., Greenbaum, R., Pao, M., Bridge, J., Reynolds, W., Horowitz. L. (2012). Suicide Risk in Youth with Intellectual Disabilities. *Journal of Developmental & Behavioral Pediatrics, 33*(5), 431–440.

Matson, J. L. (1988). *The Matson evaluation of social skills with youngsters (MESSY)*. Baton Rouge, LA: Disability Consultants, LLC.

Matson, J. L. (1995). *The Matson evaluation of social skills for individuals with severe retardation (MESSIER)*. Baton Rouge, LA: Disability Consultants, LLC.

Matson, J. L., Horovitz, M., Mahan, S., & Fodstad, J. (2013). Reliability of the Matson evaluation of social skills with youngsters (MESSY) for children with autism spectrum disorders. *Research in Autism Spectrum Disorders, 7*(2), 405–410.

McClure, K. S., Halpern, J., Wolper, P. A., & Donahue, J. J. (2009). Emotion regulation and intellectual disability. *Journal on Developmental Disabilities, 15*, 38–44.

Merrell, K. W. (2002a). *Home and community social behavior scales*. Eugene, OR: Assessment Intervention Resources.

Merrell, K. W. (2002b). *School social behavior scales* (2nd ed.). Eugene, OR: Assessment Intervention Resources.

Moore, D. G. (2001). Reassessing emotion recognition performance in people with mental retardation: A review. *American Journal of Mental Retardation, 106*, 481–502.

Mundy, P. (2018). A review of joint attention and social-cognitive brain systems in typical development and autism spectrum disorder. *European Journal of Neuroscience, 47*, 497–514.

Mundy, P., & Newell, L. (2007). Attention, joint attention, and social cognition. *Current Directions in Psychological Science, 16*, 269–274.

Mundy, P., Sigman, M., & Kasari, C. (1990). A longitudinal study of joint attention and language development in autistic children. *Journal of Autism and Developmental Disorders, 20*, 115–128.

Mundy, P., & Willoughby, J. (1998). Nonverbal communication, affect, and social-emotional developlemtn. In A. Weatherby, S. Warren, & J. Reichle (Eds.), *Transitions in prelinguistic communication*. Baltimore, MD: Brookes Publishing.

Naber, F., Bakermans-Kranenburg, M. J., van Ljzendoorn, M. H., Dietz, C., Can Daalen, E., Swinkels, S. H., … Van Engeland, H. (2008). Joint attention development in toddlers with autism. *European Child & Adolescent Psychiatry, 17*, 143–152.

Nadel, J. (2002). Imitation and imitation recognition: Functional use in preverbal infants and nonverbal children with autism. *The imitative mind: Development, evolution, and brain bases*. In A. Meltzoff & W. Prinz (Eds.), *The imitative mind* (pp. 42–62). Cambridge University Press: Cambridge.

Owen, A., Browning, M., & Jones, R. S. P. (2001). Emotion recognition in adults with mild-moderate learning disabilities: An exploratory study. *Journal of Intellectual Disabilities, 5*, 267–281.

Ozkan, S. Y. (2013). Comparison of peer and self-video modeling in teaching first aid skills to children with intellectual disability. *Education and Training in Autism and Developmental Disabilities, 48*, 88–102.

Paparella, T., & Kasari, C. (2004). Joint attention skills and language development in special needs populations: Translating research to practice. *Infants and Young Children, 17*, 269–280.

Pottie, C., & Sumarah, J. (2004). Friendships between persons with and without developmental disabilities. *Mental Retardation, 42*, 55–66.

Raymond, K. L., & Matson, J. L. (1989). Social skills in the hearing impaired. *Journal of Clinical Child Psychology, 18*, 247–258.

Rehfeldt, R. A., Latimore, D., & Stromer, R. (2003). Observational learning and the formation of classes of reading skills by individuals with autism and other developmental disabilities. *Research in Developmental Disabilities, 24*(5), 333–358.

Rosales-Ruiz, J., & Baer, D. M. (1997). Behavioral cusps: A developmental and pragmatic concept for behavior analysis. *Journal of Applied Behavior Analysis, 30*, 533–544.

Rutter, M., Bailey, A., & Lord, C. (2003). *The social communication questionnaire: Manual*. Los Angeles, CA: Western Psychological Services.

Sappok, T., Brooks, W., Heinrich, M., McCarthy, J., & Underwood, L. (2017). Cross-cultural validity of the social communication questionnaire for adults with intellectual development disorder. *Journal of Autism and Developmental Disorders, 47*, 393–404.

Sappok, T., Diefenbacher, A., Gaul, I., & Bölte, S. (2015). Validity of the social communication questionnaire in adults with intellectual disabilities and suspected autism spectrum disorder. *American Journal on Intellectual and Developmental Disabilities, 120*, 203–214.

Schalock, R. L. (2004). The concept of quality of life: What we know and do not know. *Journal of Intellectual Disability Research, 48*, 203–216.

Sigstad, H. M. H. (2016). Significance of friendship for quality of life in adolescents with mild intellectual disability: A parental perspective. *Journal of Intellectual and Developmental Disability, 41*, 289–298.

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.

Somogyi, E., & Esseily, R. (2014). Mimicry enhances observational learning in 16-month-old infants. *PLoS One, 9*(12), 1–18.

Sparrow, S. S., Cicchetti, D. V., & Balla, D. A. (2005). *Vineland adaptive behavior scales: Second edition (Vineland II)*. Livonia, MN: Pearson Assessments.

Sparrow, S. S., Cicchetti, D. V., & Saulnier, C. A. (2016). *Vineland-3: Vineland adaptive behavior scales* (3rd ed.). Bloomington, MN: NCS Pearson.

Summers, J., & Impey, J. (2011). Assessing joint attention responding and initiation in children with Angelman syndrome. *Journal of Applied Research in Intellectual Disabilities, 24*, 450–458.

Taubman, M. T., Rafuse, J., Leaf, J., & Leaf, R. (2011). True friendships. In M. T. Taubman, R. B. Leaf, & J. McEachin (Eds.), *Crafting connections: Contemporary applied behavior analysis for enriching the social lives of persons with autism spectrum disorder* (pp. 51–57). New York, NY: DRL Books.

Taylor, B. A., & DeQuinzio, J. A. (2012). Observational learning and children with autism. *Behavior Modification, 36*, 341–360.

Taylor, B. A., LeBlanc, L. A., & Nosik, M. R. (2018). Compassionate care in behavior analytic treatment: Can outcomes be enhanced by attending to relationships with caregivers? *Behavior Analysis in Practice. Advanced online publication*. https://doi.org/10.1007/s40617-018-00289-3

Thirion-Marissiaux, A. F., & Nader-Grosbois, N. (2008). Theory of mind "emotion", developmental characteristics and social understanding in children and adolescents with intellectual disabilities. *Research in Developmental Disabilities, 29*, 414–430.

Tomasello, M., & Farrar, M. J. (1986). Joint attention and early language. *Child Development, 57*, 1454–1463.

van Asselt-Goverts, A. E., Embregts, P. J., & Hendriks, A. H. (2015). Social networks of people with mild intellectual disabilities: Characteristics, satisfaction, wishes, and quality of life. *Journal of Intellectual Disability Research, 59*, 450–461.

van Duijn, G., Dijkxhoorn, Y., Scholte, E. M., & van Berckelaer-Onnes, I. A. (2010). The development of adaptive skills in young people with down syndrome. *Journal of Intellectual Disability Research, 54*, 943–954.

Whitehouse, R., Chamberlain, P., & O'Brien, A. (2001). Increasing social interactions for people with more severe learning disabilities who have difficulty developing personal relationships. *Journal of Learning Disabilities, 5*, 209–220.

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*, 1819–1831.

Yirmiya, N., Erel, O., Shaked, M., & Solomonica-Levi, D. (1998). Meta-analyses comparing theory of mind abilities of individuals with autism, individuals with mental retardation, and normally developing individuals. *Psychological Bulletin, 124*, 283–307.

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.

Characteristics and Assessment of Pica in Individuals with Intellectual Disability

26

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Definition and Topography

Pica is the act of ingesting nonfood substances that lack nutritional value (Sturmey & Williams, 2016). The American Psychiatric Association (2013) classifies pica among feeding and eating disorders when the behavior (a) persists for at least 1 month; (b) is age-inappropriate; and (c) is not culturally or socially sanctioned and (d) when the severity warrants clinical attention. Pica may emerge at any point from childhood to adulthood and can be comorbid with other conditions, including intellectual disability (American Psychiatric Association, 2013). There are several reasons why an individual might ingest inedible items that would not be considered pica, for example, participation in a cultural or religious practice, during starvation, or during the typical early oral motor development observed during infancy and early childhood (Sturmey & Williams, 2016; Young, 2011).

Some individuals with pica consume a variety of different nonfoods, such as rocks, plastic, trash, and metal objects (e.g., Falcomata, Roane, & Pabico, 2007). Others exclusively ingest only one specific nonfood item or only items that share a physical property. For example, a person with pica might only ingest cigarette butts (single item pica; Piazza, Hanley, & Fisher, 1996) or might ingest only sharp metal items such as safety pins, nails, and tacks (e.g., Goh, Iwata, & Kahng, 1999; Sturmey & Williams, 2016). When pica is limited to consuming a specific item or consuming different items that all share a physical property, it may be referenced by a more specific term or subtype. For example, cautopyreiophagia is a subtype of pica that involves the consumption of burnt matches, and acuphagia is a subtype that refers to consuming sharp objects. Another taxonomical system for pica involves classification by the composition or texture of items (McAdam, Breibord, Levine, & Williams, 2012). For example, an individual may only ingest biological nonfoods with a specific texture (e.g., soft tissue). Tables 26.1 and 26.2 display categories that have been used to classify pica in previous literature.

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Table 26.1 Pica terminology and referents

Pica terminology	Term referent
Acuphagia	Sharp objects
Amylophagia	Laundry starch
Coprophagia	Human feces, animal dung
Cautopyreiophagia	Burnt matches
Foliophagia	Leaves, grass, acorns, pinecones
Geomelophagia	Raw potatoes
Geophagia	Earth, soil-like
Hyalophagia	Glass
Lithophagia	Rocks, gravel, pebbles
Lignophagia	Word, bark, twigs
Plumbophagia	Paint chips (lead)
Pagophagia	Ice, freezer frost
Trichophagia	Hair
Tobaccophagia	Cigarette butts

Table 26.2 Pica categories

Category	Examples
Biological secretions	Feces, urine, blood, mucous and vomit
Biological solids	Fingernails, bone, and hair
Chemicals	Cooper and lead chips
Inedible food stuffs	Uncooked, rotten, or spoiled foods
Organic materials	Laundry starch, paper, and leaves
Physical items	Glass, matches, and metal objects

Prevalence and Risk Factors

Studies aimed at identifying the commonality of pica among individuals with intellectual disability have reported a wide range of prevalence estimates. For example, Ali (2001) reviewed the literature and found estimates of pica prevalence to range from 0.3% to 25%. The wide range may be partially attributed to differences in operational definitions of pica across studies. For example, some studies have included eating frozen or raw food as well as excessive or compulsive eating in their definition of pica (e.g., Tewari, Krishnan, Valsalan, & Roy, 1995). Sample characteristics may also influence prevalence estimates and may reveal potential risk factors for the emergence of pica. For example, the highest rates of pica have been reported among individuals that reside in institutional facilities, suggesting risk of pica may increase as intellectual disability becomes more severe (Ali, 2001; Swift, Paquette, Davison, & Saeed, 1999). Similarly, males, individuals with autism spectrum disorder, and those with more severe social and leisure skill deficits appear more likely to engage in pica (Ashworth, Hirdes, & Martin, 2009; Matson, Belva, Hattier, & Matson, 2011).

Adverse Effects of Pica

Pica is a life-threatening behavior (e.g., Ashworth et al., 2009). The physical risks of pica include poisoning, hemorrhaging, choking, parasitic infection, and death (Decker, 1993; McLoughlin, 1988). In many cases, the ingested items must be removed surgically. For example, Decker (1993) reviewed treatment outcomes for 48 people with comorbid intellectual disability and pica and reported that 75% ($n = 42$) of cases required surgical intervention. Of those cases, the complication rate was 30% and the mortality rate was 11%. Kamal, Thompson, and Paquette (1999) reported the extent of injury and the surgical management necessary following ingestion of vinyl gloves by five patients with intellectual disability. One of the five patients died of an upper gastrointestinal tract hemorrhage before surgery. Autopsy results revealed a large gastric ulcer caused by blockage from the vinyl gloves. The other four cases all required endoscopic exploration to confirm the presence of gastric blockages. Of those four cases, three required further surgical intervention (i.e., laparotomy, gastronomy) to remove the blockages.

Pica may persist across time, posing an ongoing threat to an individual's health. For example, McLoughlin (1988) studied pica as a cause of death in three men with intellectual disability. The first patient died at 36 years old of a large tear in his esophagus caused by a sharp piece of bone and crumpled foil in a blood clot. The second patient required a thoracotomy at 10 years of age to remove multiple metal screws that had been ingested. Because of pica, the patient developed bronchiectasis and had recurring chest infections later in life. He required surgery again at 20 years old to remove plastic from his esophagus. He died at 22 years of age due to a chest

infection caused by pica. The final patient had severe vomiting attributed to an obstruction resulting from pica and died at 38 years of age.

In addition to adverse acute and longstanding health outcomes, pica may contribute to social isolation and stigmatization as well as more restrictive residential placement and an increased cost of care (Sturmey & Williams, 2016). A study on the social and recreational characteristics of people with pica living in institutional facilities found that individuals with intellectual disability who engaged in pica were less likely to have supportive relationships with their families and were less likely to participate in social activities, recreation, and a structured day program (Ashworth et al., 2009). Matson and Bamburg (1999) used the Matson Evaluation of Social Skills in Persons with Severe Retardation (MESSIER) to assess the social skills of 45 people with intellectual disability who engaged in pica and found significantly lower scores in positive verbal, positive nonverbal, and general positive domains when compared to persons without pica.

Assessment of Pica

The assessment of pica can be considered in terms of (a) assessment of severity; (b) screening and diagnostic assessment; (c) medical assessment (e.g., to confirm blockage and identify trace metals in blood); and (d) behavioral assessment to identify operant function (Matson et al., 2011).

Pica Severity Sturmey and Williams (2016) published a pica severity index that can be used to classify the health and safety risks posed by an individual's pica. The scale operationally defines five levels of pica severity ranging from mild (1) to life-threatening (5). The scale has not been systematically evaluated in research, but, as noted by Issarraras and Matson (2018), the scale could be clinically useful for describing the nature of a client's pica in communications between service providers as well as for informing research aimed at better understanding the differences in pica presentation across groups of individuals. Table 26.3 displays the operational definitions for each level of severity.

Screening The Screening Tool of Feeding Problems (STEP; Matson & Kuhn, 2001) consists of 23 statements related to feeding problems (e.g., aspiration risk, food refusal, and nutrition-related items). Parents or other stakeholders who know the individual well are asked to rate each statement in terms of frequency and severity. For example, in terms of pica frequency, one statement is "he/she eats or attempts to eat items that are not food," and the respondent is asked to identify how often that behavior has occurred in the last month. Next, the respondent rates how serious the behavior has been during the month on a scale from 0 (no problem) to 2 (caused serious problems). Although the STEP was not intended for the diagnosis of pica, it may be a useful screening tool. Issarraras and Matson (2018) identified several similar informant-based screening tools that contain items related to pica including the Behavior Problems Inventory (BPI; Rojahn et al., 2012) and Conners Comprehensive Behavior Rating

Table 26.3 Pica severity index

Severity	Pica description
Mild	Mouths objects and has swallowed small pieces of paper or strings without choking and passed the items with no known difficulty
Moderate	Mouths objects and has swallowed small pieces of paper, strings, or other items considered non-dangerous in small quantities. Has experienced one or two incidents of choking and coughing up items
Severe	Mouths objects and has swallowed small pieces of paper, strings, or other items considered non-dangerous in small amounts. Has experienced one or two incidents of choking and coughing up items. Has also had X-rays to rule out pica on more than one occasion
Dangerous	Ingests foreign objects during probes at least weekly. History shows several X-rays and documented ingestion of foreign objects considered dangerous (e.g., metal items and jewelry)
Life-threatening	Has had one or more surgeries for the removal of foreign objects and continues to engage in pica at least once every 30–90 days

Scale originally published in Sturmey and Williams (2016) and reprinted in Issarraras and Matson (2018)

Scales (CBRS; Conners, 2008). Programs serving individuals at risk for pica (e.g., institutional facilities) should consider using a screening instrument that includes items that address pica as part of client intake procedures and/or at regular intervals for people at risk of pica.

Diagnostic Assessment Pica can be diagnosed as a standalone disorder but may also be comorbid with other diagnoses, including intellectual and developmental disability. Pica is formally diagnosed by psychological and medical professionals. Although medical assessment procedures (e.g., blood tests and X-rays) may reveal warning signs of pica (e.g., trace metals in blood) or confirm a blockage from a nonfood item, there is not a laboratory or medical test used to diagnosis pica (The National Eating Disorders Association [NEDA], 2018). Instead, pica is diagnosed using criteria outlined in the *Diagnostic and Statistical Manual of Mental Disorders* (American Psychiatric Association, 2013). The DSM-5 classifies pica among feeding and eating disorders and details four diagnostic criteria. Specifically, to be diagnosed with pica, a person must persist in eating nonfood items for at least 1 month. Further, eating nonnutritive items must be developmentally inappropriate. For example, individuals less than 2 years of age would not typically be given a pica diagnosis because mouthing of nonfood items is common at that age. Additionally, the practice of eating nonfood items must be outside of accepted cultural or social practice. For example, individuals who consume earth (mud) as part of a spiritual practice would not be given a diagnosis of pica (Sturmey & Williams, 2016). Finally, when pica occurs in tandem with another medical condition (e.g., pregnancy) or disorder (e.g., intellectual disability), it must be severe enough to warrant individual attention and treatment (American Psychiatric Association, 2013). The World Health Organization's (WHO) International Classification of Diseases, 11th Revision (ICD-11), differentiates between pica that emerges during childhood and pica that emerges later in life but is otherwise similar to DSM-5's description of pica (WHO, 2018).

Medical Assessment Individuals who engage in pica or who are suspected of engaging in pica may benefit from medical assessment. Specifically, the NEDA (2018) suggests that individuals with a pica diagnosis be assessed for pregnancy, anemia, intestinal blockage, and potential toxicity resulting from inedible items (e.g., lead from ingestion of paint chips). Medical tests focused specifically on detecting iron-deficiency anemia, and malnutrition may be particularly valuable. In such cases, pica may stem from a specific nutrient deficiency, which may be addressed with medication or vitamins. For example, Swift et al. (1999) surveyed 152 people in a residential facility who engaged in pica and found a significant association between the occurrence of pica and low levels of zinc and iron.

Functional Behavioral Assessment For people with intellectual disability, pica appears more likely to occur because of contingencies of reinforcement than because of nutritional deficiencies (Matson et al., 2011). For example, an individual may learn that people in their environment (e.g., group home staff, family, and teachers) will react to pica by providing attention, giving a preferred food, or allowing escape from a non-preferred task. A functional analysis is a behavioral assessment designed to identify reinforcement contingencies maintaining a problem behavior so that intervention efforts can be tailored to address those specific contingencies. For example, if a functional analysis identifies attention from staff as a reinforcer maintaining pica, then intervention would focus on providing attention for behaviors other than pica and changing how staff respond to pica in the natural environment (Carter, Wheeler, & Mayton, 2004).

A functional analysis, which may be administered as a standalone assessment or as part of a comprehensive Functional Behavioral Assessment (FBA), involves a series of test conditions that are administered until a pattern of responding across conditions reveals the operant function of a target behavior. A functional analysis may be applied to both problem behavior (e.g., pica) and appropriate behavior, for exam-

ple, throwing nonfood items in the trash (Issarraras & Matson, 2018). A functional analysis is designed to identify both social and automatic (non-social) operant functions. Obtaining preferred stimuli (attention and tangibles) and avoiding unwanted stimuli (e.g., escaping chores) are forms of socially mediated reinforcement that have been demonstrated to maintain or increase the frequency of many topographies of problem behavior, including pica (Ledford et al., 2019). Automatic reinforcement is not socially mediated because the relevant reinforcement contingency is not dependent on the actions of another person. Instead, automatic reinforcement involves obtaining desirable sensations (e.g., pleasurable taste or texture) and/or avoiding undesirable sensations (e.g., reducing hunger pains). Like socially mediated consequences, contingent bodily sensations are capable of maintaining or increasing problem behavior. Automatic reinforcement is the most commonly identified operant function of pica in previous research (Sturmey & Williams, 2016). However, because treatment of automatically reinforced pica differs substantial from treatment of pica maintained by socially mediated reinforcement, an automatic function should not be merely assumed.

In a functional analysis, a behavior's occurrence is measured and compared across a series of assessment conditions. Although functional analysis conditions can be designed to test a wide range of potential reinforcement contingencies, the most common functional analysis conditions include tests for positive reinforcement (e.g., provision of preferred tangibles and attention), negative reinforcement (e.g., escape from task demands), and automatic reinforcement (Beavers, Iwata, & Lerman, 2013). Each assessment condition involves contriving a motivating operation and programming a specific reinforcement contingency related to that motivating operation. For example, to evaluate the extent to which a behavior may be influenced by attention from caregivers, a functional analysis attention condition involves measuring the occurrence of the behavior when another person (usually the assessor) is in the assessment room but is not attending to the individual being assessed. When the individual engages in the behavior, the assessor delivers attention, usually by expressing concern (e.g., "Please do not do that. You might hurt yourself."). Behavior that occurs in the attention condition but does not occur in a control condition, where attention is provided at regular frequent intervals regardless of behavior (i.e., noncontingent attention), is likely maintained by positive reinforcement in the form of attention. Table 26.4 describes the purpose and general procedures for the most common functional analysis test conditions found in the literature.

Typically, each functional analysis test condition is between 5 and 20 minutes in duration, and each condition is administered a minimum of three times, usually in a random (or semi-random) sequence, or until the data paths that denote the occurrences of behavior within a condition diverge to demonstrate experimental control (Beavers et al., 2013). The operant function is determined by comparing the level of behavior in a test condition to the level of behavior in the play (control) condition, where all putative reinforcers are freely available (see Table 26.4).

Previous research has demonstrated that functional analysis procedures can be modified and still accurately identify function (Lydon, Healy, O'Reilly, & Lang, 2012). Functional assessment of pica often requires modification to the typical functional analysis procedures (Donnelly & Olczak, 1994). Specifically, in a traditional functional analysis, a problem behavior is allowed to occur for the purpose of assessment. However, because of health and safety risks inherent to pica, a functional analysis of pica would not typically allow the person to ingest real inedible items. For example, it is clearly unethical to bait an assessment room with sharp metal objects to provide opportunity for acuphagia. Instead, edibles can be disguised to look like the nonfood items typically ingested by the person being assessed. For example, the assessment environment for an individual who engages in cigarette pica might be baited with candy cigarettes as opposed to real cigarettes, and a chocolate bar shaped to look like feces could be used to bait the assessment room for a person who engages in coprophagia (e.g., Donnelly & Olczak, 1994). In

Table 26.4 Functional analysis conditions and basic procedures

Condition	Reinforcement contingency evaluated	Procedures
Attention	Socially mediated positive reinforcement in the form of social interaction	Assessor is available to provide attention but engages in a solitary activity (e.g., reading a magazine or browsing on cell phone) Assessor tells person being assessed to play or engage quietly with items that have been placed in the assessment environment Contingent on occurrence of problem behavior, assessor delivers attention and then returns to their solitary activity All other behaviors are ignored by assessor
Tangible	Socially mediated positive reinforcement in the form of provision of items	Assessor has possession of an item identified as a potential reinforcer (see Kodak, Fisher, Kelley, and Kisamore (2009) to identified reinforcers) for the person being assessed Contingent on occurrence of problem behavior, the assessor gives the item to the individual for 10–30 seconds and then takes it back All other behaviors are ignored by assessor, and attempts to obtain the item in other ways are blocked
Escape	Socially mediated negative reinforcement in the form of avoidance of undesired activities or stimuli	Assessor presents a task demand that has been identified as potentially aversive to the person being assessed (e.g., academic task or chores) Contingent on occurrence of problem behavior, the assessor removes the task materials and turns away from the person to allow escape from the task demand for 10–30 seconds All other behaviors are ignored by assessor
Alone/ ignore	Automatic (non-social) reinforcement	The individual being assessed is alone in the assessment environment and is monitored remotely or covertly for safety and data collection. If remote observation is not possible or if leaving the individual alone presents a safety risk, then the assessor may remain in the room but ignore all behavior (no socially mediated consequences) aside from what may be necessary to prevent injury (referred to as ignore condition) There are no programmed contingencies
Play	Serves as control condition where reinforcers evaluated in other conditions are provided independent (noncontingent) of behavior	The person being assessed has free access to the preferred items used in the tangible condition Assessor does not give any task demands, does not ask any questions, and does not tell the person to do anything Assessor delivers attention on a set schedule (e.g., every 10 seconds) or continually regardless of behavior There are no programmed contingencies for problem behavior Problem behavior is ignored

some cases, nonedible items that have been evaluated by physicians as safe for human consumption in small amounts (e.g., small bits of paper) have been used in the assessment (Kern, Starosta, & Adelman, 2006). Table 26.5 provides examples from previous research wherein pica items have been modified or replaced with disguised edible items in order to conduct functional analyses.

A number of studies have conducted functional analyses to determine the operant function of pica and then used the results to successfully reduced or eliminated pica for people with intellectual disability. For example, Ing et al. (2011) conducted a functional analysis to identify the operant function of coprophagia (ingestion of feces) by a 6-year-old girl with autism and intellectual disability. The functional analysis consisted of test conditions designed to identify positive reinforcement contingencies (i.e., provision of tangibles and attention), negative reinforcement contingencies (i.e., escape from demands), and automatic

Table 26.5 Disguised pica bait items used in previous research

Inedible item ingested in the natural environment	Disguised pica bait[a] or modified pica items[b] used on functional analysis
Feces	Mixture of flour, water, and food coloring
Rocks	Uncooked beans
Cotton/lint	Cotton candy
Rocks	Chocolate, licorice jelly beans
Small twigs	Beef jerky
Dirt	Crumbled pieces of cookies and brownies
Leaves	Lettuce
Cigarettes	Long white pieces of candy (candy cigarettes)
Paper	Small bits of paper deemed safe for consumption by proper professional

[a]The disguised pica bait items in Table 26.4 are described in Donnelly and Olczak (1994); Ing, Roane, and Veenstra (2011)
[b]The modified pica item (small pieces of paper) is described in Kern et al. (2006)

reinforcement. A play condition where toys and attention were freely available and no demands were made of the child was used as the control condition (see Table 26.4). Sessions were 10 minutes each, and the assessment room was baited with a tray of artificial feces (i.e., chocolate bars melted and sculpted to resemble feces). High-preference toys were available in the tangible and toy play condition, and lower-preference toys were used in the attention condition. The rate per minute of coprophagia was measured within each condition and then compared across conditions. Test conditions and the control condition were run three times each (15 sessions), but data paths did not diverge, and the operant function was not initially evident. Because automatic reinforcement contingencies may influence behavior across functional analysis conditions, four consecutive ignore conditions (modified alone conditions) were conducted. Because pica continued to occur in the absence of socially mediated consequences during the extended ignore conditions, it was concluded that coprophagia was maintained by automatic reinforcement. Treatment based on the results of the functional analysis decreased coprophagia to zero and increased the consumption of real food.

Indirect Behavioral Assessment In some cases, it may not be possible to conduct a functional analysis, perhaps due to unavailable expertise, safety concerns, or objections from stakeholders (Lloyd, Weaver, & Staubitz, 2016). In cases where a functional analysis is not possible or desirable, questionnaires capable of indirectly identifying operant function from caregiver responses to items may be indicated. The Questions About Behavioral Function (QABF; Paclawskj, Matson, Rush, Smalls, & Vollmer, 2000) lists 25 questions that can be addressed to a person who knows the individual with pica well (e.g., parent, teacher, or facility staff). The 25 items refer to situations in which the individual might engage in the target behavior or to the potential motives of the individual engaged in the target behavior. For example, items, directed at identifying socially mediated functions, ask if the person "engages in the behavior [pica] to get attention," "to escape learning situations," and to "get access to items such as preferred toys, food, or beverages." Examples of items directed at automatic functions (referred to as "non-social" on the QABF) include engages in the behavior because "he/she is in pain," "even if he/she thinks no one is in the room," and "when he/she is ill." Respondents are asked to answer by scoring each item on a Likert scale from 0 (never occurs) to 3 (often occurs). Items are grouped by the operant function they addressed, and a total score for each group is then summed. Specifically, the QABF provides scores for social reinforcement functions (i.e., attention, escape, and tangible) and for automatic reinforcement (i.e., non-social and physical). The category with the highest total score is likely to be the function of pica for that individual. Previous research has demonstrated that the results of the QABF often align with results from a functional analysis. Further, when applied to pica, the QABF most often suggests an automatic function, which aligns with the most common result from functional analyses of pica (Matson et al., 2005).

In addition to the QABF, the Motivational Assessment Scale and the Functional Analysis Screening Tool (FAST; Iwata, Deleon, & Rosco, 2013) may be used. The MAS and FAST vary in terms of the number and phrasing of questions but are otherwise comparable in process, form, and function to the QABF. The QABF and MAS have both been used to assess pica, and results have been shown to align with direct functional analysis. However, the QABF has a notably supportive evidence base (Matson, Tureck, & Rieske, 2012; Paclawskj et al., 2000) and appears to be used more often than other options in published research (e.g., Matson et al., 2005). It is important to note, however, that both the MAS and QABF are indirect respondent-informed questionnaires and direct observation with experimental control – as in a functional analysis – is likely the most accurate approach (Koritsas & Iacono, 2013) and should be used when possible and acceptable.

Conclusion

Pica is a dangerous behavior that is prevalent in samples of individuals with intellectual disability, particularly among individuals living in institutional facilities. Considerations for the assessment of pica include (a) screening for pica during intake and/or at regular intervals for individuals in residential facilities; (b) assessing for specific medical conditions that may cause pica (nutrient deficiency) or result from pica (e.g., intestinal blockage); (c) rating the severity of pica; and (d) identifying the operant function and reinforcement contingencies maintaining pica. Given the acute and long-term health risks associated with pica, treatment is often a high priority, and treatment informed by functional assessment tends to be more effective (Hurl, Wrightman, Haynes, & Virues-Ortega, 2016). Functional analysis may be the most accurate form of functional assessment, but indirect questionnaires are more efficient because they can be administered in a single session by staff with less expertise and often align with the direct functional analysis results.

References

Ali, Z. (2001). Pica in people with intellectual disability: A literature review of aetiology, epidemiology and complications. *Journal of Intellectual and Developmental Disability, 26*, 205–215.

American Psychiatric Association. (2013). *Diagnostic and statistical manual of mental disorders* (5th ed.). Arlington, VA: American Psychiatric Publishing.

Ashworth, M., Hirdes, J. P., & Martin, L. (2009). The social and recreational characteristics of adults with intellectual disability and pica living in institutions. *Research in Developmental Disabilities, 30*, 512–520.

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.

Carter, S. L., Wheeler, J. J., & Mayton, M. R. (2004). Pica: A review of recent assessment and treatment procedures. *Education and Training in Developmental Disabilities, 39*, 346–358.

Conners, C. K. (2008). *Conners comprehensive behaviour rating scales manual*. North Tonawanda, NY: Multi-Health Systems Inc..

Decker, C. J. (1993). Pica in the mentally handicapped: A 15-year surgical perspective. *Canadian Journal of Surgery/Journal Canadien de Chirurgie, 36*, 551–554.

Donnelly, D. R., & Olczak, P. V. (1994). A placebo stimulus for the assessment and treatment of pica for tobacco. *Behavioral Interventions, 9*, 247–250.

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*, 350–359.

Goh, H. L., Iwata, B. A., & Kahng, S. W. (1999). Multicomponent assessment and treatment of cigarette pica. *Journal of Applied Behavior Analysis, 32*, 297–316.

Hurl, K., Wrightman, J., Haynes, S., & Virues-Ortega, J. (2016). Does a pre-intervention functional assessment increase intervention effectiveness? A meta-analysis of within-subject interrupted time-series studies. *Clinical Psychology Review, 47*, 71–84.

Ing, A. D., Roane, H. S., & Veenstra, R. A. (2011). Functional analysis and treatment of coprophagia. *Journal of Applied Behavior Analysis, 44*, 151–155.

Issarraras, A., & Matson, J. L. (2018). Assessment of pica. In J. L. Matson (Ed.), *Handbook of clinical psychopathology and developmental disabilities assessment*. New York, NY: Springer International Publishing.

Iwata, B. A., Deleon, I. G., & Rosco, E. M. (2013). Reliability and validity of the functional analysis

screening tool. *Journal of Applied Behavior Analysis, 46*, 271–284.

Kamal, I., Thompson, J., & Paquette, D. M. (1999). The hazards of vinyl glove ingestion in the mentally retarded patient with pica: New implications for surgical management. *Canadian Journal of Surgery, 42*, 201–204.

Kern, L., Starosta, K., & Adelman, B. E. (2006). Reducing pica by teaching children to exchange inedible items for edibles. *Behavior Modification, 30*, 135–158.

Kodak, T., Fisher, W. W., Kelley, M. E., & Kisamore, A. (2009). Comparing preference assessments: Selection- versus duration-based performance procedures. *Research in Developmental Disabilities, 30*, 1068–1077.

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*, 747–757.

Ledford, J. R., Barton, E. E., Rigor, M. N., Stankiewicz, K. C., Chazin, K. T., Harbin, E. R., & Taylor, A. L. (2019). Functional analysis and treatment of pica on a preschool playground. *Behavior Analysis in Practice, 12*, 176–181.

Lloyd, B. P., Weaver, E. S., & Staubitz, J. (2016). A review of functional analysis methods conducted in public school classroom settings. *Journal of Behavioral Education, 25*, 324–356.

Lydon, S., Healy, O., O'Reilly, M. F., & Lang, R. (2012). Variations in functional analysis methodology: A systematic review. *Journal of Developmental and Physical Disabilites, 24*, 301–326.

Matson, J. L., & Bamburg, J. W. (1999). A descriptive study of pica behavior in persons with mental retardation. *Journal of Developmental and Physical Disabilities, 11*, 353–361.

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., & Kuhn, D. E. (2001). Identifying feeding problems in mentally retarded persons: Development and reliability of the screening tool for feeding problems (STEP). *Research in Developmental Disabilities, 22*, 165–172.

Matson, J. L., Mayville, S. B., Kuhn, D. E., Sturmey, P., Laud, R., & Cooper, C. (2005). The behavioral function of feeding problems as assessed by the questions about behavioral function (QABF). *Research in Developmental Disabilities, 26*, 399–408.

Matson, J. L., Tureck, K., & Rieske, R. (2012). The questions about behavior function (QABF): Current status as a method of functional assessment. *Research in Developmental Disabilities, 33*, 630–634.

McAdam, D. B., Breibord, J., Levine, M., & Williams, D. E. (2012). Pica. In P. Sturmey & M. Hersen (Eds.), *Handbook of evidence-based practice in clinical psychology, Vol. 1: Child and adolescent disorders*. Hoboken, NJ: John Wiley & Sons.

McLoughlin, I. J. (1988). Pica as a cause of death in three mentally handicapped men. *The British Journal of Psychiatry, 152*, 842–845.

Paclawskj, T. R., Matson, J. L., Rush, K. S., Smalls, Y., & Vollmer, T. R. (2000). Questions about behavior function (QABF): A behavioral checklist for functional assessment of aberrant behavior. *Research in Developmental Disabilities, 21*, 223–229.

Piazza, C. C., Hanley, G., & Fisher, W. W. (1996). Functional analysis and treatment of cigarette pica. *Journal of Applied Behavior Analysis, 29*, 437–450.

Rojahn, J., Rowe, E. W., Sharber, A. C., Hastings, R., Matson, J. L., Didden, R., & Dumont, E. L. M. (2012). The behavior problems inventory-short form for individuals with intellectual disabilities: Part I: Development and provisional clinical reference data: Behavior problems inventory-S: Part I. *Journal of Intellectual Disability Research, 56*, 527–545.

Sturmey, P., & Williams, D. E. (2016). *Pica in individuals with developmental disabilities*. Cham, Switzerland: Springer International Publishing.

Swift, I., Paquette, D., Davison, K., & Saeed, H. (1999). Pica and trace metal deficiencies in adults with developmental disabilities. *British Journal of Developmental Disabilities, 45*, 111–117.

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, 45*, 52–62.

The National Eating Disorders Association. (2018). *Pica*. Retrieved May 1, 2019 from: https://www.nationaleatingdisorders.org/learn/by-eating-disorder/other/pica

World Health Organization. (2018). *International statistical classification of diseases and related health problems* (11th Revision). Retrieved from: https://icd.who.int/browse11/l-m/en

Young, S. L. (2011). *Craving earth-understanding pica: The urge to eat clay, starch, ice and chalk*. New York, NY: Columbia University Press.

Treatment of Anxiety

27

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Introduction

One of the most widely researched areas of psychological intervention has been the treatment of anxiety disorders; however, to date there has been limited research examining the treatment of anxiety disorders in individuals with intellectual disability (ID). This chapter will briefly outline the different anxiety disorders and prevalence rates associated with these disorders in individuals with ID. This chapter then will discuss the research examining evidence-based treatments (e.g., behavioral therapy, cognitive behavioral therapy, applied behavior analysis) for a multitude of anxiety disorders of individuals with ID. We end the chapter by addressing other methods that may be adapted for use with this population. For a more preliminary and detailed description and discussion of ID itself, see Chap. 15.

Anxiety disorders manifest when feelings of anxiety/fear cause distress and impairment in typical situations or around specific stimuli (Barlow, 1988). An individual can be diagnosed with multiple anxiety disorders at the same time or across his or her lifespan (Kendall et al., 2010).

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There are ten different anxiety disorders that are in the *Diagnostic and Statistical Manual of Mental Disorders, 5th Edition (DSM-5).* The anxiety disorders are sequenced developmentally, with the order of disorders reflecting the typical age of onset. They each are briefly outlined below. Anxiety disorders are typically diagnosed when anxiety or fear occurs, interferes with functioning, and is out of proportion to the potential stressor or threat.

Separation anxiety disorder has the earliest onset, typically in young childhood, but it can develop during later adolescence/adulthood (Mash & Barkley, 2014). It is characterized by significant impairment (APA, 2013) due to a consistent, excessive, and developmentally inappropriate amount of anxiety when separating from a major attachment figure (e.g., caregiver) or the home environment itself. The 12-month prevalence of separation anxiety disorder in adolescence is 1.6% and ranges from 0.9% to 1.9% in adults (APA, 2013).

Selective mutism is an uncommon anxiety disorder (point prevalence of 0.3% to 1%). It captures individuals that consistently fail to speak to others in a given social situation where there is an ability and expectation to speak (e.g., school) despite speaking in other situations (e.g., home) (APA, 2013; Mash & Barkley, 2014). This disorder typically leads to deficits in academic or occupational achievement and is marked by high levels of social anxiety (APA, 2013). It is

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especially important to highlight this disorder, because individuals with selective mutism must have the ability to speak. An individual with ID might not be able to verbally communicate at all; thus, this individual would not be considered selectively mute.

Specific phobia is one of the most prevalent anxiety disorders in childhood (12-month prevalence: 5% in children and approximately 16% in adolescents), and the 12-month prevalence rate in adulthood is 3–5% (APA, 2013). A specific phobia develops when an individual fears a particular object or situation, and this fear is inconsistent with the actual level of danger attributed to this object or situation, which causes significant distress and impairment for the individual (e.g., stimulus or situation is avoided or suffered through with intense anxiety or fear) (APA, 2013). In the *DSM-5*, there are five different types of phobias which are identified by specifiers: animal (e.g., insects, dogs), natural environment (e.g., storms, heights), blood-injection-injury (e.g., needles, blood), situational (e.g., closed spaces), and other (e.g., loud sounds, costumed characters, toilets; APA, 2013).

Social anxiety disorder (previously known as social phobia) is characterized by significant impairment due to a consistent and excessive fear of negative evaluation that transcends different social situations (APA, 2013). This marked anxiety of social situations can persist in situations including meeting new people, eating or writing in public, using public restrooms, and/or speaking in public (Reid, Smiley, & Cooper, 2011). The 12-month prevalence rate of social anxiety disorder is 7% (APA, 2013).

Panic disorder is diagnoseable when an individual experiences recurrent panic attacks that are marked by an abrupt surge of intense fear or anxiety that occurs concurrently with at least four physiological symptoms (e.g., sweating, chest pain or discomfort, dizziness, and nausea) (APA, 2013). Additionally, one must experience intense worry about future panic attacks or must make significant changes in behavior related to the attacks (e.g., behaviors to avoid having panic attacks). The 12-month prevalence rate of panic disorder is 2–3% in adolescents and adults (APA, 2013).

The panic attack specifier is different than panic disorder and can occur with any anxiety disorder. A panic attack is a sudden surge of intense fear or discomfort that peaks within minutes and is marked by multiple physical (e.g., feeling dizzy, chest pain or discomfort, sweating) or cognitive (e.g., fear of going crazy or fear of dying) symptoms (APA, 2013). Panic attacks can be expected or can occur unexpectedly, without some specific trigger. The 12-month prevalence rate for panic attacks is 11.2% in adults (APA, 2013).

Agoraphobia (12-month prevalence rate of 1.7%) is defined as intense fear or anxiety in regard to two or more of the following situations: being outside the house alone, standing in a line or being in a crowd, using public transportation, being in open spaces (e.g., parking lots), and/or being in enclosed spaces (e.g., movie theater) (APA, 2013). The individual does not fear the situations, but rather fears an inability to escape these situations. These situations are avoided or suffered through with intense fear or anxiety, and this fear or anxiety is disproportionate to the actual danger posed by the situation (Doerfler, Connor, Volungis, & Toscano, 2007).

Generalized anxiety disorder *(GAD)* is characterized by the persistent, uncontrollable worry about many different events or activities (APA, 2013). Individuals with generalized anxiety disorder tend to experience significant physiological symptoms when they are anxious and tend to overestimate the probability of these anxiety-provoking events actually occurring. The 12-month prevalence rate for generalized anxiety disorder is 0.9% in adolescents and 2.9% among adults (APA, 2013).

Anxiety disorder due to another medical condition is defined as either panic attacks or anxiety that is a prominent feature of the current psychological problem, and there is evidence that panic attacks or anxiety is a direct pathopsychological consequence of a medical condition (APA, 2013). The medical condition must precede the panic attacks or anxiety for it to be diagnoseable. To date, the prevalence rates are unclear, but there

are specific medical conditions that are known to cause anxiety (e.g., asthma, hypertension) (APA, 2013).

Other specified anxiety disorder includes all of the symptoms that are characteristic of an anxiety disorder which causes significant distress and impairment but is only used when the full criteria for that specific anxiety disorder have not been met (APA, 2013). For this diagnostic category to be used, the clinician must communicate the specific reason(s) that full criteria are not met for the given anxiety disorder.

Unspecified anxiety disorder is utilized when the symptoms of anxiety disorder are present and cause significant distress and impairment, but the clinician does not specify the reason that the full criteria have not been met; this includes situations when sufficient information is unable to be obtained to make a specific diagnosis (e.g., in an acute intervention) (APA, 2013).

Research has demonstrated that children and adolescents with ID have significantly higher prevalence rates of emotional and behavior problems compared to typically developing peers (Emerson, 2003; Linna et al., 1999). These rates may be due to deficits in interpersonal coping skills and the overall impairment of the central nervous system (Linna et al., 1999). But the overall prevalence rates (based on epidemiological studies) for psychopathology in individuals with ID vary drastically, with ranges between 10% and 60% compared to rates of 6–17% in typically developing youth (Green, Berkovits, & Baker, 2015; Koskentausta, Iivanainen, & Almqvist, 2002). The variation in prevalence rates may be due to differences in the samples being assessed (e.g., specific subject characteristics such as intelligence quotient scores [IQ], how samples are derived), deficits in the ability of the individual to clearly communicate about his or her symptoms, as well as how the construct of psychopathology is being measured (Dykens, 2000; Unwin, Tsimopoulou, Kroese, & Azmi, 2016). Even so, children with mild to moderate ID are more likely to have specific fears and generalized worry than children without ID (Borthwick-Duffy, Lane, & Widaman, 1997; Ramirez & Kratochwill, 1997). Anxiety disorders have been found to occur at a rate of 8.7% in children ages 5–15 years with co-occurring ID and have reached estimated rates of 22% compared to just 3–7% of typical youth (Emerson & Hatton, 2007; Emerson, 2003).

Rates of specific anxiety disorders in individuals with ID have been examined to a limited extent. In a study examining children aged 5–9 years, children with ID were found to have higher levels of anxiety rated on the *Child Behavior Checklist (CBCL* Achenbach 1991); overall, children with ID were generally found to be four times more likely to be diagnosed with social anxiety disorder compared to typically developing peers (Green et al., 2015). Children with ID show elevated levels of fear related to specific phobias and generalized anxiety disorder as they get older. Dekker and Koot (2003) examined the 1-year prevalence rates of psychopathology in children ages 6–18 with ID. They found that 21.9% of children met diagnostic criteria for any anxiety disorder; more specifically, 17.5% of the children examined met diagnostic criteria for specific phobia (the most common anxiety disorder found in this sample). Additionally, there are genetic syndromes that are typically marked by cognitive deficits that appear to have higher rates of specific anxiety disorders. Individuals with fragile X syndrome and individuals with Angelman syndrome have been found to experience more social anxiety, while individuals with Williams syndrome are more likely to be diagnosed with a specific phobia (Dykens, 2000; Pitts, Klein-Tasman, Osborne, & Mervis, 2016).

There have been mixed results regarding the prevalence of anxiety disorders in individuals with ID by sex. Males with ID have been found to have higher rates of comorbidity, but there appears to be no sex difference in the development of an anxiety disorder (Strømme & Diseth, 2000). Other studies have found no sex differences (Green et al., 2015). Similarly, when considering severity of ID and the co-occurrence of mental disorders, studies have failed to yield consistent results. Most of the research conducted in this area appears to support the notion that the severity of ID does not influence the association with comorbid conditions, although there is mar-

ginal support for an increased risk for anxiety in those individuals with ID who have higher cognitive abilities (Dekker & Koot, 2003; Ehrenreich-May & Remmes, 2013; Strømme & Diseth, 2000).

Anxiety and Intellectual Disabilities

As mentioned, there has been limited research examining the presentation of anxiety disorders in individuals with ID; this could be due to diagnostic overshadowing (as clinicians attribute the anxiety symptoms to the diagnosis of ID as well as individuals with ID presenting with atypical presentations of anxiety disorders; see Unwin et al., 2016). Clinicians have also historically misdiagnosed or left undiagnosed certain psychopathology in individuals with severe or profound ID due to misattributing symptoms of aggression, self-injury, and noncompliance to their developmental disability rather than a separate mental disorder (Matson, Smiroldo, Hamilton, & Baglio, 1997). In children with and without ID, anxiety symptoms can be precipitated by stressful life events (Stavrakaki & Mintsioulis, 1997). Children with ID and comorbid GAD appear to present similarly to those children without ID (Masi, Brovedani, Mucci, & Favilla, 2002). Those with ID and GAD are more likely to also meet diagnostic criteria for panic disorder, while children with ID have been found to have equal rates of anxiety disorders (Masi et al., 2002). One area of research has focused on fear and specific phobias in individuals with ID. As typically developing children mature, the nature and content of a child's fear or anxieties change and morph into fears that are developmentally appropriate (Davis III, Munson, & Tarcza, 2009; Davis III, White, & Ollendick, 2014; Mash & Barkley, 2014). Specific phobias seem to present similarly in individuals with and without ID; however, adults with ID tend to have more concrete fears that are more developmentally similar to those of children (Ramirez & Kratochwill, 1997; Stavrakaki & Lunsky, 2007). Children with ID have been found to more frequently have phobias involving animals (Ehrenreich-May & Remmes, 2013). In adults with ID, much of the research has focused on dog phobia, blood-injection-injury phobia, and acrophobia (Erfanian & Miltenberger, 1990; Hagopian, Crockett, & Keeney, 2001; Hurley, 2004). Less research has focused on social anxiety, but individuals with ID are likely susceptible to developing social anxiety disorder due to high prevalence of social isolation and exclusion (Ehrenreich-May & Remmes, 2013). Individuals with ID who have social anxiety disorder display interpretation biases similar to those individuals without ID who also have social anxiety (Houtkamp, van der Molen, Salemink, de Voogd, & Klein, 2017).

Regardless of which anxiety diagnosis an individual may meet criteria for, there are fortunately many treatment techniques that can be helpful. However, in planning appropriate treatment, clinicians should consider the client's foundational skills necessary to engage in any one particular intervention. For example, ID may make it more difficult for a client to engage in the cognitive components of therapy due to one's overall level of impairment; more profoundly impaired individuals may not possess the language skills needed to communicate clearly and may have a poor understanding of emotions (Joyce, Globe, & Moody, 2006). Typically, individuals with ID selected for treatment studies tend to vary based on differences in impairment (mild/moderate to severe/profound). Most studies examining treatment in this population focus on individuals with mild to moderate ID. This is mainly a function of the ability to differentiate between thoughts, emotions, and behavior, with more profoundly impaired individuals having the most difficulty with these skills (Hassiotis et al., 2013).

However, the growing literature surrounding the treatment of people with intellectual disabilities has provided clinicians with various empirically supported options. Moreover, the increased prevalence of anxiety among those with ID has provided a considerable range of treatment options. Regardless of the level of intellectual disability (i.e., mild, moderate, severe, profound), evidence-based treatments and techniques for

anxiety should be considered and used if possible. Modifying treatment based on the severity of ID hopefully provides the client with a custom treatment plan allowing for a higher success rate; however, practitioners should be cautious of overextending treatments designed specifically for typically developing populations (Davis III, 2012). There are various options available depending on cognitive ability as well as adaptive functioning (e.g., verbal abilities).

Behavioral Treatment

Behavior therapy has enjoyed high success rates among individuals with low cognitive functioning. In a review of the literature, Hagopian and Jennett (2008) found that the majority of the studies focusing on ID objectively defined anxiety as avoidance of situations and stimuli, while prescribed treatment was graduated exposure and reinforcement. According to APA Divisions 12 and 16, graduated exposure with the use of reinforcement is included as an empirically supported treatment for populations with intellectual disabilities (Jennett & Hagopian, 2008). Jennett and Hagopian (2008) termed this avoidance as "phobic avoidance" stating that little literature focuses on other aspects of anxiety disorders outside of avoidance for individuals with intellectual disabilities.

Hagopian and Jennett (2008) have previously recommended a variety of tools for assessing an individual's fear hierarchy through nonverbal methods. The behavioral avoidance task (BAT; for reviews see Castagna, Davis III, & Lilly, 2017 and Davis III et al., 2013) is a widely used behavioral observation tool administered to assess avoidant behavior associated with a specific fear. The observation includes structured exposure to a feared stimulus to detect the level at which the person will attempt to avoid the fear-provoking stimuli. This behavioral method can be implemented with those with ID across a variety of severity levels. While BATs are primarily used as an assessment tool for typically developing individuals with anxiety, the qualities that allow the BAT to nonverbally assess an individual's fear of a situation can be useful in building a fear hierarchy further allowing the behavioral modification process to be successful. The fear hierarchy should include many variations of the feared stimulus. For example, if the individual fears dogs, the fear hierarchy could include variations of the individual approaching the dog with increasingly shorter distances or various levels of touching the dog. Natural observations can also be used to determine the severity of the behavior around the avoidant stimulus. While this may be difficult to observe naturally, a parent or caregiver may be helpful in reporting the anxious behavior (Hagopian & Jennett, 2008). See Chap. 16 for more information regarding assessing anxiety for populations with intellectual disabilities.

A systematic preference assessment may be beneficial in nonverbally assessing the quality of various reinforcers for use with an individual (Hagopian, Long, & Rush, 2004). Hagopian, Long, and Rush (2004) described preference assessments as identifying a person's preferred stimuli, which can function as reinforcers through indirect assessment or direct assessment (see Fig. 27.1). Indirect assessment may include parent, caregiver/guardian, direct care staff, or advocate interviews as well as client interviews if the individual is verbal. A direct preference assessment may include systematically exposing the individual to the stimuli to determine which is preferred. The direct preference assessment can be broken down further into approach-based procedures (recording whether they approach the stimuli alone or in conjunction with another stimulus) and engagement-approach procedures (recording how long the individual engages with the stimuli). The systematic assessment of preferred stimuli allows contingent reinforcers to be placed into behavior therapy regardless of cognitive function and verbal abilities. In addition to a preference assessment, a reinforcer assessment may be necessary. A reinforcer assessment determines the effects of the reinforcer on the level of engagement or amount the targeted behavior is performed when provided contingently (Hagopian, Long, & Rush, 2004). This may be necessary to determine if the preferred stimuli are actually effective during the exposure sessions.

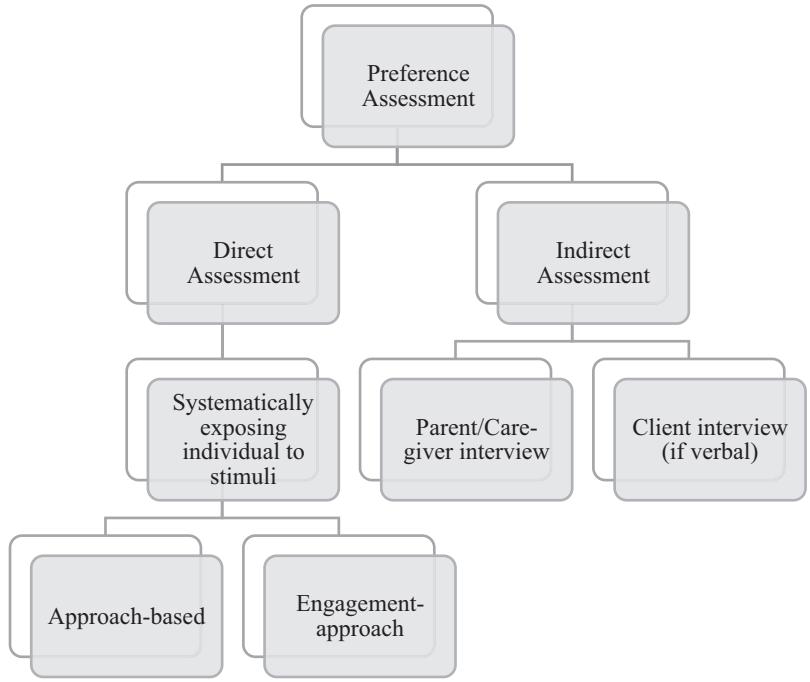

Fig. 27.1 Types of preference assessments. (Adapted from Hagopian, Long, & Rush, 2004)

Once a fear hierarchy and the reinforcers are established, the main components of behavior therapy can be implemented, including exposure and reinforcements. Exposure therapy is an evidenced-based treatment for anxiety and phobias (American Psychiatric Association [APA], 2013). Exposure therapy focuses on reducing the fear a stimulus provokes; there are multiple forms of exposure therapy including graduated live exposure, imaginal, virtual reality, and in vivo exposure (Craske, Treanor, Conway, Zbozinek, & Vervliet, 2014; Davis III, May, & Whiting, 2011; Davis III & Ollendick, 2005).

Graduated exposure among typically developing individuals with anxiety includes being gradually exposed to fear-inducing stimuli of increasing intensity as the patients are able to handle their fear. The strength of the exposure will vary on some physical nature (e.g., duration of exposure, proximity, mode of presentation; Hagopian, Lilly, & Davis, 2017). The continuous building onto the fear hierarchy can ultimately lead to extinction of the avoidance of the feared stimuli. The current step should be mastered before moving on to the next, although the complete absence of fear does not need to be accomplished (Hagopian & Jennett, 2008). There is contradicting evidence on whether flooding is beneficial. In fact, learning to experience anxiety without avoidance while working through the hierarchy will be beneficial to the individual (Chorpita, 2007).

Systematic desensitization can be an alternative option when using exposure, particularly with individuals who are limited in their verbal abilities. This form of exposure has been successfully used with people who have various disabilities, even with nonverbal individuals (Williams, Lewis, Marcham, & Palicka, 2018). Systematic desensitization is the process of systematically exposing an individual to feared stimuli in a least to most fashion. During this process, the individual should not be experiencing fear or anxiety (Davis III, 2009; Davis III et al., 2011; Davis III & Ollendick, 2005). The lack of fear or anxiety extinguishes the link between the feelings and the feared stimuli, therefore reducing the individual's overall avoidance (Burton, Palicka, & Williams, 2017). It should be noted that while similar to graduated exposure, systematic desensitization should be absent of said fear, while an individual may experience fear through graduated exposure.

When working with individuals who are intellectually disabled, it may be difficult to distinguish if the individual has an absence of fear, especially if the individual is nonverbal. Careful consideration should be taken to insure the individual is not being flooded by increasing the level of the hierarchy too quickly.

Similar to treatment with typically developing individuals, the goal of therapy should be adjusted based on the individual and his or her capabilities. Goals may be set based on concrete stimuli, such as the behavioral aspects of anxiety or fear or level of avoidance. Presentations of behaviors should be considered when assessing for effectiveness of the graduated exposure or systematic desensitization while working through the hierarchy. Individuals with ID and comorbid anxiety present symptoms in a variety of ways. Characteristics may include appearance of distress, agitation or aggression, physical resistance or avoidance, yelling, crying, or destructive behavior. Being informed on the client's presentation of symptoms (and for a unique, particular individual) can be key since these behaviors may be present while working through the graduated exposure, which can ultimately indicate if the treatment is being performed correctly or if progress is being made (for more information on the presentation of anxiety symptoms in populations with intellectual disabilities, see Chap. 16).

In addition to graduated exposure, Hagopian and Jennett (2008) have suggested the importance of including reinforcements. In anxiety disorders, avoidance or escape-maintained behavior often acts as a negative reinforcer. For example, if someone is anxious around a stimulus, when he or she avoids the stimulus, his or her anxiety goes down. Therefore, the avoidance or the alleviation of symptoms is a negative reinforcer of fear: the presence of the stimulus. For typically developing individuals, implementing a strong reinforcer contingent on the exposure can eliminate the negative reinforcement the individual may have in place regarding the feared stimuli. The same may be true for populations with ID. For verbal individuals, picking reinforcers can be done by having the individual name them, while for nonverbal individuals, a preference assessment may need to be conducted. After performing the preference assessment, the positive reinforcers (e.g., edible or tangible items) should be administered on a contingent on increased approach responses (Hagopian et al., 2017). It is important to be cognizant of the effectiveness of chosen reinforcers. The potency of some reinforcers may fade over time so reassessment of potent reinforcers may be necessary.

Other behavioral treatment components may be useful to treat anxiety in populations with ID. Modeling, which is based on learning principles, allows the individual to observe and learn from someone else engaging a fear-provoking stimulus (Hagopian & Jennett, 2008). Conyers et al. (2004) used video modeling to treat avoidance of dental procedures in adults with severe to profound ID to watch a person they were familiar with engage in the procedures appropriately while being praised. Modeling can also be observed live (Love, Matson, & West, 1990), by either a person the client is familiar with or by the clinician in session. Observations of exposure and the extinction of avoidance can aid the individual in understanding how to encounter the fear hierarchy. Despite the method of modeling, the important distinction is that the component should not be used in a stand-alone manner, but rather in addition to graduated exposure and reinforcement.

Additionally, prompting has been touted as an additional treatment component to graduated exposure and reinforcement. Prompting includes assisting an individual to comply with the exposure hierarchy either through verbal (e.g., "touch the stimuli") or physical (e.g., holding hands, touching arm) prompts (Hagopian & Jennett, 2008). Prompting is often done in systematic desensitization. Systematic desensitization includes a least-to-most prompting hierarchy, which involves prompting a client to move closer to the feared stimuli while engaging in reinforcing activities (Davis III et al., 2009). In a treatment conducted for specific phobia of dogs in two individuals with moderate to profound ID, Erfanian and Miltenberger (1990) used a "least-to-most" prompting hierarchy to guide the clients in their exposure. For the two individuals

with dog phobias, the clients started the first session with the use of modeling by watching the owner interact with his dog while engaging in preferred activities on the opposite side of the room. In the next sessions, the clients were prompted verbally to move closer to the dog and then prompted physically to move closer to the dog while engaging in the preferred activities. Erfanian and Miltenberger (1990) brought the clients' attention to the dog with each increased level of the hierarchy and then praised the clients for their participation. The least-to-most prompting proved to be successful in treating the specific phobia of dogs in the two individuals with intellectual disabilities (Erfanian & Miltenberger, 1990). Prompting may be a beneficial component for treatment of all severity levels of ID despite language and cognitive skills as there are a wide variety of prompting methods that can be used with this population (MacDuff, Krantz, & McClannahan, 2001).

Response prevention can also be used to increase interactions with the fear-provoking stimuli. Response prevention refers to preventing escape-maintained behavior as well as encouraging the client to engage with the fear-provoking stimuli. Prompting is used to discourage the client to escape the situation and instead complete the current level of the fear hierarchy. Once the current step of the exposure is complete, the individual should be reinforced (Hagopian & Jennett, 2008). Rapp, Vollmer, and Hovanetz (2005) used response prevention while treating adolescents with severe ID for behaviors associated with avoidance of swimming pools. The individual was physically directed to approach the pool until he or she was able to physically enter the pool. Prompting was used as response prevention for any escape-attempting behavior. As the individual got closer in proximity to the pool (e.g., progressing on the hierarchy), reinforcers (e.g., edible or tangible items) were provided (Rapp et al., 2005). No other published studies have included response prevention in the treatment of anxiety for individuals with intellectual disabilities. Further research should be conducted to determine the utility of response prevention for individuals with intellectual disabilities.

Distracting stimuli are also typically used in systematic desensitization. Individuals participating in treatment are allowed to engage with distracting stimuli during exposure to increase or maintain their level of relaxation. The access to the distracting stimuli should not be contingent on anything and should not be used as the reinforcement for the exposure (Hagopian & Jennett, 2008). Distracting stimuli should include items that the individual enjoys (e.g., toys, books, media) so that the stimuli continue to keep him or her engaged. In a study by Luscre and Center (1996), individuals in treatment were given distracting stimuli during systematic desensitization to treat avoidance of dental procedures. Items used as distracting stimuli included music and toys which were effectively used to increase relaxation in the distressing situation. There are various reasons why distracting stimuli might be effective. The distracting stimuli may take away attention from the fear-provoking stimulus, increase the reinforcement for participating in exposure, and align the anxiety-provoking situation with something more preferred. While the distracting stimuli may be beneficial, careful consideration should be taken when deciding to incorporate this behavioral component to treatment. Since the individual is being provided complete access to his or her preferred item, there is the possibility of contingent reinforcements being weakened or being less effective in encouraging the individual in the exposure altogether (Hagopian & Jennett, 2014).

Ehrenreich-May and Remmes (2013) have suggested that the use of relaxation techniques is an effective behavioral treatment component for individuals with intellectual disabilities. progressive relaxation (Jacobsen, 1938) and abbreviated progressive relaxation (Bernstein & Borkovec, 1973) have been successfully applied to individuals with ID, which include muscle relaxation and deep breathing to treat phobic avoidance (Guralnick, 1973; Peck, 1977). Relaxation techniques have shown success being implemented in combination with other behavioral treatment components; however, when implemented alone, there has only been success shown in individuals with mild ID (Rickard, Thrasher, & Elkins,

1984). For more severe levels of ID, Behavioral Relaxation Training (BRT) has been found to be more effective than other relaxation techniques (Lindsay, Baty, Michie, & Richardson, 1989). With BRT, the instructor models the tense and relaxed states so that the individual will mimic the actions and is then reinforced for doing so (Ehrenreich-May & Remmes, 2013).

Relaxation techniques can be effective since the individual will be focusing on the physical sensations occurring, which can be a more concrete approach. The individual should be walked through deep breathing as well as tensing and relaxing muscles in his or her body. With limited cognitive functioning, it may be beneficial to have the individual to bring a stress ball into session so that there is a clear understanding of what tensing means. The individual should then be instructed to focus on the physical sensation of releasing the tensed hand so that he or she can then perceive what relaxing means. These relaxation techniques can be applied to other body parts as well and should be used as an additional component to graduated exposure and reinforcement.

Cognitive Treatment

There has been a recent trend to utilize more cognitive treatment approaches with individuals with ID in addition to both behavioral and pharmacological interventions. One of the main types of evidence-based treatments for anxiety disorders in the typical population is cognitive behavioral therapy (CBT) (APA, 2013; National Institute for Health and Clinical Excellence, 2011). CBT is based on a cognitive model that suggests that an individual has dysfunctional cognitions (e.g., beliefs or thoughts that might automatically come into his or her mind) that lead to particular reactions, both physical (e.g., heart racing), emotional (e.g., feeling very nervous), and behavioral (e.g., avoiding a particular situation) (see Fig. 27.2) (Beck, 2011). The goals of CBT are to combine cognitive (mental) processes with behavioral techniques in order to aid an individual in gaining new perspectives of their underlying beliefs about themselves and the world around them it is a collaborative and goal-oriented approach to problem-solving (Beck, 2011) (see Fig. 27.2). Typical treatment using CBT can include techniques such as graduated exposure, social skills training, assertiveness training, conflict resolution, and stress management techniques, such as relaxation and visual imagery (Beck, 2011). Individual and group CBT has been used to treat individuals with ID with a variety of comorbid mental disorders (Hassiotis et al., 2013).

Research has supported the use of CBT in individuals with ID to varying degrees. Willner and colleagues (2002) found that those adults with moderate ID who received CBT for anger management showed a decrease in both their self-report and caregiver or caretaker report on measures of anger and provocation compared to a control group. Willner and colleagues (2002) concluded that more research is needed to determine the efficacy of CBT with this population, given the inconsistency of this sample's understanding of the cognitive components of treatment; qualitatively they report that clients had difficulty understanding the concept of cognitive restructuring. Individuals with ID may be able to develop an understanding of the more complex cognitive concepts with time and modification to the existing protocol as well as patience and collaboration with the treatment provider. Within the context of treatment, there has been some debate as to which skills (e.g., understanding and labeling emotions, ability to communicate, understanding of the cognitive model) an individual with ID needs to learn in psychotherapy. Moreover, there is evidence to suggest that individuals with ID can be successful in CBT as long as they have the support of the clinician teaching them the necessary skills.

Generally, there have been more studies conducted examining the use of CBT for individuals with ID targeting a variety of disorders, compared to those specifically targeting anxiety-related difficulties. Four studies have been conducted using CBT in individuals with ID and comorbid anxiety disorders; one study was a quantitative design using individualized adapted CBT (Lindsay, 1999), another one was random-

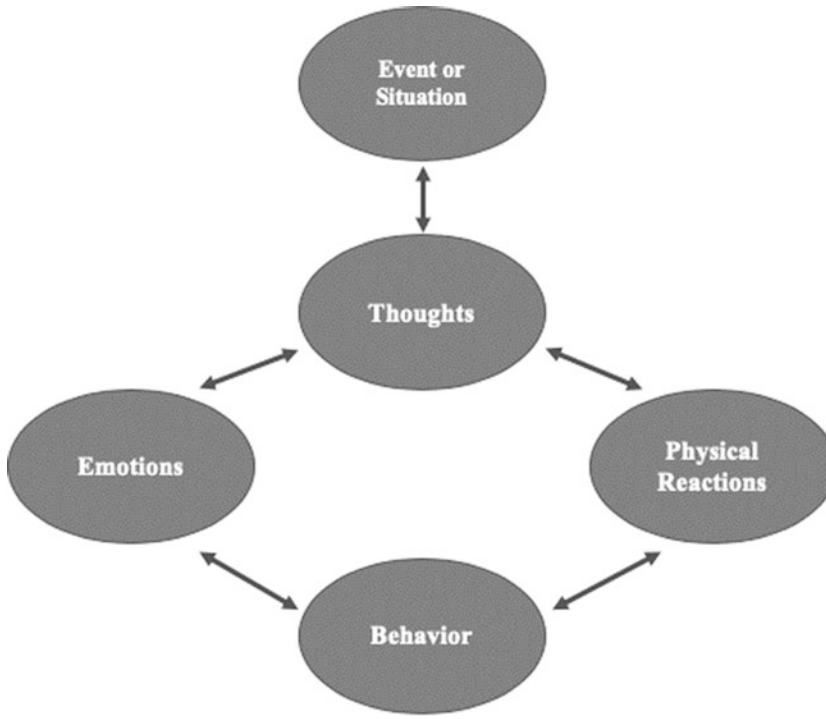

Fig. 27.2 Cognitive behavioral therapy model. (Adapted from Beck, 2011)

ized controlled trial using manualized individual CBT (Hassiotis et al., 2013), and the two other studies were mixed method designs using group CBT (Douglass, Palmer, & O'connor, 2007; Marwood & Hewitt, 2013). All of the studies had pre-post-, and follow-up assessments to measure the anxiety levels of the clients and were relatively small in sample sizes (7–15 clients, although the study conducted by Hassiotis and colleagues featured 32 clients). Hassiotis et al. (2013) conducted a feasibility randomized controlled trial using a manualized individual CBT with adults with mild to moderate ID and demonstrated that this intervention was acceptable to use with this population. Studies including those individuals with ID who have mixed clinical presentations (anxiety and/or depression) have had varying results.

There are, however, many limitations to the current research in this area. First, the inability to use standardized manuals or protocol to deliver CBT complicates the efficacy work since most applications of CBT with individuals with ID need to be modified; there are minimal modified manualized CBT manuals currently being used (Unwin et al., 2016). The small sample sizes are a second limitation which underpower the results; larger sample sizes would also allow for better generalization of the current findings. Lastly, the present research suggests the need for additional studies that focus on the efficacy of cognitive treatment in individuals with ID to ensure reliability.

However, there has been increasing literature suggesting that populations with mild to moderate ID can participate in modified CBT to treat anxiety with success. In a review by Dagnan, Jackson, and Eastlake (2018), the most common CBT components used to treat anxiety in a population with ID included psychoeducation and approaches to counter beliefs or anxious thoughts. Moree and Davis (2010) also reviewed types of modifications to CBT for anxiety in population with autism spectrum disorder, which have been shown to be successful. Treatment for typically developing individuals would include countering cognitive

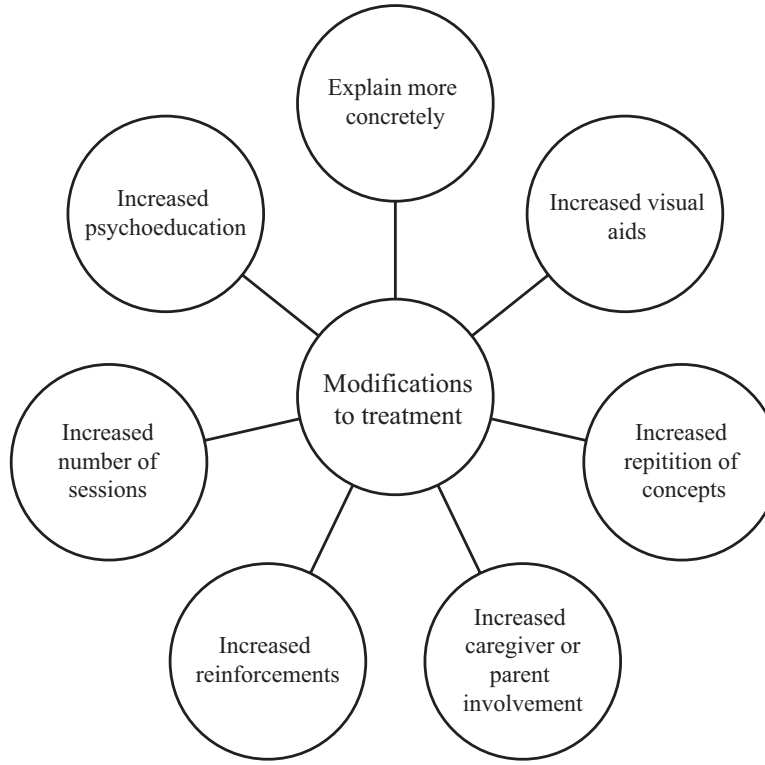

Fig. 27.3 Modifications to treatment for individuals with ID. (Adapted from Davis, Saeed, & Antonacci, 2008)

distortions through Socratic dialogue and positive self-talk. Individuals participating in this evidence-based treatment use cognitive challenging by rationalizing the evidence to deter unrealistic thoughts that often are at the root of anxiety disorders (Beck, 1995). Because the cognitive component is such a vital part of CBT, it was previously thought that the use of this treatment would not be efficacious in populations with ID due to difficulties with language and cognitive abilities (Taylor, Lindsay, & Willner, 2010). Emerging research has argued that some of these basic skills can be taught to individuals with mild ID so that the discrimination between thoughts, feelings, and behaviors can occur (Vereenooghe, Gega, Reynolds, & Langdon, 2016). Roberts and Kwan (2018) also propose that modified CBT has been shown to be successful as well.

There are various modifications to the treatment of anxiety that can be effective for individuals with ID (see Fig. 27.3). Davis et al. (2008) suggested that individuals with ID may need things explained more concretely. Often in CBT, anxiety-provoking situations are parsed out into thoughts, feelings, and behaviors. This may be difficult for someone with limited cognitive ability. Providing a concrete scenario through the use of movies, books, or materials eliminates the extra step of self-generating the examples. Having concrete examples of how to react in a situation may be helpful as well (e.g., watching movies with appropriate social interactions for social anxiety or relating the situation to favorite game or cartoon) particularly in learning the differences between an appropriate reaction and an overreaction (Ehrenreich-May & Remmes, 2013). Drawing concepts in the form of graphs or other visual aids may be helpful in linking thoughts and feelings with a clear connection between the two so that the differences may be emphasized. Focusing on the physiology of the anxiety symptoms instead of the cognitions may be also more successful since physiological symptoms are more concrete (Davis et al., 2008).

Psychoeducation is another major component of treatment, which becomes even more crucial when working with populations with ID. Psychoeducation may include what constitutes

anxiety as well as what creates and maintains the avoidance behaviors (Moskowitz et al., 2017). Psychoeducation should be provided in a concrete manner. For example, it is important to explain emotions or feelings in psychoeducation when working with an individual with anxiety; however, this may be more complex when working with an individual with ID. Drawing emotions or having stickers with facial expressions may be a beneficial way to describe something slightly abstract in a more concrete manner. Operational definitions of emotions may help bridge the gap between what is being taught and the individual's actual understanding of the concepts. The length of psychoeducation may be extended since the grasping of each concept might take longer or need to be broken down into smaller steps. Another modification is to increase repetition of the concepts over a greater number of sessions. Previous case studies have used a range of 20–40 sessions of cognitive behavioral therapy for populations with intellectual disabilities. The extension of sessions allows the concepts to be repeated in order to ensure each technique has been mastered (Davis et al., 2008).

Treatment modifications may also include increased participation and involvement of the parent or caregiver (Davis et al., 2008). While most child CBT programs have a parent component, this may need to be amplified for populations with ID. The parent or caregiver may be involved in all or most sessions, which allows the skills to be cued or used in other settings. Increased psychoeducation should be implemented for the parent or caregiver as well as the individual receiving treatment. Psychoeducation may include simple and brief information or something more creative such as a psychoeducational game or quiz (Dagnan et al., 2018). Caregiver involvement may also increase compliance of the individual with exposure to the feared stimulus. The parent or caregiver should be an active agent in fostering the utilization of the techniques outside of the treatment. Additional treatment modifications presented are increased reinforcement, which is related to the behavioral component of cognitive behavioral therapy through operant conditioning.

Donoghue, Stallard, and Kucia (2011) created a modified approach to CBT for children with autism spectrum disorder (ASD), which can be applied to some populations with ID. The publication used the acronym PRECISE to detail the modifications needed for CBT to accommodate someone with ASD. The "P" represents collaborative partnership vital to utilize a person's strengths and weaknesses. The "R" represents the right developmental or cognitive level; the "E" represents the empathy required; "C" represents the creativity needed in the approach to keep the individual's interest; "I" represents the investigation and experimentation; "S" represents self-discovery to determine what the individual already knows; and the "E" represents making the experience enjoyable for the individual (Donoghue et al., 2011). The acronym may be applicable when modifying treatment for an individual with ID experiencing anxiety (depending on cognitive abilities).

Klein et al. (2018) suggested that cognitive bias modification training can be successful for individuals with mild ID, particularly for treating social anxiety. Interpretational bias, or the tendency to inappropriately analyze ambiguous situations or events which can increase anxiety, can be reduced through cognitive bias modification training (CBM-I). CBM-I does not rely on cognitive reflection, meta-cognition, or other abstract concepts resulting in a more appealing treatment for those with mild ID. In CBM-I training, individuals learn to restructure the way they analyze ambiguous situations, particularly those that are increasing their anxiety, through computerized pictures. Klein et al. (2018) tested the effects of cognitive bias modification for interpretation bias on 69 socially anxious adolescents with mild ID and found that there was a reduction in negative interpretation bias in just 5 sessions over a 3-week period where they covered 40 items in each session. CBM-I coupled with modifications to treatment (e.g., increased number of sessions) may result in even higher reduction of symptoms; however, more research should be conducted to determine the effectiveness as current research is limited.

Pharmacotherapy

There is limited research using pharmacotherapy to treat anxiety for those having ID (Ehrenreich-May & Remmes, 2013). While some research suggests that optimal treatment may be the combination of therapy and pharmacological treatment similar to CAMS, the same approach may not be optimal for populations with ID experiencing anxiety. Primary medications often prescribed for anxiety include serotonin reuptake inhibitors (SSRIs), serotonin-norepinephrine reuptake inhibitors (SNRIs), benzodiazepines, and buspirone (Vanin & Helsley, 2008). Davis et al. (2008) conducted a review of studies looking at the effectiveness of treating anxiety among youth with pervasive developmental disorders (PDDs). The researchers identified only three studies looking at the effects of SSRIs, which found positive effects of SSRIs with the PDD population. These findings were consistent among all levels of intellectual functioning (mild, moderate, severe, and profound). Anxiolytics are often prescribed to people with intellectual disabilities to treat disruptive behavior as well as symptoms of generalized anxiety disorder (Aman, Collier-Crespin, & Lindsay, 2000). Due to limited literature on the ID population, careful considerations should be made when determining treatment components (e.g., pharmacotherapy, behavioral therapy, etc.) for anxiety when working with individuals with intellectual disabilities.

Conclusion

This chapter has explored the various options available to treat anxiety in individuals with ID. Behavioral therapy has been shown to be effective in treating anxiety among individuals with ID through the use of graduated exposure, systematic desensitization, and reinforcement (Hagopian & Jennett, 2008). Reinforcers may be assessed through preference assessments (Hagopian, Long, & Rush, 2004). Additional treatment components have shown promise when paired with graduated exposure and reinforcement, such as relaxation, prompting, modeling, response prevention, and distracting stimuli (Ehrenreich-May & Remmes, 2013; Hagopian & Jennett, 2008). Although behavioral treatment has been and continues to be the primary mode of treatment for individuals with ID, growing literature has suggested promise in modifying CBT as well.

There are important ethical considerations to consider during the use of behavioral treatment for anxiety among individuals with ID, particularly for individuals who are nonverbal. Increased psychoeducation may help eradicate distress when at the forefront (Ehrenreich-May & Remmes, 2013). The behavioral treatment should be taken slowly and move along the fear hierarchy while mastering each level. It may be helpful to repeat the rationale behind the treatment at the start of each session (Ehrenreich-May & Remmes, 2013). When conducting systematic desensitization, the individual, regardless of cognitive abilities, should feel minimal to no fear at any point in the treatment if done correctly (Davis III et al., 2009).

Due to the necessary cognitive abilities for CBT, modified CBT should only be attempted in individuals with ID in the mild to moderate range with verbal abilities. Modifications to CBT may include increased sessions, concrete explanations, increased parental or caregiver involvement, or increased reinforcements (Moree & Davis, 2010). CBT should involve the differentiation between thoughts, emotions, and behaviors and the ability to distinguish between distressing thoughts and realistic thoughts. For those with more severe or profound ID, this may not be feasible (Hassiotis et al., 2013). For populations with more severe deficits, the use of behavioral treatments without the cognitive component may be more effective, particularly if the individual is nonverbal.

Regardless of the treatment for anxiety used, pharmacotherapy should be carefully considered before implementation due to the limited research among populations with ID. Current research has shown that SSRIs can be effective when treating anxiety for an individual with intellectual disabilities regardless of severity levels (i.e., mild, moderate, severe, profound). However, there are

only three published studies reviewing this dynamic; the effects may not be completely accurate. The use of pharmacotherapy should always be consulted with a licensed physician.

Future directions should include extending the literature on various techniques effective for treating anxiety among populations with ID. The current literature surrounding the treatments has been growing and further examining treatment moderators and mechanisms of change. However, it still lags behind the studies of those who are typically developing.

References

Achenbach, T. M. (1991). The Child Behavior Checklist—1991. Burlington: University of Vermont

Aman, M. G., Collier-Crespin, A., & Lindsay, R. L. (2000). Pharmacotherapy of disorders in mental retardation. *European Child & Adolescent Psychiatry, 9*, 198–1107.

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

Barlow, D. H. (1988). *Anxiety and its disorders: The nature and treatment of anxiety and panic*. Guilford press.

Beck, J. S. (1995). *Cognitive therapy: Basics and beyond*. New York, NY: The Guilford Press.

Beck, J. S. (2011). *Cognitive behavior therapy: Basics and beyond*. New York, NY: Guilford press.

Bernstein, D., & Borkovec, T. D. (1973). *Progressive relaxation training*. Champaign, IL: Research Press.

Borthwick-Duffy, S. A., Lane, K. L., & Widaman, K. F. (1997). Measuring problem behaviors in children with mental retardation: Dimensions and predictors. *Research in Developmental Disabilities, 18*(6), 415–433.

Burton, P., Palicka, A., & Williams, T. I. (2017). Treating specific phobias in young people with autism and severe learning difficulties. *The Cognitive Behaviour Therapist, 10*(21), 1–8.

Castagna, P., Davis, T. E., III, & Lilly, M. (2017). The behavioral avoidance task with anxious youth: A review of procedures, properties, and criticisms. *Clinical Child and Family Psychology Review, 20*(2), 162–184.

Chorpita, B. F. (2007). *Modular cognitive-behavioral therapy for childhood anxiety disorders*. New York, NY: Guilford Press.

Conyers, C., Miltenberger, R. G., Peterson, B., Gubin, A., Jurgens, M., Selders, A., et al. (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. https://doi.org/10.1901/jaba.2004.37-233

Craske, M. G., Treanor, M., Conway, C. C., Zbozinek, T., & Vervliet, B. (2014). Maximizing exposure therapy: An inhibitory learning approach. *Behaviour Research and Therapy, 58*, 10–23.

Dagnan, D., Jackson, I., & Eastlake, L. (2018). A systematic review of cognitive behavioural therapy for anxiety in adults with intellectual disabilities. *Journal of Intellectual Disability Research, 62*(11), 974–991.

Davis, E., Saeed, S. A., & Antonacci, D. J. (2008). Anxiety disorders in persons with developmental disabilities: Empirically informed diagnosis and treatment. *Psychiatric Quarterly, 79*, 249–263.

Davis, T. E., III. (2009). PTSD, anxiety, and phobias. In J. Matson, F. Andrasik, & M. Matson (Eds.), *Treating childhood psychopathology and developmental disorders* (pp. 183–220). New York, NY: Springer Science and Business Media, LLC.

Davis, T. E., III. (2012). Where to from here for ASD and anxiety? Lessons learned from child anxiety and the issue of DSM-5. *Clinical Psychology: Science and Practice, 19*, 358–363.

Davis, T. E., III, May, A. C., & Whiting, S. E. (2011). Evidence-based treatment of anxiety and phobia in children and adolescents: Current status and effects on the emotional response. *Clinical Psychology Review, 31*, 592–602.

Davis, T. E., III, Munson, M. S., & Tarcza, E. V. (2009). Anxiety disorders and phobias. In J. L. Matson (Ed.), *Social behavior and skills in children* (pp. 219–243). New York, NY: Springer Science + Business Media.

Davis, T. E., III, & Ollendick, T. H. (2005). Empirically supported treatments for specific phobia in children: Do efficacious treatments address the components of a phobic response? *Clinical Psychology: Science and Practice, 12*, 144–160.

Davis, T. E., III, Reuther, E., May, A., Rudy, B., Munson, M., Jenkins, W., & Whiting, S. (2013). The Behavioral Avoidance Task using Imaginal Exposure (BATIE): A paper-and-pencil version of traditional in vivo behavioral avoidance tasks. *Psychological Assessment, 25*, 1111–1119.

Davis, T. E., III, White, S. W., & Ollendick, T. H. (2014). Handbook of Autism and Anxiety. New York: Springer.

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.

Doerfler, L. A., Connor, D. F., Volungis, A. M., & Toscano, P. F. (2007). Panic disorder in clinically referred children and adolescents. *Child Psychiatry and Human Development, 38*(1), 57–71.

Donoghue, K., Stallard, P., & Kucia, J. (2011). The clinical practice of Cognitive Behavioural Therapy with children and young people with a diagnosis of Asperger's Syndrome. *Clinical Child Psychology and Psychiatry, 16*, 89–102.

Douglass, S., Palmer, K., & O'connor, C. (2007). Experiences of running an anxiety management group for people with a learning disability using a cognitive behavioural intervention. *British Journal of Learning Disabilities, 35*(4), 245–252.

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.

Ehrenreich-May, J., & Remmes, C. S. (2013). Treatment of childhood anxiety in the context of limited cognitive functioning. In *Handbook of treating variants and complications in anxiety disorders* (pp. 149–161). New York: Springer.

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.

Emerson, E., & Hatton, C. (2007). Mental health of children and adolescents with intellectual disabilities in Britain. *Britain Journal of Psychiatry, 191*, 493–499.

Erfanian, N., & Miltenberger, R. G. (1990). Contact desensitization in the treatment of dog phobias in personal who have mental retardation. *Behavioral Residential Treatment, 5*, 55–60.

Green, S. A., Berkovits, L. D., & Baker, B. L. (2015). Symptoms and development of anxiety in children with or without intellectual disability. *Journal of Clinical Child & Adolescent Psychology, 44*(1), 137–144.

Guralnick, M. J. (1973). Behavior therapy with an acrophobia mentally retarded young adult. *Journal of Behavior Therapy and Experimental Psychiatry, 4*, 263–265.

Hagopian, L. P., Crockett, J. L., & Keeney, K. M. (2001). Multicomponent treatment for blood-injury-injection phobia in a young man with mental retardation. *Research in Developmental Disabilities, 22*(2), 141–149.

Hagopian, L. P., Long, E. S., & Rush, K. S. (2004). Preference assessment procedures for individuals with developmental disabilities. *Behavior Modification, 28*, 668–677. https://doi.org/10.1177/0145445503259836

Hagopian, L. P., & Jennett, H. K. (2008). Behavioral assessment and treatment of anxiety in individuals with intellectual disability and autism. *Journal of Developmental and Physical Disabilities, 20*(467), 467–483.

Hagopian, L. P., & Jennett, H. K. (2014). Behavioral assessment and treatment of anxiety for those with autism spectrum disorder. In T. E. Davis, S. W. White, & T. H. Ollendick (Eds.), *Handbook of autism and anxiety* (pp. 155–169). New York, NY: Springer.

Hagopian, L. P., Lilly, M., & Davis, T. E. (2017). Behavioral treatment for anxiety in minimally verbal children with ASD. In C. Kerns, P. Renno, E. A. Storch, P. Kendall, & J. Wood (Eds.), *Anxiety in children and adolescents with autism spectrum disorder* (pp. 193–210). Academic Press.

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.

Houtkamp, E. S., van der Molen, M. J., Salemink, E., de Voogd, E. L., & Klein, A. M. (2017). Interpretation biases in socially anxious adolescents with a mild intellectual disability. *Research in Developmental Disabilities, 67*, 94–98.

Hurley, A. D. (2004). Treatment of multiple phobias and agoraphobia in a man with Down syndrome. *Mental Health Aspects of Developmental Disabilities, 7*(4), 142–148.

Jacobsen, E. (1938). *Progressive relaxation*. Chicago, IL: University of Chicago Press.

Jennett, H. K., & Hagopian, L. P. (2008). Identifying empirically supported treatments for phobic avoidance in individuals with intellectual disabilities. *Behavior Therapy, 39*(2), 151–161.

Joyce, T., Globe, A., & Moody, C. (2006). Assessment of the component skills for cognitive therapy in adults with intellectual disability. *Journal of Applied Research in Intellectual Disability, 19*, 17–23.

Kendall, P. C., Compton, S. N., Walkup, J. T., Birmaher, B., Albano, A. M., Sherrill, J., … Keeton, C. (2010). Clinical characteristics of anxiety disordered youth. *Journal of Anxiety Disorders, 24*(3), 360–365.

Klein, A. M., Salemink, E., de Hullu, E., Houtkamp, E., Papa, M., & van der Molen, M. (2018). Cognitive bias modification reduces social anxiety symptoms in socially anxious adolescents with mild intellectual disabilities: A randomized controlled trial. *Journal of Autism and Developmental Disorders, 48*, 3116–3126.

Koskentausta, T., Iivanainen, M., & Almqvist, F. (2002). Psychiatric disorders in children with intellectual disability. *Nordic Journal of Psychiatry, 56*(2), 126–131.

Lindsay, W. R. (1999). Cognitive therapy. *Psychologist, 12*(5), 238–241.

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*(2), 129–140.

Linna, S. L., Moilanen, I., Ebeling, H., Piha, J., Kumpulainen, K., Tamminen, T., & Almqvist, F. (1999). Psychiatric symptoms in children with intellectual disability. *European Child & Adolescent Psychiatry, 8*(4), S77–S82.

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.

Luscre, D. M., & Center, D. B. (1996). Procedures for reducing dental fear in children with autism. *Journal of Autism and Developmental Disorders, 26*, 527–546.

MacDuff, G. S., Krantz, P. J., & McClannahan, L. E. (2001). Prompts and prompt-fading strategies for people with autism. In C. Maurice, G. Green, & R. M.

Foxx (Eds.), *Making a difference: Behavioral intervention for autism* (pp. 37–50). Austin, TX: PRO-ED.

Marwood, H., & Hewitt, O. (2013). Evaluating an anxiety group for people with learning disabilities using a mixed methodology. *British Journal of Learning Disabilities, 41*(2), 150–158.

Mash, E. J., & Barkley, R. A. (Eds.). (2014). *Child psychopathology*. Guilford Publications.

Masi, G., Brovedani, P., Mucci, M., & Favilla, L. (2002). Assessment of anxiety and depression in adolescents with mental retardation. Child *Psychiatry and Human Development, 32*, 227–237. https://doi.org/10.1023/A:1017908823046

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.

Moree, B., & Davis, T. E. (2010). Cognitive-behavioral therapy for anxiety in children diagnosed with autism spectrum disorders: Modification trends. *Research in Autism Spectrum Disorders, 4*, 346–354.

Moskowitz, L. J., et al. (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.

National Institute for Health and Clinical Excellence (2011). Clinical guidelines. Retreived from https://www.ncbi.nlm.nih.gov/books/NBK11822/

Peck, C. L. (1977). Desensitization for the treatment of fear in the high level adult retardate. *Behaviour Research and Therapy, 15*(2), 137–148.

Pitts, C. H., Klein-Tasman, B. P., Osborne, J. W., & Mervis, C. B. (2016). Predictors of specific phobia in children with Williams syndrome. *Journal of Intellectual Disability Research, 60*(10), 1031–1042.

Ramirez, S. Z., & Kratochwill, T. R. (1997). Self-reported fears in children with and without mental retardation. *Mental Retardation, 35*(2), 83–92.

Rapp, J. T., Vollmer, T. R., & Hovanetz, A. N. (2005). Evaluation and treatment of swimming pool avoidance exhibited by an adolescent girl with autism. *Behavior Therapy, 36*, 101–105.

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.

Rickard, H. C., Thrasher, K. A., & Elkins, P. D. (1984). Responses of persons who are mentally retarded to four components of relaxation instruction. *Mental Retardation, 22*(5), 248–252.

Roberts, L., & Kwan, S. (2018). Putting the C into CBT: Cognitive challenging with adults with mild to moderate intellectual disabilities and anxiety disorders. *Clinical Psychology Psychotherapy, 25*, 662–671.

Stavrakaki, C. & Mintsioulis, G. (1997). Implications of a clinical study of anxiety disorders in persons with mental retardation. *Psychiatric Annals, 27*(3): 182–189.

Stavrakaki, C., & Lunsky, Y. (2007). Depression, anxiety and adjustment disorders in people with intellectual disabilities. In N. Bouras & G. Holt (Eds.), *Psychiatric and behavioural disorders in intellectual and developmental disabilities* (2nd ed., pp. 113–130). Cambridge, UK: Cambridge University Press.

Strømme, P., & Diseth, T. H. (2000). Prevalence of psychiatric diagnoses in children with mental retardation: Data from a population-based study. *Developmental Medicine & Child Neurology, 42*(4), 266–270.

Taylor, J. L., Lindsay, W. R., & Willner, P. (2010). CBT for people with intellectual disabilities: Emerging evidence, cognitive ability and IQ effects. *Behavioural and Cognitive Psychotherapy, 36*, 723–733.

Unwin, G., Tsimopoulou, I., Kroese, B. S., & 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*, 60–75.

Vanin, J. R., & Helsley, J. D. (Eds.). (2008). *Anxiety disorders: A pocket guide for primary care*. Totowa, NJ: Humana Press.

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.

Williams, T. I., Lewis, S., Marcham, L., & Palicka, A. (2018). Treatment of dog phobia in young people with autism and severe intellectual disabilities: An extended case series. *Contemporary Behavioral Health Care, 3*(1), 1–5.

Willner, P., Jones, J., Tams, R., & Green, G. (2002). A randomized controlled trial of a cognitive-behavioural anger management group for clients with learning difficulties. *Journal of Applied Research in Intellectual Disabilities, 15*, 224–235.

28

Depression Treatment Evidence and Application to Individuals with Intellectual Disability

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Depression is the leading cause of disability in the general population of adults worldwide (Smedley, Stith, & Nelson, 2003; Office of the Surgeon General, 2001; Curren et al., 2018), with 1-year major depressive episode prevalence rates of 6.7% among US adults in 2016, representing over 16 million individuals (Ahrnsbrak, Bose, Hedden, Lipari, & Park-Lee, 2017). The cost to society is also large and growing. The economic burden due to depression was estimated at $83.1 billion in 2000, increasing to over $200 billion in 2010. These figures reflect direct costs associated with depression and comorbid conditions, in addition to lost workplace productivity (Greenberg, Fournier, Sisitsky, Pike, & Kessler, 2015).

At one point viewed as a disorder of adulthood, clinical professionals now widely recognize depression as a disorder affecting adolescents and children as well. Prevalence rates for depressive disorders in 2016 for adolescents was estimated at 12.7%, representing over three million US adolescents aged 12–17 (Ahrnsbrak et al., 2017). While rates among pre-pubertal children are much lower, it is critical not to overlook depression in the youngest demographic, with estimated prevalence rates around 1–1.5% (Garber, Gallerani, & Frankel, 2009).

Depression alone can lead to significant distress in persons of all ages, and effects are further compounded with psychiatric comorbidity. For individuals with intellectual disability (ID), characterized by significantly below average IQ, interference with functional behaviors, and pre-adult onset, depression is one of the most common psychological disorders (Austin, Hunter, Gallagher, & Campbell, 2018; Hamers, Festen, & Hermans, 2018). Prevalence estimates of depression comorbid with ID vary considerably, with adult rates mostly falling in the 2–8% range (Hamers et al., 2018). Notably, some researchers have found much higher rates, with prevalence estimates as high as 64%, based on self-report cut-scores, which tend to yield higher prevalence rates (Austin et al., 2018). It should also be noted that these prevalence rates found by Austin et al. (2018) were derived from individuals in the developmental stage of emerging adulthood, which is generally a stressful developmental transition with high rates of depressive disorders, and potentially even more so for individuals with ID. With regard to children and adolescents, prevalence rates among youth meeting criteria for ID follow similar trends as those of their typically developing peers, with higher rates for adolescents than children. Maïano et al. (2018)

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conducted a meta-analysis to calculate the pooled prevalence estimates for depression in youth with ID. Estimates for major depressive disorder in adolescents and children were 5.7% (95% CI 2.4–13.1) and 3.2% (95% CI 1.0–9.4), respectively (Maïano et al., 2018).

As depression can lead to significant distress and impairment, treatment for depression is critical and a range of options for treatment modalities is available; however, there is an imbalance in the evidence available for individuals with depression. The largest research base comes from the general adult population, followed by typically developing adolescents, and much less for children with depression. Research specific to adults with comorbid ID and depression lags far behind, with even fewer studies focused on adolescents and children in this population. There is a clear need to address this gap with an increased focus on designing and implementing rigorous studies to assess the efficacy of various interventions in individuals with ID. Nonetheless, practical considerations warrant examining the broader literature on depression treatment efficacy, considering its applicability to individuals with ID, and comparing this with the few studies that focus on depression treatment research for those with an ID diagnosis.

This chapter begins with an overview of depression treatment efficacy research in the general adult population followed by the same for children and adolescents. This is followed by a review of treatment studies for depression among adults and youth with ID. In each of these sections, evidence for psychosocial and pharmacological interventions are presented. Finally, each section also explores developmental considerations pertinent to depression course and potentially relevant for treatment outcomes.

Adult Depression Treatment

Psychosocial Interventions

Within the realm of psychotherapy research, interventions for depression have received the lion's share of attention (Cuijpers, Karyotaki, Reijnders, & Ebert, 2019). Two interventions in particular have been the focus of much of this research: cognitive behavioral therapy (CBT) and interpersonal psychotherapy (IPT). In this next section, descriptions of and evidence for both interventions are presented.

Cognitive Behavioral Therapy Cognitive behavioral therapy (CBT) is a short-term, often manualized, psychotherapy that has been established as an efficacious treatment for adult depression (Craighead, Miklowitz, & Craighead, 2008; Cuijpers et al., 2013). While CBT is sometimes described as a unitary treatment approach, in reality, it includes a wide range of interventions. Compton et al. (2004) note that CBT interventions have five characteristics in common: (1) a reflection of the scientist-practitioner approach, which selects treatments based on evidence; (2) a comprehensive idiographic assessment of presenting problems and the situational, cognitive, and behavioral factors that lead to or maintained the target symptoms; (3) psychoeducation; (4) problem-specific treatments that aim to reduce the target symptoms; (5) relapse prevention and strategies and interventions to promote generalization. In these ways, CBT for depression aims to address maladaptive cognitions and behaviors, which typically include helplessness, hopelessness, and social withdrawal and maintains the goal of promoting positive relationships.

CBT is the most widely researched modality of psychotherapy for adult depression and therefore carries the greatest weight of evidence (Cuijpers et al., 2019; Cuijpers et al., 2013). A meta-analysis of 115 randomized controlled trials (RCTs) examining treatment for adult depression compared CBT versus no treatment, CBT versus control groups, CBT versus other psychotherapies, and CBT versus pharmacotherapy (Cuijpers et al., 2013).

The authors found that comparison of CBT and control conditions (wait-list, CAU, placebo, and other control groups) yielded a large effect size in favor of CBT (g = 0.71), indicating that CBT is more effective than control conditions for the treatment of adult depression (Cuijpers et al.,

2013). However, the authors cautioned that the large effect size may have been overestimated due to the effects of publication bias. In fact, later research yielded support for this hypothesis. Cuijpers et al. (2019) examined the mean effect size for CBT based on studies with a low bias risk, which excluded over three quarters of the 192 CBT trials they identified. Based on their analyses, mean CBT effect sizes were small.

The presently described meta-analysis indicated that CBT was found to be no more or less effective than the other evaluated psychotherapy modalities, which included nondirective supportive therapy, behavioral activation (BA) therapy, psychodynamic psychotherapy, interpersonal psychotherapy (IPT), problem-solving therapy (PST), and other psychotherapies (Cuijpers et al., 2013). However, due to limitations of the evidence base, some comparisons between psychotherapies were based on a very small number of studies: supportive therapy (n = 3), behavioral activation (BA) therapy (n = 8), psychodynamic psychotherapy (n = 5), interpersonal psychotherapy (IPT) (n = 5), problem-solving therapy (PST) (n = 3), and other psychotherapies (n = 9).

No difference in efficacy was found between CBT and pharmacotherapy when compared directly for the treatment of adult depression (Cuijpers et al., 2013). While this finding differs from earlier meta-analyses that found CBT to be superior to anti-depressants (Dobson, 1989; Gloaguen, Cottraux, Cucherat, & Blackburn, 1998), these studies may have over-estimated the efficacy of CBT relative to medication due to earlier methodological factors (Butler, Chapman, Forman, & Beck, 2006). Despite the similar effect sizes of CBT and pharmacotherapy for the treatment of adult depression immediately post-treatment, CBT may be more effective than pharmacotherapy when evaluating long-term outcomes. While the present study only evaluated short-term outcomes, the authors point out that a meta-analysis of longer-term effects of CBT (Vittengl, Clark, Dunn, & Jarret, 2007; Dobson et al., 2008) reported that individuals treated with CBT for depression experienced lower relapse rates at 1- and 2-year follow-up than individuals treated with medication alone. Consistent with the research base, the combination of CBT and pharmacotherapy is superior to pharmacotherapy alone for the treatment of adult depression (g = 0.49) (Cuijpers et al., 2013).

In sum, the extant literature, heavily weighted toward CBT, has historically highlighted this intervention as an efficacious treatment for depression. However, these results are attenuated when meta-analytic procedures exclude studies with a higher bias risk, a wait-list control, and which were conducted in non-Western countries (due to an effect of country type on effect size) (Cuijpers et al., 2019). This leaves a much smaller pool of studies from which to draw conclusions about the ultimate efficacy of CBT alone, which is even further reduced when trying to make head-to-head comparisons against other interventions that also have low risk of bias. Nonetheless, CBT effect sizes were not inconsequential and reported to be in the small range (Cuijpers et al., 2019). When pharmacotherapy is factored in, the research suggests similar outcomes between CBT and medications, with CBT seemingly providing greater relapse prevention. Finally, combination treatments that include CBT and pharmacotherapy may provide the strongest results.

Interpersonal Psychotherapy Interpersonal psychotherapy (IPT) was developed as a time-limited, weekly outpatient treatment for major depressive disorder (de Mello, de Jesus Mari, Bacaltchuk, Verdeli, & Neugebauer, 2005). IPT helps patients to identify how depression may be initiated and maintained by social relationships and interpersonal contexts. Examples of these social contexts that may contribute to depression include the loss of a loved one, relocating, and relationship disputes (Weissman et al., 2005). Skills are developed to help individuals identify problematic patterns of dealing with others and adopt new and more helpful interpersonal skills to manage relationships. IPT focuses on patients' immediate social context and conceptually links presenting symptomatology with one or more of the following four domains: (1) unresolved grief, (2) role transitions, (3) interpersonal role disputes, or (4) interpersonal deficits (Frank &

Spanier, 1995). A primary interpersonal problem area linked to the current depressive episode is identified, and interpersonal skill-development is tailored to this target area with the intention that skills will generalize to other relationships and create positive change (Weisz & Kazdin, 2017). For example, a person experiencing an interpersonal role dispute will be taught first to identify whether the relationship is at the stage of renegotiation, impasse, or dissolution. The therapist then guides the individual in selecting appropriate response patterns and action plans related to interpersonal goals. In comparison, an emphasis on transitions includes working on such tasks as skill acquisition to navigate new roles, emotion expression, and developing new supports. Finally, work on interpersonal deficits is centered around identifying problematic socialization patterns and adopting new ones, improving social skills, and building self-efficacy in interpersonal relationships (Weissman, 2005).

A meta-analysis of 38 RCTs of treatment for adult depression compared IPT versus no treatment or usual care, IPT versus other psychotherapies, and IPT versus pharmacotherapy (Cuijpers et al., 2011). In comparison with a waitlist control, placebo control, and usual care, IPT evidenced a significant, medium effect size ($n = 16$, $d = 0.63$, 95% CI = 0.36 to 0.68). While not statistically significant, the authors also found that IPT demonstrated a larger effect when compared with a waitlist control than when compared with studies that employed usual care or other control conditions. When compared with other psychotherapies, IPT was not shown to be significantly more effective ($n = 13$, $d = 0.04$, 95% CI = −0.14 to 0.21). When IPT was compared with pharmacotherapy, a nonsignificant effect size favored pharmacotherapy ($n = 5$, $d = -0.12$, 95% CI = −0.36 to 0.12). Subgroup analyses indicated that selective serotonin reuptake inhibitors (SSRIs) were significantly more effective than IPT, whereas tricyclic antidepressants were significantly less effective than IPT, illustrating important differences in efficacy between these classes of medications. IPT alone was compared with combination treatment of IPT and pharmacotherapy, and the difference between these treatments was not statistically significant. The authors suggested that the nonsignificant result may have been affected by the small number of studies and consequent low statistical power.

In conclusion, based on the meta-analysis by Cuijpers et al. (2011), IPT performed similarly to other forms of psychotherapy, as well as in comparison to pharmacotherapy alone or combination with IPT. However, further examination revealed that IPT may demonstrate better results than some antidepressants, but not others. Finally, as expected, IPT was found to be more effective than waitlist control, placebo control, and usual care.

Psychopharmacological Interventions

While both pharmacological and non-pharmacological treatment options are available for adult depression, antidepressants are used more often than psychological interventions because of the relatively fewer required resources to administer them (Cipriani et al., 2018). A systematic review and meta-analysis of 522 double-blind RCTs compared the efficacy of 21 antidepressant medications to placebo and also compared the medications against one another in "head-to-head trials." Head-to-head trials produced more variability in efficacy than when antidepressants were evaluated against placebo, and all antidepressants evaluated were shown to be more efficacious than placebo in adults, with modest summary effect sizes (Cipriani et al., 2018).

In evaluating the acceptability of these medications based on patient attrition, fluoxetine (Prozac; Sarafem) and agomelatine (Melitor; Thymanax; Valdoxan) were shown to be significantly more acceptable than placebo, and clomipramine (Anafranil) was shown to be significantly less acceptable than placebo. The other medications were not shown to be significantly different than placebo in terms of acceptability. In evaluating the efficacy of these medications, agomelatine (Melitor; Thymanax; Valdoxan),

amitriptyline (Elavil), escitalopram (Lexapro), mirtazapine (Remeron; Remeronsoltab), paroxetine (Paxil; Pexeva; Brisdelle), venlafaxine (Effexor), and vortioxetine (Trintellix; Brintellix) were found to be more effective than the other antidepressants evaluated (range of ORs 1.19–1.96), whereas fluoxetine (Prozac; Sarafem), fluvoxamine (Luvox), reboxetine (Edronax), and trazodone (Oleptro) were found to be the least efficacious drugs evaluated (OR 0.51–0.84) (Cipriani et al., 2018).

When considering efficacy and acceptability in conjunction, escitalopram, mirtazapine, paroxetine, agomelatine, and sertraline had a relatively higher response and lower dropout rate than the other medications evaluated. By contrast, reboxetine, trazodone, and fluvoxamine were associated with generally inferior efficacy and acceptability profiles compared with the other antidepressants evaluated (Cipriani et al., 2018). These results can be used to guide the initial choice of antidepressant medication for adults with major depressive disorder. It is further important to note that acceptability in drug trials could only be examined for patients who found at least some acceptability in a medication approach to depression treatment, as those who find pharmacological interventions unacceptable would not be included in such studies.

Lifespan Developmental Considerations

While this section focuses on efficacy evidence for adults with depression, it is important to appreciate the variability found within this broad developmental range and how such variability may influence treatment efficacy. Many biological, psychosocial, identity, and role changes occur during adulthood. Researchers have found it fruitful at times to consider adulthood as encompassing three distinct developmental categories due to differences that have been observed across biological, psychological, and social domains at different stages of adulthood.

Emerging adulthood is the period from 18 to 25 in which young adults experience biological changes in the brain that increase emotional regulation (Galambos, Barker, & Krahn, 2006). Emerging adults navigate many developmental tasks that often lead to role changes and identity exploration during this time. While the prevalence of major depressive disorder is highest during the teen years, the rates of depression appear to be less in adulthood comparatively. Studies have shown significant decreases in depression in students from their senior year of high school to 1-year post-graduation and have also shown a steady decrease in depressive symptoms from ages 19 and 20 to the mid-30s (Galambos et al., 2006).

During the period of emerging adulthood, general psychological well-being and self-esteem tend to increase, as depressive symptoms and anger decrease. In addition, the gender divergence that marks adolescent depression reverses during this time. Whereas girls experience higher rates of depression in adolescence than boys, by emerging adulthood, the prevalence rates of depression across both sexes improve, with women improving at faster rates than men and therefore evidencing a convergence of prevalence rates among the sexes. This depression gender gap is narrowest between ages 18 and 30, yet diverges again after age 30, which may point to differential impacts of life transitions such as employment, housework, child care, and economic hardship that put women at a disadvantage (Galambos et al., 2006).

Midlife is often conceptualized through seemingly disparate lenses, both as a time of peak functioning for adults and also as a time of crisis for adults (Lachman, 2004). This variability may point to differential gains and losses that adults in midlife experience in domains such as cognitive functioning, personality, emotions, social relationships, work, and physical health. During midlife, rates of major depression tend to increase with age. Kesser and colleagues (2004) showed that differential exposure to stress, rather than differential stress reactivity, explains the relationship between age and stress (Lachman, 2004). Gender moderates the relationship between biopsychosocial factors in midlife and the incidence of depression: for men, work and finances have

the most important associations with depression, whereas for women, health, family relationships, work, and finances all contribute significantly to depression. Marital separation or divorce in midlife is associated with elevated risk for depression, though these effects are greater for men than they are for women. Being unemployed in midlife is associated with elevated risk for depression over being employed or a homemaker, though there are no effects of retirement or parental status on depression. In summary, life circumstances in midlife play an important role in the incidence of depression, and the effect of these circumstances on rates of depression for men versus women varies by domain.

While depressive symptoms in late life are less frequent than in midlife, depression may be the most frequent cause of emotional suffering in older adults, significantly decreasing their quality of life (Blazer, 2003). Among community-dwelling older adults, prevalence rates of clinically significant depressive symptoms range from approximately 8% to 16%. When considering depression in late life, it is important to consider the high frequency of medical comorbidities, especially in the oldest elderly adults. Depression in the medically ill in late life is common, and it is therefore important to consider the medical illness when conceptualizing the etiology of depression at this stage.

In terms of treatment, antidepressant medications provide the most commonly used foundational treatment for late-in-life depression. While virtually all antidepressant medications are equally effective for treating serious major depression, the drugs fluoxetine (Prozac; Sarafem), sertraline (Zoloft), paroxetine (Paxil; Pexeva; Brisdelle), citalopram (Celexa), and fluvoxamine (Luvox) have been specifically demonstrated to be efficacious for older adults. Acceptability and efficacy of antidepressants for older adults are greater in the context of more severe depression than in the context of less severe depression. It should also be noted that antidepressants have been found to be less efficacious in this demographic according to meta-analytic data (Calati et al., 2013), which may be due to age-related neurological changes (Ribeiz et al., 2013). However, not all researchers have replicated the finding of age-related declines in treatment efficacy (Tunvirachaisakul et al., 2018).

Despite greater acceptability of psychotherapy in late life adults than adults in midlife, psychotherapy remains an infrequently prescribed therapy for older adults with depression due to the reticence of physicians to prescribe it. When psychotherapy is employed, research suggests that older adults tend to prefer psychotherapists who embody an educator role over those who adopt the reflective posture of more psychodynamically-oriented psychotherapies (Blazer, 2003). Furthermore, the effectiveness of psychotherapy for this cohort appears comparable to other age groups (Haigh, Bogucki, Sigmon, & Blazer, 2018).

Child and Adolescent Depression Treatment

Psychosocial Interventions

The American Psychiatric Association and the American Academy of Child and Adolescent Psychiatry recommend that psychotherapy be used either alone or in conjunction with pharmacological interventions to treat youth depression (Birmaher et al., 2007). Cognitive behavioral therapy (CBT) and interpersonal psychotherapy (IPT) are considered among the most effective psychotherapy approaches for youth depression (Weersing, Jeffreys, Do, Schwartz, & Bolano, 2017). Current guidelines stipulate that evidence-based approaches, such as CBT and IPT, be used at milder levels of depression, although nondirective supportive therapy and symptom monitoring in a primary care setting may be sufficient in some cases (Cheung, Kozloff, & Sacks, 2013; Zuckerbrot, Cheung, Jensen, Stein, & Laraque, 2018). However, for moderate and severe symptom presentations, practice parameters suggest that pharmacotherapy and psychotherapy are best used in combination (Birmaher et al., 2007).

Cognitive Behavioral Therapy CBT is an often-recommended treatment for youth depres-

sion that can be viewed from a social learning perspective (Compton et al., 2004). It is a present-focused, problem-oriented, and skills-based treatment. The theory holds that depression is caused and maintained by how a child or adolescent perceives situations and events, as well as the presence of emotional and behavioral skills deficits. These deficits may include low involvement in pleasant activities, poor problem-solving and assertion skills, and cognitive distortions. Therefore, symptom reduction can be achieved with treatments that modify patterns of behavior through skills acquisition and altering patterns of cognition.

CBT for child and adolescent depression is supported by the American Academy of Child and Adolescent Psychiatry, and numerous clinical trials have demonstrated its efficacy compared to waitlist control (Clarke, Rohde, Lewinsohn, Hops, & Seeley, 1999; Lewinsohn, Clarke, Hops, & Andrews, 1990; Rosselló & Bernal, 1999; Smith et al., 2015) and monitor and control conditions (Poppelaars et al., 2016). However, even though professional guidelines recommend psychological therapy as a first-line treatment for youth depression, a recent meta-analysis examining psychological therapies for youth internalizing and externalizing disorders and problems found that psychological treatments for depression had relatively small effect sizes. These findings clearly underscore the need to enhance the effectiveness of existing treatments perhaps by incorporating a greater focus on surrounding systems, like families and schools, that have the potential to strengthen their impact.

Adolescent depression tends to be associated with a high risk of relapse. In terms of long-term efficacy of CBT, in trials of CBT for adolescent depression, post-treatment gains are typically maintained in studies with shorter follow-up periods (9 months or less) than for longer follow-up periods (9 months to 2 years). Variables that have been shown to predict relapse or lack of recovery include conflict between parents and adolescents (Birmaher et al., 2000), comorbidity at post-treatment (e.g., anxiety disorders, oppositional defiant disorder) (Vostanis, Feehan, & Grattan, 1998), depression severity (Birmaher et al., 2000), low self-esteem (Vostanis, Feehan, Grattan, & Bickerton, 1996), parental depression (Clarke et al., 2002), and subsyndromal or not fully resolved depression (Brent, Birmaher, Kolko, Baugher, & Bridge, 2001).

Given the greater prevalence of adolescent, as opposed to pre-adolescent depression, much of the work on treatment has focused on adolescents. While some of what works for adolescent youth may be effective for pre-adolescent youth, there is reason for caution and a need to consider developmental factors in implementing treatment. First, compared to adolescent youth, pre-adolescent children are more deeply embedded in family contexts, relying on parents and guardians to identify relevant concerns, interact with larger social structures, access resources, and advocate on their behalf. Individually-focused treatments need to educate caregivers on depression and its treatment, to integrate them in efforts to promote generalization of learned skills, and to assist them in altering the environment (e.g., reducing stressors, introducing supports) to promote recovery. Second, preadolescent children are developmentally limited cognitively, restricted in their ability to abstract and generalize, and may not be able to apply learned strategies to novel situations. Practitioners are further responsible for evaluating what milestones children and adolescents may or may not have achieved in terms of their cognitive and social-emotional development and to aid in recharting the youth's appropriate developmental trajectory. In implementing CBT interventions with younger children, behavioral components, rather than cognitive elements, may be more amenable to the limited cognitive skills of younger children.

Interpersonal Psychotherapy Interpersonal psychotherapy for adolescents (IPT-A; Mufson, Dorta, Moreau, & Weissman, 2004a) was adapted from interpersonal psychotherapy for depressed adults (Klerman, Rounsaville, Chevron, Neu, & Weissman, 1984) and is based on the interpersonal theory of depression. Relationships with those around us are critical to our well-being

(Bowlby, 1978) and depression may in part result from disruptions in those relationships. Adolescence is a time in which relationships are particularly important, yet are often challenging (Jacobson & Mufson, 2010). As a result, treatments that target relational processes may be particularly warranted in youth depression given substantial evidence that interpersonal stressors are strongly linked with youth depression symptoms (Rudolph et al., 2000; Sheeber, Davis, Leve, Hops, & Tildesley, 2007). Specifically, peer and parent conflict are related to elevated depressive symptoms in youth (Klomek, Marrocco, Kleinman, Schonfeld, & Gould, 2007; Sheeber et al., 2007), and IPT-A works through intervening in problematic interpersonal relationships that may be contributing to mood symptoms. The three main components of IPT-A are education, affect identification, and interpersonal skills building (Jacobson & Mufson, 2010).

IPT-A is considered to be an effective and efficacious treatment for adolescent depression, with numerous research studies having demonstrated it is better than treatment as usual and waitlist conditions (Mufson et al., 2004b; O'Shea, Spence, & Donovan, 2015; Zhou et al., 2015). Furthermore, it has been found to be just as effective as CBT for adolescent depression, with treatment effects maintained in both the short- and long-term (Zhou et al., 2015). Moreover, IPT has been demonstrated to work via the mechanisms outlined in the interpersonal theory of depression. Adolescents undergoing IPT show improvements in social functioning, social skills, peer relationships, and attachment style (Spence, O'Shea, & Donovan, 2016), and improvements in depression symptoms correspond with changes in social skills, parent-child relationships, and attachment style (Spence et al., 2016).

More recently, a number of studies have begun to examine interpersonal-based interventions for pre-adolescents, recognizing (as noted previously) that family relationships are especially important in this age group. Dietz and colleagues (Dietz, Weinberg, Brent, & Mufson, 2015) have developed a family-based version of IPT (FB-IPT) that includes an active parent component, involving parents in weekly sessions and focusing on parent-child conflict. In a recent trial, FB-IPT outperformed child-centered therapy, with youth receiving FB-IPT showing greater decreases in depressive symptoms, lower symptoms at post-treatment, and greater decreases in interpersonal impairment compared to youth receiving child-centered therapy (Dietz et al., 2015). Tompson and colleagues (Tompson, Langer, Hughes, & Asarnow, 2017) also developed a family-focused treatment for childhood depression (FFT-CD) in which a major focus of the treatment is interpersonal factors that contribute to and maintain depressive symptoms. Specifically, families identify problematic patterns of interaction and work to develop skills to enhance family functioning and reduce stress. A recent RCT showed that FFT-CD outperformed individual supportive psychotherapy, with youth receiving FFT-CD showing higher rates of treatment response and greater reported knowledge and skills for handling depression (Tompson et al., 2017). Ultimately, much of the research demonstrates that while we are able to make a short-term impact on depression, long-term effects are less impressive. Further work is needed to support models that can have a more sustained impact.

Psychopharmacological Interventions

In regard to pharmacological approaches, selective serotonin reuptake inhibitors (SSRIs) are considered the first-line treatment for moderate to severe youth depression and have the most evidence to support their effectiveness (Birmaher et al., 2007; Bridge et al., 2007; Cheung et al., 2013; Varigonda et al., 2015). There is also some evidence to support effectiveness of serotonin-norepinephrine reuptake inhibitors in comparison to placebo for child and adolescent depression (Locher et al., 2017). Of the SSRIs, fluoxetine has received the most consistent support for its effectiveness in comparison to placebo in decreasing child and adolescent depression symptoms (Emslie et al., 1997; Hetrick, Merry, McKenzie, Sindahl, & Proctor, 2007).

Escitalopram has also been found to be effective for treating adolescent depression specifically (Emslie, Ventura, Korotzer, & Tourkodimitris, 2009), and in addition to fluoxetine, it is now approved by the Food and Drug Administration for use in adolescents with depression (Cheung et al., 2013). Tricyclic antidepressants, previously used to treat youth depression, are no longer recommended as research studies to indicate limited efficacy compared to placebo (Hazell & Mirzaie, 2013).

Several studies have shown the effectiveness of using a combination of psychotherapy and pharmacotherapy to treat youth depression. Results from the TADS study, a rigorous study with a large sample of depressed youth, showed that the combination of fluoxetine plus CBT demonstrated greater symptom improvement than fluoxetine or CBT alone (March et al., 2004). At 3 months post-treatment, positive response to treatment was seen in 71% of the participants in the combined group compared to only 43.2% of the participants who received CBT alone and 60.6% of the participants who received fluoxetine. However, by 9 months post-treatment, there was little difference in remission rates across treatment modality with fluoxetine alone, CBT alone, and combination treatment showing 55%, 64%, and 60% remission rates, respectively (Kennard et al., 2009; March et al., 2004).

In sum, although psychopharmacological approaches are often used in isolation as a first line of treatment, research suggests that immediate results are best for moderate to severe levels of depression when combined with psychosocial interventions. For milder presentations of depression, psychosocial approaches alone may be sufficient. A number of psychosocial interventions have been proven to be effective for both children and adolescents with depression. CBT and IPT have the most support in their effectiveness, and there have been a number of recent studies in which family-based treatments targeting younger children show promise. However, despite a growing number of effective treatment options, a number of youth do not respond, and the long-term impact of existing interventions are less impressive. More work is needed in order to determine the treatment ingredients, and in what combination, that will have the most lasting impacts for the greatest number of families and youth.

Depression Treatment for Individuals with ID

A sizeable amount of research has been devoted to investigating treatments for depression in the general population. Unfortunately, research on treatment efficacy for individuals with ID has lagged behind. With at least similar rates of depression as the general population (Hamers et al., 2018), but low rates of receiving specialty mental health services for co-morbid conditions (McCarthy & Boyd, 2002), it is important to examine the data that does exist for this population in light of evidence from studies in the general population.

Psychosocial Interventions

Cognitive Behavioral Therapy Similar to intervention research in the general population, CBT has been the most widely investigated psychosocial treatment for individuals with comorbid depression and intellectual disabilities (Osugo & Cooper, 2016; Roberts & Kwan, 2018). The assumption at one point was that individuals with ID would not be able to engage sufficiently with CBT due to cognitive limitations (Taylor, Lindsay, & Willner, 2008). However, research demonstrates that individuals with ID may indeed have the capacity to learn and utilize many of the skills essential for this therapeutic approach (Roberts & Kwan, 2018), and there is a growing body of evidence supporting CBT (with some modification) for adults with comorbid ID and depression, particularly for those with mild, and possibly moderate, ID (James, 2017; Osugo & Cooper, 2016; Roberts & Kwan, 2018; Vereenooghe & Langdon, 2013).

Recent reviews have examined the efficacy of CBT for adults with depression and ID (James, 2017; Osugo & Cooper, 2016). The consensus has been that CBT is effective at reducing depres-

sion symptoms within this population, similar to findings from the general population. In a review of six studies, James (2017) reported that CBT in all studies resulted in reductions in depression symptoms from the mild to minimal range as assessed by the BDI-II. Effect sizes were calculated for each of the six studies, with two falling in the large range, two in the medium range, and two showing no effect (James, 2017). Of these last two, one study compared three active treatments that included a cognitive treatment, a behavioral treatment, and a combined cognitive-behavioral treatment (McGillivray & Kershaw, 2015); while the other compared CBT versus treatment as usual (Hassiotis et al., 2013). While this may give cause for optimism, the author of the review cites methodological issues as justification for cautionary interpretations, such as few RCTs (only two fell into this category), overreliance upon symptom rather than diagnostic assessment, lack of control for psychotropic medication use, and potential bias due to lack of assessor blinding (James, 2017; Osugo & Cooper, 2016). Nonetheless, the conclusion of the review is that CBT can be considered as an option for clinicians, but that individual factors (such as cognitive capacity and ability to identify and examine problematic thought patterns) should receive the greater weight of consideration over the research base, which the author notes is lacking in rigor for this population (James, 2017). This review was consistent with Osugo and Cooper's (2016) conclusions after examining some of the same studies in their review of a wide range of interventions for individuals with ID and comorbid diagnoses/symptoms. Furthermore, Osugo and Cooper (2016) note the importance of recognizing the distinction between a limited amount of evidence as opposed to evidence of ineffectiveness, the latter of which is not currently suggested by the findings.

While CBT as a whole may be supported, undoubtedly many clinicians will wonder about specific components of this treatment that may be most applicable to the population of individuals with ID and comorbid depressive disorders. Specific main components of CBT include identifying and labeling emotions, linking thoughts, feelings, and behaviors, identifying problematic patterns of thinking with the goal of restructuring these, and modifying maladaptive behavioral responses to problematic patterns of thinking. The research to date supports that individuals with ID are able to engage in many of these skills, including emotion identification (Hartley et al., 2015; Joyce, Globe, & Moody, 2006; Sams, Collins, & Reynolds, 2006), differentiating between thoughts, feelings, and behaviors (Hartley et al., 2015; Oathamshaw & Haddock, 2006; Roberts & Kwan, 2018; Sams et al., 2006), and linking emotions with thoughts (Dagnan, Chadwick, & Proudlove, 2000; Oathamshaw & Haddock, 2006) and situations (Dagnan et al., 2000; Joyce et al., 2006; Roberts & Kwan, 2018). Furthermore, individuals with ID have demonstrated an increase in skills such as distinguishing thoughts, feelings, and behaviors after instruction (Vereenooghe, Gega, Reynolds, & Langdon, 2016; Vereenooghe, Reynolds, Gega, & Langdon, 2015), with some data (Taylor et al., 2008) supporting awareness of cognitive mediation (i.e., that how one thinks of situations influences one's reaction). However, much of this research has focused on individuals with mild ID. Some researchers who included those with moderate ID in their samples found that this subgroup had greater difficulty learning CBT, requiring much more modeling, repetition, simplification, and prompting as compared to those with mild ID (Roberts & Kwan, 2018).

The potential difference in CBT efficacy for those with mild versus moderate ID may be partially explained by differences in verbal IQ. Verbal abilities appear to predict success with CBT. Researchers have found that receptive vocabulary and verbal IQ positively correlate with acquisition of CBT skills (Taylor et al., 2008). For verbal IQ, Taylor et al. (2008) suggest that a verbal IQ of 50 or greater seems associated with better prognosis.

In addition to the CBT-specific skills that seem particularly moderated by verbal abilities, Willner and Goodey (2006) emphasize the need to consider other, more general, cognitive deficits that may influence treatment accessibility. More specifically, they argue that it is critical for the

behavioral health treatment provider to distinguish between deficits due to ID and distortions that are errors in thinking rooted in incorrect assumptions and beliefs. When the latter is at play (referred to as a "cognitive distortion model"), CBT techniques are well designed to target these (Taylor et al., 2008). In the instance of the former (a "cognitive deficit model"), adaptations to treatment delivery are warranted (Taylor et al., 2008). Willner and Goodey's (2006) work on the effects of broader cognitive deficits on CBT treatment for anxiety resulted in the identification of several areas for the clinician and researcher to consider in tailoring CBT. More specifically, they noted seven modifications related to the following: (1) pacing and linguistic complexity, (2) level of abstraction, (3) support for episodic memory, (4) temporal sequencing assistance, (5) directive approaches for adaptive thoughts, (6) support for prospective memory, and (7) support for self-direction. To address each of these, the authors list the following adaptations: (1) the simplification of content, slowing of pace, and increased number of sessions, (2) in vivo practice and the elimination of imaginal scenarios, (3) the involvement of caregiver/collateral individual to increase recall, (4) the use of event timelines, (5) therapist provision of alternate realistic thoughts, (6) reminders and guidance on homework from caregiver, and (7) self-instructional scripts to rehearse adaptive thoughts (Willner & Goodey, 2006).

With the emerging evidence for CBT with adults with ID, clinical professionals may wonder about the suitability of this intervention for youth with comorbid depression and ID. Notably, the literature on psychosocial interventions for youth with ID and depression is fairly underdeveloped. Nonetheless, there is some evidence pointing to CBT as a viable option with this demographic as well. CBT for depression is rooted in an assumption that negative cognitions about the self, world, and future maintain depression, which serves as the foundation for the cognitive error/distortion model (Beck, 2005). Some support for this Beckian model of depression in youth with ID and depression was found by Weeland, Nijhof, Otten, Vermaes, and Buitelaar (2017). Specifically, their evidence pointed to a particular vulnerability based on a greater tendency of youth with ID to underestimate coping abilities (i.e., negative beliefs about the self) as compared to typically developing peers, a vulnerability which was predictive of depressive symptoms.

Although such similarities in depressive models and mechanisms may exist, the particular cognitive limitations in this population warrant treatment adaptations to better fit the unique cognitive profiles of youth with ID (Hronis, Roberts, & Kneebone, 2017; Weeland et al., 2017; Weeland, Nijhof, Vermaes, Engels, & Buitelaar, 2015). Hronis and colleagues (2017) provided an overview of suggested adaptations based on particular cognitive deficits identified from their review of neuropsychological literature. Specific modifications were enumerated for the following five domains: (1) attention (e.g., shorter sessions, breaks, and contingency management of behaviors), (2) working memory (e.g., simplified sentence structures and mnemonic devices), (3) learning and memory (e.g., overlearning, enriched learning through role-plays, and incorporation of collateral individuals), (4) executive functions (e.g., consistent session structure and overt schedules), and (5) written, expressive, and receptive language (e.g., visual aids and concepts deconstructed into smaller units). In an RCT for youth in residential treatment who were diagnosed with ID and comorbid depression and anxiety, Weeland and colleagues incorporated adaptations similar to these. Specifically, their treatment incorporated simplified language, reliance upon collateral support, concepts broken down into smaller components, and the use of more concrete steps such as self-instruction over more traditional cognitive restructuring (Weeland et al., 2015).

In sum, for youth with ID and comorbid depression, researchers have provided some initial support for a similar model of depression as that which is found in typically developing peers, as well as theoretical and practical guidance on potentially important modifications in light of the particular cognitive profiles for youth with ID. Nonetheless, it is clear that specific research

on the efficacy of such interventions is lacking. As compared to typically developing peers, youth with ID are at increased risk for experiencing a greater number of adverse life events (Hatton & Emerson, 2004), higher rates and severity of bullying (Tipton-Fisler, Rodriguez, Zeedyk, & Blacher, 2018), greater self-reported underestimation of their likability, and a lower self-reported estimation of their ability to cope, all increasing the risk of and vulnerability for depressive disorders. In light of such risk factors, the need for more research investigating the potential efficacy of CBT and other psychosocial interventions for depression in youth with ID is evident.

Based on the extant literature, CBT seems to be an efficacious treatment for depression in adults with mild ID in particular and therefore should be considered a viable option for depression treatment. Although the studies have some methodological inconsistencies (James, 2017), the consistent reductions in depressive symptoms support the consideration of CBT for this population. The clinician considering CBT for a client with ID and depression would be right to weigh the client's ability to engage in treatment. A careful consideration of both CBT-specific skills and broader cognitive deficits will help guide modifications in the delivery of CBT. Particular areas for consideration include such skills as discriminating between and linking thoughts, feelings, and behaviors and generating adaptive thoughts after weighing evidence. Verbal abilities and IQ also predict success (Taylor et al., 2008), but should not be the sole consideration. Other general considerations are also necessary, such as motivation, sense of efficacy, and caregiver support (Lindsay, Jahoda, & Willner, 2012; Scott et al., 2019; Taylor et al., 2008; Willner & Goodey, 2006). In summation, having an ID diagnosis does not preclude the use of CBT for co-morbid depression, although the severity and nature of ID deficits likely warrant modifications and adaptations. These modifications seem to be important in increasing the efficacy of CBT for adults with ID (James, 2017; Lindsay et al., 2012; Roberts & Kwan, 2018; Taylor et al., 2008; Willner & Goodey, 2006), while additional research is needed to understand the treatment possibilities for youth with ID and depression.

Other Psychosocial Interventions There are a few other psychosocial interventions that have been investigated in this population, such as exercise therapy (Carraro & Gobbi, 2014) and mindfulness-based cognitive therapy (Idusohan-Moizer, Sawicka, Dendle, & Albany, 2015), many of which have some relationship to or overlap with CBT. One that may show promise is behavioral activation. Behavioral activation is a treatment strategy designed to target the activity withdrawal behavioral response to depression. The rationale is that depression leads to diminished enjoyment of and motivation to engage in activities that one used to enjoy. This lessened participation then feeds into the depressive cycle by decreasing opportunities for naturally rewarding experiences and increasing isolation. In behavioral activation, clients are encouraged to engage in activities that confer a sense of achievement, increase socialization opportunities, fulfill obligations, and/or provide a potentially pleasurable experience. At times, behavioral activation is included as a component of CBT for depression but also has been used as a stand-alone treatment. One particular advantage of behavioral activation as a depression treatment for individuals with ID is that there is less reliance on verbal skills (Jahoda et al., 2015).

In a small pilot feasibility study, Jahoda et al. (2015) investigated behavioral activation as a depression intervention for 21 adults with ID. Most participants (n = 16) were in the mild range for ID, with three and two categorized as moderate and severe, respectively. The intervention, based on a behavioral activation treatment (Lejuez, Hopko, & Hopko, 2001), consisted of 10–12 sessions spanning three phases. Phase one focused on assessing pre-intervention activity levels, identifying impediments to activity engagement, and determining severity of withdrawal. The emphasis of phase two included activity scheduling and addressing obstacles to activity engagement. The final phase focused on celebrating success and planning for continued

progress. Participants monitored mood and activity levels throughout and also engaged in goal setting. Adaptations to ensure better treatment fit for this population included the use of graphical representations of activities and the inclusion of a support person, who also learned the steps of behavioral activation and supported the person with ID in accomplishing activities. Jahoda et al. (2015) noted that participants experienced significant improvement in depression symptoms from pre- to post-intervention based on paired sample t-tests resulting in strong effect sizes (r = 0.78), which were maintained at 3-month follow-up (r = 0.86). This study was followed up with a single-blind RCT comparing behavioral activation to a guided self-help intervention in a sample of 161 adults with mild to moderate ID (Jahoda et al., 2017). The self-help intervention focused on psychoeducation for depression, including information on factors relating to low mood, sleep, physical activity, and problem-solving. Both treatments included the involvement of a support person. Ultimately, participants in both treatments experienced significant reductions in depression symptoms, with large effect sizes, but no difference between treatments (Jahoda et al., 2017). This study further demonstrates the potential efficacy of behavioral activation as a stand-alone depression treatment for persons with ID, but suggests that it may be no more effective than a guided self-help. The role of a supportive individual may also be a common and important factor in treatment for this population.

Notably, although IPT has been found to be an effective treatment for adult and adolescent depression in the general population, this intervention has not been investigated within the ID population, despite preliminary support for problems in interpersonal functioning related to depression in this population (Ailey, Miller, Heller, & Smith, 2006). Given the important predictive relationship between social strain and depression (Lunsky & Benson, 2001), an approach focused on enhancing interpersonal skills/coping may be particularly appropriate. A quasi "off-label" usage of IPT may be warranted to the extent that a client's particular presentation of depression seems to be reflective of the interpersonal rationale in which IPT is rooted. Further, similar to CBT, the clinician would need to consider the cognitive capacity of the individual to identify and understand interpersonal relational dynamics and underlying cognitions and emotions that influence these. It may be useful when applying an IPT framework to include sessions with caregivers to further enhance coping and application of learned skills.

Family-based interventions are another understudied area of investigation (Dykens, 2015). As parenting a child with an ID diagnosis may be associated with increased stress, which can influence the parent-child interaction and exacerbate comorbid psychological conditions, such as depression, family-based interventions may be an important adjunct or even primary treatment methodology. Of the studies that exist, few focus on intervention, and the ones that do tend to mainly employ psychoeducation strategies (Dykens, 2015). This highlights a clear gap in the literature to be addressed by researchers.

Psychopharmacological Interventions

Numerous studies report on the psychotropic medication efficacy for individuals with ID and comorbid depression. Based on the research, there seems to be a consensus that SSRIs should be the first line of treatment, when medications are warranted (Matson, Rivet, & Fodstad, 2009; Verhoeven, Veendrik-Meekes, Jacobs, van den Berg, & Tuinier, 2001). Researchers have found that SSRIs seem to be associated with lessened depressive symptoms for persons with ID and comorbid depression symptoms (Janowsky, Shetty, Barnhill, Elamir, & Davis, 2005). Some specific medications receiving support include citalopram (Hamers et al., 2018; Verhoeven et al., 2001), fluoxetine (Sovner, Fox, Lowry, & Lowry, 2008) and paroxetine for adolescents (Masi, Marcheschi, & Pfanner, 1997). However, much of this data is based on case reports, uncontrolled studies, and retrospective chart reviews (Janowsky et al., 2005).

Prescribing psychotropic medications for persons with ID must be carefully monitored, as there is some evidence for increased likelihood of side effects (Hamers et al., 2018). These side effects may be particularly likely based on higher dosages and polypharmacy (Matson et al., 2009). Concerns about potential increased sensitivity to side effects has resulted in a general prescribing rule of starting dosages low and raising them more gradually than in the typical population (Osugo & Cooper, 2016). However, researchers are quick to point out that this "low and slow" approach is based on clinician judgment and has yet to be put to the test of empirical assessment (Osugo & Cooper, 2016). The concern that arises is whether individuals with ID and comorbid psychiatric disorders, including depression, go undertreated.

At earlier stages in the treatment of psychiatric disorders in this population, there was an overreliance on antipsychotic medications, with prescription rates for antipsychotics far outpacing rates of antidepressant prescriptions (Aman, Van Bourgondien, Wolford, & Sarphare, 1995; Robertson et al., 2000). In contrast, these trends may be changing, with antidepressant prescriptions increased according to somewhat more recent studies (Hurley, Folstein, & Lam, 2003; Tsiouris, Kim, Brown, Pettinger, & Cohen, 2013). Even with these developments, there still appears to be a disproportionately high prescription rate of antipsychotic medications without a diagnosis of psychosis as compared to the general population (Tsiouris et al., 2013).

Of the research that exists on psychopharmacological interventions for persons with comorbid-depression and ID, the predominant focus is on adults. Nonetheless, it is important to consider a wider developmental perspective to ensure sufficient attention to treatment implications for individuals at both ends of the developmental trajectory. As for the efficacy of medication use in children and adolescents with ID, there is little research to guide this work, and much must be extrapolated from the general population for whom fluoxetine has received the most consistent support (Cipriani et al., 2018; Hetrick et al., 2007), highlighting a clear need for additional research. Prescribing trends may be replete with the same over-reliance on antipsychotics that characterized adult treatments, particularly in a climate where off-label prescription of antipsychotics has surged among youth with a variety of mental health disorders (Comer, Olfson, & Mojtabai, 2010). Although there is evidence of increased balance for adults with ID and other psychiatric conditions, this may not yet be the case with children and adolescents. For this younger demographic, the trend of particularly high rates of antipsychotic prescriptions may be even more pronounced, especially for youth in residential treatment settings (Scheifes et al., 2013). In a review of psychotropic drug prescriptions, Scheifes et al. (2013) noted that antidepressants were rarely prescribed for the 185 youth with a depression or anxiety diagnosis. In contrast, a sizeable number of youths were prescribed antipsychotic medications, despite few of them having a diagnosis of psychosis (Scheifes et al., 2013). Without more treatment efficacy studies, particularly RCTs focused on youth with ID and depressive disorders, it is harder to mount a convincing case for changing such practices.

For older adults with ID and comorbid depression, there is some support for the use of antidepressants, such as SSRIs (Aman & Singh, 1991). However, several factors need careful consideration when incorporating psychotropic medications in this population. Changing metabolic patterns with age require careful monitoring of dosage and side effects (Eady, Courtenay, & Strydom, 2015; LeBlanc & Matson, 1997). Treatment may be further complicated by long-term chronic health conditions which increase the likelihood of polypharmacy (Eady et al., 2015). Prescribers considering a psychotropic medication are advised to take into account the following considerations from Eady et al. (2015): (1) the extent to which a health-care proxy is needed depending on the capacity of the older adult to make health-care decisions, (2) the capacity to adhere to a medication regimen, (3) the need to notify all caregivers of prescriptions and modifications, and (4) the need to monitor carefully side effects and to instruct collateral supports on how to monitor these as well. While

there is generally limited research on psychopharmacological interventions in this population (Eady et al., 2015), these clinical recommendations can assist the prescriber in utilizing clinical judgment based on data from the general population, limited research on older adults with ID and depression, and individual factors.

Conclusion

Treatment studies over the last several decades have resulted in a significantly improved landscape for treatment of depressive disorders across the lifespan. Across development, psychosocial and pharmacological treatments for depression demonstrate efficacy when compared to no treatment or waitlist control conditions. Important developmental differences and considerations are notable, including greater family involvement for youth, attention to comorbid conditions (i.e., psychiatric and developmental comorbidity common in children and adolescents, and medical comorbidity, common in the elderly). For many depressed individuals – both youth and adults – combination treatment including pharmacological and psychosocial components may be particularly helpful in reducing immediate symptoms and enhancing longer term skills. Given that suicidality is a frequent concomitant of depression, including psychotherapeutic interventions along with pharmacological ones, provides greater opportunity to monitor regularly and address suicidality. This increased occasion for risk monitoring appears as a particular strength of this combined approach. However, there are limitations in the current knowledge base. First, despite evidence that active treatment is better than no treatment or waitlist, there is less evidence supporting differences between active treatments. Second, there is a scarcity of data on moderators of treatment, limiting our understanding of factors that may assist us in determining which treatment would be better for particular individuals. By identifying these moderators, we may be in a better position to match treatments to individuals more effectively. Third, few studies have examined treatment of depression in specific populations, particularly those with neurodevelopmental disorders, such as ID. Existing treatments may need adjustments when working with these populations. Fourth and finally, many of the studies of both adults and youth have short-term follow-up; more studies are needed to identify the impact of treatment longer term. Relatedly, existing studies with longer term follow-up data suggest significant chronicity and relapse of depression. New and efficacious strategies are needed to extend the impact of interventions and provide more long-term models aimed at ameliorating symptoms, reducing relapse and improving overall functioning and life adjustment.

References

Ahrnsbrak, R., Bose, J., Hedden, S. L., Lipari, R. N., & Park-Lee, E. (2017). Key substance use and mental health indicators in the United States: Results from the 2016 National Survey on drug use and health. In *Center for Behavioral Health Statistics and Quality*. Rockville, MD: Substance Abuse and Mental Health Services Administration.

Ailey, S. H., Miller, A. M., Heller, T., & Smith, E. V. (2006). Evaluating an interpersonal model of depression among adults with down syndrome. *Research and Theory for Nursing Practice, 20*(3), 229–246.

Aman, M. G., & Singh, N. N. (1991). Pharmacological intervention. In J. L. Matson & J. A. Mulick (Eds.), *Handbook of mental retardation., 2nd ed.* (pp. 347–372). Retrieved from http://search.ebscohost.com/login.aspx?direct=true&db=psyh&AN=1991-97816-023&site=ehost-live

Aman, M. G., Van Bourgondien, M. E., Wolford, P. L., & Sarphare, G. (1995). Psychotropic and anticonvulsant drugs in subjects with autism: Prevalence and patterns of use. *Journal of the American Academy of Child & Adolescent Psychiatry, 34*(12), 1672–1681. https://doi.org/10.1097/00004583-199512000-00018

Austin, K. L., Hunter, M., Gallagher, E., & Campbell, L. E. (2018). Depression and anxiety symptoms during the transition to early adulthood for people with intellectual disabilities: Depression and anxiety in young adults with ID. *Journal of Intellectual Disability Research, 62*(5), 407–421. https://doi.org/10.1111/jir.12478

Beck, A. T. (2005). The current state of cognitive therapy: A 40-year retrospective. *Archives of General Psychiatry, 62*(9), 953. https://doi.org/10.1001/archpsyc.62.9.953

Birmaher, B., Brent, D. A., Kolko, D., Baugher, M., Bridge, J., Holder, D., … Ulloa, R. E. (2000). Clinical

outcome after short-term psychotherapy for adolescents with major depressive disorder. *Archives of General Psychiatry, 57*(1), 29–36.

Birmaher, B., Brent, D., Bernet, W., Bukstein, O., Walter, H., Benson, R. S., … Medicus, J. (2007). Practice parameter for the assessment and treatment of children and adolescents with depressive disorders. *Journal of the American Academy of Child & Adolescent Psychiatry, 46*(11), 1503–1526. https://doi.org/10.1097/chi.0b013e318145ae1c

Blazer, D. G. (2003). Depression in late life: Review and commentary. *The Journals of Gerontology: Series A: Biological Sciences and Medical Sciences, 58*(3), 249–265. https://doi.org/10.1093/gerona/58.3.M249

Bowlby, J. (1978). Attachment theory and its therapeutic implications. *Adolescent Psychiatry.*

Brent, D. A., Birmaher, B., Kolko, D., Baugher, M., & Bridge, J. (2001). Subsyndromal depression in adolescents after a brief psychotherapy trial: Course and outcome. *Journal of Affective Disorders, 63*(1–3), 51–58.

Bridge, J. A., Iyengar, S., Salary, C. B., Barbe, R. P., Birmaher, B., Pincus, H. A., … Brent, D. A. (2007). Clinical response and risk for reported suicidal ideation and suicide attempts in pediatric antidepressant treatment: A meta-analysis of randomized controlled trials. *JAMA, 297*(15), 1683–1696.

Butler, A. C., Chapman, J. E., Forman, E. M., & Beck, A. T. (2006). The empirical status of cognitive-behavioral therapy: A review of meta-analyses. *Clinical Psychology Review, 26*(1), 17–31. https://doi.org/10.1016/j.cpr.2005.07.003

Calati, R., Salvina Signorelli, M., Balestri, M., Marsano, A., De Ronchi, D., Aguglia, E., & Serretti, A. (2013). Antidepressants in elderly: Metaregression of double-blind, randomized clinical trials. *Journal of Affective Disorders, 147*(1–3), 1–8. https://doi.org/10.1016/j.jad.2012.11.053

Carraro, A., & Gobbi, E. (2014). Exercise intervention to reduce depressive symptoms in adults with intellectual disabilities. *Perceptual and Motor Skills, 119*(1), 1–5. https://doi.org/10.2466/06.15.PMS.119c17z4

Cheung, A. H., Kozloff, N., & Sacks, D. (2013). Pediatric depression: An evidence-based update on treatment interventions. *Current Psychiatry Reports, 15*(8), 381. https://doi.org/10.1007/s11920-013-0381-4

Cipriani, A., Furukawa, T. A., Salanti, G., Chaimani, A., Atkinson, L. Z., Ogawa, Y., … Geddes, J. R. (2018). Comparative efficacy and acceptability of 21 antidepressant drugs for the acute treatment of adults with major depressive disorder: A systematic review and network meta-analysis. *The Lancet, 391*(10128), 1357–1366. https://doi.org/10.1016/S0140-6736(17)32802-7

Clarke, G. N., Hornbrook, M., Lynch, F., Polen, M., Gale, J., O'Connor, E., … Debar, L. (2002). Group cognitive-behavioral treatment for depressed adolescent offspring of depressed parents in a health maintenance organization. *Journal of the American Academy of Child & Adolescent Psychiatry, 41*(3), 305–313. https://doi.org/10.1097/00004583-200203000-00010

Clarke, G. N., Rohde, P., Lewinsohn, P. M., Hops, H., & Seeley, J. R. (1999). Cognitive-behavioral treatment of adolescent depression: Efficacy of acute group treatment and booster sessions. *Journal of the American Academy of Child & Adolescent Psychiatry, 38*(3), 272–279. https://doi.org/10.1097/00004583-199903000-00014

Comer, J. S., Olfson, M., & Mojtabai, R. (2010). National Trends in child and adolescent psychotropic polypharmacy in office-based practice, 1996-2007. *Journal of the American Academy of Child & Adolescent Psychiatry, 49*(10), 1001–1010. https://doi.org/10.1016/j.jaac.2010.07.007

Compton, S. N., March, J. S., Brent, D., Albano, A. M., Weersing, V. R., & Curry, J. (2004). Cognitive-behavioral psychotherapy for anxiety and depressive disorders in children and adolescents: An evidence-based medicine review. *Journal of the American Academy of Child & Adolescent Psychiatry, 43*(8), 930–959. https://doi.org/10.1097/01.chi.0000127589.57468.bf

Craighead, W. E., Miklowitz, D. J., & Craighead, L. W. (2008). *Psychopathology: History, diagnosis, and empirical foundations*. Hoboken, N.J: John Wiley & Sons.

Cuijpers, P., Karyotaki, E., Reijnders, M., & Ebert, D. D. (2019). Was Eysenck right after all? A reassessment of the effects of psychotherapy for adult depression. *Epidemiology and Psychiatric Sciences, 28*(1), 21–30. https://doi.org/10.1017/S2045796018000057

Cuijpers, P., Berking, M., Andersson, G., Quigley, L., Kleiboer, A., & Dobson, K. S. (2013). A meta-analysis of cognitive-Behavioural therapy for adult depression, alone and in comparison with other treatments. *The Canadian Journal of Psychiatry, 58*(7), 376–385. https://doi.org/10.1177/070674371305800702

Cuijpers, P., Geraedts, A. S., van Oppen, P., Andersson, G., Markowitz, J. C., & van Straten, A. (2011). Interpersonal psychotherapy for depression: A meta-analysis. *American Journal of Psychiatry, 168*(6), 581–592. https://doi.org/10.1176/appi.ajp.2010.10101411

Curren, L., Huz, I., McKee, M., Traeger, L., Bedoya, C. A., Chang, T. E., … Trinh, N.-H. (2018). Patient primary language in a culturally focused intervention for Latino Americans with depression. *Annals of Clinical Psychiatry, 30*(2), 84–90.

Dagnan, D., Chadwick, P., & Proudlove, J. (2000). Toward an assessment of suitability of people with mental retardation for cognitive therapy. *Cognitive Therapy and Research, 24*(6), 627–636. https://doi.org/10.1023/A:1005531226519

de Mello, M. F., de Jesus Mari, J., Bacaltchuk, J., Verdeli, H., & Neugebauer, R. (2005). A systematic review of research findings on the efficacy of interpersonal therapy for depressive disorders. *European Archives of Psychiatry and Clinical Neuroscience, 255*(2), 75–82. https://doi.org/10.1007/s00406-004-0542-x

Dietz, L. J., Weinberg, R. J., Brent, D. A., & Mufson, L. (2015). Family-based interpersonal psychotherapy for depressed preadolescents: Examining efficacy

and potential treatment mechanisms. *Journal of the American Academy of Child & Adolescent Psychiatry, 54*(3), 191–199.

Dobson, K. S. (1989). A meta-analysis of the efficacy of cognitive therapy of depression. *Journal of Consulting and Clinical Psychology, 57*, 414–419.

Dobson, K. S., Hollon, S. D., Dimidjian, S., Schmaling, K. B., Kohlenberg, R. J., Gallop, R. J., … Gollan, J. K. (2008). Randomized trial of behavioral activation, cognitive therapy, and antidepressant medication in the prevention of relapse and recurrence in major depression. *Journal of Consulting and Clinical Psychology, 76*, 468–477.

Dykens, E. M. (2015). Family adjustment and interventions in neurodevelopmental disorders. *Current Opinion in Psychiatry*, 1. https://doi.org/10.1097/YCO.0000000000000129

Eady, N., Courtenay, K., & Strydom, A. (2015). Pharmacological Management of Behavioral and Psychiatric Symptoms in older adults with intellectual disability. *Drugs & Aging, 32*(2), 95–102. https://doi.org/10.1007/s40266-014-0236-7

Emslie, G. J., Rush, A. J., Weinberg, W. A., Kowatch, R. A., Hughes, C. W., Carmody, T., & Rintelmann, J. (1997). A double-blind, randomized, placebo-controlled trial of fluoxetine in children and adolescents with depression. *Archives of General Psychiatry, 54*(11), 1031–1037.

Emslie, G. J., Ventura, D., Korotzer, A., & Tourkodimitris, S. (2009). Escitalopram in the treatment of adolescent depression: A randomized placebo-controlled multisite trial. *Journal of the American Academy of Child & Adolescent Psychiatry, 48*(7), 721–729.

Frank, E., & Spanier, C. (1995). Interpersonal psychotherapy for depression: Overview, clinical efficacy, and future directions. *Clinical Psychology: Science and Practice, 2*(4), 349–369. https://doi.org/10.1111/j.1468-2850.1995.tb00048.x

Galambos, N. L., Barker, E. T., & Krahn, H. J. (2006). Depression, self-esteem, and anger in emerging adulthood: Seven-year trajectories. *Developmental Psychology, 42*(2), 350–365. https://doi.org/10.1037/0012-1649.42.2.350

Garber, J., Gallerani, C. M., & Frankel, S. A. (2009). Depression in children. In I. H. Gotlib & C. L. Hammen (Eds.), *Handbook of depression., 2nd ed.* (pp. 405–443). Retrieved from http://search.ebscohost.com/login.aspx?direct=true&db=psyh&AN=2008-18597-018&site=ehost-live

Gloaguen, V., Cottraux, J., Cucherat, M., & Blackburn, I-M. (1998). A meta-analysis of the effects of cognitive therapy in depressed patients. *Journal of Affective Disorders, 49*, 59–72

Greenberg, P. E., Fournier, A.-A., Sisitsky, T., Pike, C. T., & Kessler, R. C. (2015). The economic burden of adults with major depressive disorder in the United States (2005 and 2010). *The Journal of Clinical Psychiatry, 76*(02), 155–162. https://doi.org/10.4088/JCP.14m09298

Haigh, E. A. P., Bogucki, O. E., Sigmon, S. T., & Blazer, D. G. (2018). Depression among older adults: A 20-year update on five common myths and misconceptions. *The American Journal of Geriatric Psychiatry, 26*(1), 107–122. https://doi.org/10.1016/j.jagp.2017.06.011

Hamers, P. C. M., Festen, D. A. M., & Hermans, H. (2018). Non-pharmacological interventions for adults with intellectual disabilities and depression: A systematic review: Non-pharmacological interventions for depression. *Journal of Intellectual Disability Research, 62*(8), 684–700. https://doi.org/10.1111/jir.12502

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 randomised controlled trial. *Journal of Affective Disorders, 151*(1), 186–195. https://doi.org/10.1016/j.jad.2013.05.076

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. https://doi.org/10.1111/j.1360-2322.2004.00188.x

Hazell, P., & Mirzaie, M. (2013). Tricyclic drugs for depression in children and adolescents. *Cochrane Database of Systematic Reviews, 6*.

Hetrick, S. E., Merry, S. N., McKenzie, J., Sindahl, P., & Proctor, M. (2007). Selective serotonin reuptake inhibitors (SSRIs) for depressive disorders in children and adolescents. *Cochrane Database of Systematic Reviews, 3*.

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. https://doi.org/10.1111/bjc.12133

Hurley, A. D., Folstein, M., & Lam, N. (2003). Patients with and without intellectual disability seeking outpatient psychiatric services: Diagnoses and prescribing pattern. *Journal of Intellectual Disability Research, 47*(1), 39–50. https://doi.org/10.1046/j.1365-2788.2003.00463.x

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: Mindfulness for adults with intellectual disabilities. *Journal of Intellectual Disability Research, 59*(2), 93–104. https://doi.org/10.1111/jir.12082

Jacobson, C. M., & Mufson, L. (2010). Treating adolescent depression using interpersonal psychotherapy.

Evidence-Based Psychotherapies for Children and Adolescents, 2, 140–155.

Jahoda, A., Melville, C. A., Pert, C., Cooper, S.-A., Lynn, H., Williams, C., & Davidson, C. (2015). A feasibility study of behavioural activation for depressive symptoms in adults with intellectual disabilities: Behavioural activation for depressive symptoms. *Journal of Intellectual Disability Research, 59*(11), 1010–1021. https://doi.org/10.1111/jir.12175

Jahoda, A., Hastings, R., Hatton, C., Cooper, S.-A., Dagnan, D., Zhang, R., … Melville, C. (2017). Comparison of behavioural activation with guided self-help for treatment of depression in adults with intellectual disabilities: A randomised controlled trial. *The Lancet Psychiatry, 4*(12), 909–919. https://doi.org/10.1016/S2215-0366(17)30426-1

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

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. *The International Journal of Neuropsychopharmacology, 8*(1), 37–48. https://doi.org/10.1017/S146114570400481X

Joyce, T., Globe, A., & Moody, C. (2006). Assessment of the component skills for cognitive therapy in adults with intellectual disability. *Journal of Applied Research in Intellectual Disabilities, 19*(1), 17–23. https://doi.org/10.1111/j.1468-3148.2005.00287.x

Kennard, B. D., Silva, S. G., Tonev, S., Rohde, P., Hughes, J. L., Vitiello, B., … March, J. (2009). Remission and recovery in the treatment for adolescents with depression study (TADS): Acute and long-term outcomes. *Journal of the American Academy of Child & Adolescent Psychiatry, 48*(2), 186–195. https://doi.org/10.1097/CHI.0b013e31819176f9

Klerman, G., Rounsaville, B., Chevron, E., Neu, C., & Weissman, M. (1984). Manual for short-term interpersonal therapy for depression.

Klomek, A. B., Marrocco, F., Kleinman, M., Schonfeld, I. S., & Gould, M. S. (2007). Bullying, depression, and suicidality in adolescents. *Journal of the American Academy of Child & Adolescent Psychiatry, 46*(1), 40–49.

Lachman, M. E. (2004). Development in midlife. *Annual Review of Psychology, 55*(1), 305–331. https://doi.org/10.1146/annurev.psych.55.090902.141521

LeBlanc, L. A., & Matson, J. L. (1997). Aging in the developmentally disabled: Assessment and treatment. *Journal of Clinical Geropsychology, 3*(1), 37–55.

Lejuez, C. W., Hopko, D. R., & Hopko, S. D. (2001). A brief behavioral activation treatment for depression: Treatment manual. *Behavior Modification, 25*(2), 255–286. https://doi.org/10.1177/0145445501252005

Lewinsohn, P. M., Clarke, G. N., Hops, H., & Andrews, J. A. (1990). Cognitive-behavioral treatment for depressed adolescents. *Behavior Therapy, 21*(4), 385–401. https://doi.org/10.1016/S0005-7894(05)80353-3

Lindsay, W. R., Jahoda, A. J., & Willner, P. (2012). Adapting psychological therapies for people with intellectual disabilities II: Treatment approaches and modifications. In J. L. Taylor, W. R. Lindsay, R. P. Hastings, & C. Hatton (Eds.), *Psychological Therapies for Adults with Intellectual Disabilities* (pp. 85–100). https://doi.org/10.1002/9781118329252.ch6

Locher, C., Koechlin, H., Zion, S. R., Werner, C., Pine, D. S., Kirsch, I., … Kossowsky, J. (2017). Efficacy and safety of selective serotonin reuptake inhibitors, serotonin-norepinephrine reuptake inhibitors, and placebo for common psychiatric disorders among children and adolescents: A systematic review and meta-analysis. *JAMA Psychiatry, 74*(10), 1011–1020.

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: JIDR, 45*(Pt 2), 106–114.

Maïano, C., Coutu, S., Tracey, D., Bouchard, S., Lepage, G., Morin, A. J. S., & Moullec, G. (2018). Prevalence of anxiety and depressive disorders among youth with intellectual disabilities: A systematic review and meta-analysis. *Journal of Affective Disorders, 236*, 230–242. https://doi.org/10.1016/j.jad.2018.04.029

March, J., Silva, S., Petrycki, S., Curry, J., Wells, K., Fairbank, J., … Vitiello, B. (2004). Fluoxetine, cognitive-behavioral therapy, and their combination for adolescents with depression: Treatment for adolescents with depression study (TADS) randomized controlled trial. *JAMA, 292*(7), 807–820.

Masi, G., Marcheschi, M., & Pfanner, P. (1997). Paroxetine in depressed adolescents with intellectual disability: An open label study. *Journal of Intellectual Disability Research, 41*(3), 268–272. https://doi.org/10.1111/j.1365-2788.1997.tb00707.x

Matson, J. L., Rivet, T. T., & Fodstad, J. C. (2009). Matson evaluation of drug side-effects (MEDS) profiles of selective serotonin reuptake inhibitors (SSRI) in adults with intellectual disability. *Journal of Developmental and Physical Disabilities, 21*(1), 57–68. https://doi.org/10.1007/s10882-008-9125-5

McCarthy, J., & Boyd, J. (2002). Mental health services and young people with intellectual disability: Is it time to do better? *Journal of Intellectual Disability Research, 46*(3), 250–256. https://doi.org/10.1046/j.1365-2788.2002.00401.x

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?: Analysis of interventions for depression in people with ID. *Journal of Intellectual Disability Research, 59*(2), 105–115. https://doi.org/10.1111/jir.12110

Mufson, L., Dorta, K. P., Wickramaratne, P., Nomura, Y., Olfson, M., & Weissman, M. M. (2004a). A randomized effectiveness trial of interpersonal psychother-

apy for depressed adolescents. *Archives of General Psychiatry, 61*(6), 577–584.

Mufson, L., Dorta, K. P., Moreau, D., & Weissman, M. M. (2004b). *Interpersonal psychotherapy for depressed adolescents*. New York.

Oathamshaw, S. C., & Haddock, G. (2006). Do people with intellectual disabilities and psychosis have the cognitive skills required to undertake cognitive Behavioural therapy? *Journal of Applied Research in Intellectual Disabilities, 19*(1), 35–46. https://doi.org/10.1111/j.1468-3148.2005.00284.x

Office of the Surgeon General (US). (2001). Center for Mental Health Services (US); National Institute of Mental Health (US). Mental Health: culture, race, and ethnicity. *A supplement to mental health: a report of the Surgeon General*. Rockville, MD: Substance Abuse and Mental Health Services Administration (US).

O'Shea, G., Spence, S. H., & Donovan, C. L. (2015). Group versus individual interpersonal psychotherapy for depressed adolescents. *Behavioural and Cognitive Psychotherapy, 43*(1), 1–19.

Osugo, M., & Cooper, S.-A. (2016). Interventions for adults with mild intellectual disabilities and mental ill-health: A systematic review: Mild intellectual disabilities and mental ill-health. *Journal of Intellectual Disability Research, 60*(6), 615–622. https://doi.org/10.1111/jir.12285

Poppelaars, M., Tak, Y. R., Lichtwarck-Aschoff, A., Engels, R. C. M. E., Lobel, A., Merry, S. N., … Granic, I. (2016). A randomized controlled trial comparing two cognitive-behavioral programs for adolescent girls with subclinical depression: A school-based program (Op Volle Kracht) and a computerized program (SPARX). *Behaviour Research and Therapy, 80*, 33–42. https://doi.org/10.1016/j.brat.2016.03.005

Ribeiz, S. R. I., Duran, F., Oliveira, M. C., Bezerra, D., Castro, C. C., Steffens, D. C., … Bottino, C. M. C. (2013). Structural brain changes as biomarkers and outcome predictors in patients with late-life depression: A cross-sectional and prospective study. *PLoS One, 8*(11), e80049. https://doi.org/10.1371/journal.pone.0080049

Roberts, L., & Kwan, S. (2018). Putting the C into CBT: Cognitive challenging with adults with mild to moderate intellectual disabilities and anxiety disorders. *Clinical Psychology & Psychotherapy, 25*(5), 662–671. https://doi.org/10.1002/cpp.2196

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*(6), 666–676. https://doi.org/10.1046/j.1365-2788.2000.00307.x

Rosselló, J., & Bernal, G. (1999). The efficacy of cognitive-behavioral and interpersonal treatments for depression in Puerto Rican adolescents. *Journal of Consulting and Clinical Psychology, 67*(5), 734–745. https://doi.org/10.1037/0022-006X.67.5.734

Rudolph, K. D., Hammen, C., Burge, D., Lindberg, N., Herzberg, D., & Daley, S. E. (2000). Toward an interpersonal life-stress model of depression: The developmental context of stress generation. *Development and Psychopathology, 12*(2), 215–234.

Sams, K., Collins, S., & Reynolds, S. (2006). Cognitive therapy abilities in people with learning disabilities. *Journal of Applied Research in Intellectual Disabilities, 19*(1), 25–33. https://doi.org/10.1111/j.1468-3148.2006.00303.x

Scheifes, A., de Jong, D., Stolker, J. J., Nijman, H. L. I., Egberts, T. C. G., & Heerdink, E. R. (2013). Prevalence and characteristics of psychotropic drug use in institutionalized children and adolescents with mild intellectual disability. *Research in Developmental Disabilities, 34*(10), 3159–3167. https://doi.org/10.1016/j.ridd.2013.06.009

Scott, K., Hatton, C., Knight, R., Singer, K., Knowles, D., Dagnan, D., … Jahoda, A. (2019). Supporting people with intellectual disabilities in psychological therapies for depression: A qualitative analysis of supporters' experiences. *Journal of Applied Research in Intellectual Disabilities, 32*(2), 323–335. https://doi.org/10.1111/jar.12529

Sheeber, L. B., Davis, B., Leve, C., Hops, H., & Tildesley, E. (2007). Adolescents' relationships with their mothers and fathers: Associations with depressive disorder and subdiagnostic symptomatology. *Journal of Abnormal Psychology, 116*(1), 144.

Smedley, B. D., Stith, A. Y., & Nelson, A. R. (2003). *Committee on understanding and eliminating racial and ethnic disparities in health care. Unequal treatment: confronting racial and ethnic disparities in health care*. Washington, DC: National Academies Press.

Smith, P., Scott, R., Eshkevari, E., Jatta, F., Leigh, E., Harris, V., … Yule, W. (2015). Computerised CBT for depressed adolescents: Randomised controlled trial. *Behaviour Research and Therapy, 73*, 104–110. https://doi.org/10.1016/j.brat.2015.07.009

Sovner, R., Fox, C. J., Lowry, M. J., & Lowry, M. A. (2008). Fluoxetine treatment of depression and associated self-injury in two adults with mental retardation. *Journal of Intellectual Disability Research, 37*(3), 301–311. https://doi.org/10.1111/j.1365-2788.1993.tb01287.x

Spence, S. H., O'Shea, G., & Donovan, C. L. (2016). Improvements in interpersonal functioning following interpersonal psychotherapy (IPT) with adolescents and their association with change in depression. *Behavioural and Cognitive Psychotherapy, 44*(3), 257–272. https://doi.org/10.1017/S1352465815000442

Taylor, J. L., Lindsay, W. R., & Willner, P. (2008). CBT for people with intellectual disabilities: Emerging evidence, cognitive ability and IQ effects. *Behavioural and Cognitive Psychotherapy, 36*(06), 723. https://doi.org/10.1017/S1352465808004906

Tipton-Fisler, L. A., Rodriguez, G., Zeedyk, S. M., & Blacher, J. (2018). Stability of bullying and internalizing problems among adolescents with ASD, ID,

or typical development. *Research in Developmental Disabilities, 80*, 131–141. https://doi.org/10.1016/j.ridd.2018.06.004

Tompson, M. C., Langer, D. A., Hughes, J. L., & Asarnow, J. R. (2017). Family-focused treatment for childhood depression: Model and case illustrations. *Cognitive and Behavioral Practice, 24*(3), 269–287.

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. https://doi.org/10.1007/s10803-012-1617-6

Tunvirachaisakul, C., Gould, R. L., Coulson, M. C., Ward, E. V., Reynolds, G., Gathercole, R. L., … Howard, R. J. (2018). Predictors of treatment outcome in depression in later life: A systematic review and meta-analysis. *Journal of Affective Disorders, 227*, 164–182. https://doi.org/10.1016/j.jad.2017.10.008

Varigonda, A. L., Jakubovski, E., Taylor, M. J., Freemantle, N., Coughlin, C., & Bloch, M. H. (2015). Systematic review and meta-analysis: Early treatment responses of selective serotonin reuptake inhibitors in pediatric major depressive disorder. *Journal of the American Academy of Child & Adolescent Psychiatry, 54*(7), 557–564. https://doi.org/10.1016/j.jaac.2015.05.004

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

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. https://doi.org/10.1016/j.ridd.2013.08.030

Vereenooghe, L., Reynolds, S., Gega, L., & Langdon, P. E. (2015). Can a computerised training paradigm assist people with intellectual disabilities to learn cognitive mediation skills? A randomised experiment. *Behaviour Research and Therapy, 71*, 10–19. https://doi.org/10.1016/j.brat.2015.05.007

Verhoeven, W. M. A., Veendrik-Meekes, M. J., Jacobs, G. A. J., van den Berg, Y. W. M. M., & Tuinier, S. (2001). Citalopram in mentally retarded patients with depression: A long-term clinical investigation. *European Psychiatry, 16*(2), 104–108. https://doi.org/10.1016/S0924-9338(01)00546-6

Vittengl, J. R., Clark, L. A., Dunn, T. W., & Jarret, R. B. (2007). Reducing relapse and recurrence in unipolar depression: A comparative meta-analysis of cognitive-behavioral therapy's effects. *Journal of Consulting and Clinical Psychology, 75*, 475–488.

Vostanis, P., Feehan, C., & Grattan, E. (1998). Two-year outcome of children treated for depression. *European Child & Adolescent Psychiatry, 7*(1), 12–18.

Vostanis, P., Feehan, C., Grattan, E., & Bickerton, W.-L. (1996). Treatment for children and adolescents with depression: Lessons from a controlled trial. *Clinical Child Psychology and Psychiatry, 1*(2), 199–212.

Weeland, M. M., Nijhof, K. S., Otten, R., Vermaes, I. P. R., & Buitelaar, J. K. (2017). Beck's cognitive theory and the response style theory of depression in adolescents with and without mild to borderline intellectual disability. *Research in Developmental Disabilities, 69*, 39–48. https://doi.org/10.1016/j.ridd.2017.07.015

Weeland, M. M., Nijhof, K. S., Vermaes, I., Engels, R. C. M. E., & Buitelaar, J. K. (2015). Study protocol: a randomised controlled trial testing the effectiveness of 'Op Volle Kracht' in Dutch residential care. BMC Psychiatry, 15(1). https://doi.org/10.1186/s12888-015-0498-6

Weersing, V. R., Jeffreys, M., Do, M.-C. T., Schwartz, K. T. G., & Bolano, C. (2017). Evidence base update of psychosocial treatments for child and adolescent depression. *Journal of Clinical Child and Adolescent Psychology, 46*(1), 11–43. https://doi.org/10.1080/15374416.2016.1220310

Weissman, M. M., Wickramaratne, P., Nomura, Y., Warner, V., Verdeli, H., Pilowsky, D. J., … Bruder, G. (2005). Families at high and low risk for depression: A 3-generation study. *Archives of General Psychiatry, 62*(1), 29–36. https://doi.org/10.1001/archpsyc.62.1.29

Weisz, J. R., & Kazdin, A. E. (2017). *Evidence-based psychotherapies for children and adolescents* (3rd ed.). New York, NY: Guilford Publications.

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

Zhou, X., Hetrick, S. E., Cuijpers, P., Qin, B., Barth, J., Whittington, C. J., … Xie, P. (2015). Comparative efficacy and acceptability of psychotherapies for depression in children and adolescents: A systematic review and network meta-analysis. *World Psychiatry, 14*(2), 207–222. https://doi.org/10.1002/wps.20217

Zuckerbrot, R. A., Cheung, A., Jensen, P. S., Stein, R. E. K., & Laraque, D. (2018). Guidelines for adolescent depression in primary care (GLAD-PC): Part I practice preparation, identification, assessment, and initial management. *Pediatrics, 141*(3), 1–21.

29 The Treatment of the Dually Diagnosed: Intellectual Disability and Severe Psychopathology

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Introduction

The treatment of severe psychopathology in persons dually diagnosed with intellectual disabilities is clinically complex and challenging. While persons with intellectual disability are at greater risk of serious mental illness, their needs are more complicated and more likely to go unmet (Havercamp & Scott, 2015). Clinical complexities include patient multimorbidity, limited availability of clinicians specializing in the treatment of persons with intellectual disability, and a sparse dual diagnosis research base. Multimorbidity refers to the presence of more than two chronic conditions (Gijssen et al., 2017; McCarron et al., 2013). The Healthy Ageing and Intellectual Disabilities study of over 1000 adults with intellectual disability over the age of 50 found multimorbidity in 80%, with 47% having more than 4 conditions (Hermans & Evenhuis, 2014). Medical comorbidities complicate the diagnosis and treatment of dually diagnosed persons leading some clinicians to avoid these challenging tasks. Surveys of health-care providers have identified basic knowledge of intellectual disability, the assessment and treatment of dually diagnosed persons, and communication with patients and their caregivers as professional development needs (Hemm, Dagnan, & Meyer, 2015). Communication is not the only day-to-day treatment challenge. Persons with intellectual disability also face limited patient autonomy which impacts transportation, lifestyle choices and opportunities for work, socialization and exercise, and most importantly for this discussion, treatment/medical decision-making. The Vanderbilt Kennedy Center for Excellence in Developmental Disabilities' *Health Care Toolkit for Adults with Intellectual and Developmental Disabilities*, which addresses condition-specific health and mental health concerns as well as communication and informed consent, is available at www.IDDtoolkit.org/. This chapter will address these challenges while outlining the treatment interventions for individuals dually diagnosed with intellectual disability and severe psychopathology including schizophrenia spectrum disorders and bipolar disorder. The importance of a multidisciplinary approach combining psychopharmacotherapy and psychosocial interventions will be detailed.

Vignette: Meeting Ashley

Ashley is a 24-year-old female with moderate intellectual disability and newly diagnosed schizophrenia. She was discharged from an

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inpatient psychiatric hospital a week ago and has come to your office for follow-up. Records indicate that she went into the hospital after attacking her roommate. Intake forms completed by the group home staff note that Ashley has lived there for 2 years, working 4 days a week sorting clothes at a thrift store, and enjoying crafts and watching TV with peers until about 8 months ago. She stopped wanting to spend weekends with her parents at that time and had to be encouraged to go to work. She needed reminders to shower. She was increasingly suspicious of others. Before going into the hospital, she had accused her roommate of stealing her favorite coffee cup. Even after the cup was found in the cupboard, she called her roommate a thief. Later that night she became loud, waking her roommate and threatening her. Ashley finally calmed and went to sleep, only to wake agitated; hitting her roommate during breakfast, claiming she had messed with her food.

In your office, Ashley is quiet and reserved with limited facial expression, and reports she is doing "OK. The hospital made me well. Nothing wrong with me now." Lynda, Ashley's caregiver, tells you she is sleeping at night, eating well, and is no longer openly suspicious of others. She has yet to return to her job and does not join in activities. Vital signs are normal, except for an elevated BMI and waist circumference. Her AIMS screen for medication side effects is unremarkable. Routine lab work is ordered. Ashley's parents and staff have questions about Ashley's diagnosis and ask if she will need to continue the medication. You explain the diagnosis of schizophrenia to Ashley and tell her that her family, Lynda, her doctors, and clinicians will help her stay well. You tell Ashley that she has been doing a great job taking her medication every day and she rewards you with a faint smile. Ashley and her family agree to attend family psychoeducation with Lynda joining as well.

Overview of Severe Psychopathology in Dually Diagnosed Persons

The dual diagnosis of severe psychopathology and intellectual disability is a concept rooted in the earliest days of psychiatry. Emil Kraepelin, the father of modern psychiatry, categorized mental illness as manic depression and dementia praecox, the former is now called bipolar disorder and the latter is called schizophrenia (Kraepelin, Barclay, & Robertson, 1919). Kraepelin coined the term *pfropfschizophrenie* to refer to persons with pre-existing intellectual disability who developed schizophrenia (Kraepelin et al., 1919). Compared to the general population, psychopathology in the intellectually disabled population occurs more frequently (Cooper, Smiley, Allan, & Morrison, 2018; Matson & Shoemaker, 2011). Greater than 30% of individuals with intellectual disabilities also have psychiatric disorders, usually since childhood and continuing into adolescence and adulthood (Deb et al., 2009). A systematic review and meta-analysis of 25 studies including over 140,000 individuals with intellectual disability reported a 3.46% prevalence of psychotic disorders in persons with intellectual disability (Aman, Naeem, Farooq, & Ayub, 2016). Historically, medication has been used without behavior or psychiatric symptom specificity and has been utilized to suppress unwanted behaviors; such treatments exposed individuals to deleterious side-effect profiles (Sheehan, Strydom, Morant, Pappa, & Hassiotis, 2017). Tyrer et al. (2008) reported that placebo was superior to Haldol and Risperdal prescribed for aggressive challenging behavior, indicating no evidence of clear efficacy of antipsychotic medication in treating non-symptom-driven behavior. In addition, there have been methodological problems in the pharmacological studies of medication prescribed for behavior problems and psychopathology symptoms in individuals with intellectual disability (Matson,

Bielecki, Mayville, & Matson, 2003). Diagnostic masking and diagnostic overshadowing are common clinical problems in the treatment of persons with intellectual disability. Diagnostic masking occurs when intellectual disability conceals symptoms of illness (Manohar, Subramanian, Kandasamy, Penchilaiya, & Arun, 2016). Diagnostic overshadowing describes the tendency to attribute difficulties to intellectual disability without considering other causes (Geiss et al., 2017). Such diagnostic pitfalls must be avoided to ensure that psychopharmacotherapy is utilized only when indicated for a specific diagnosis. Screening instruments can be helpful in identifying and tracking symptoms over the course of treatment. (See Table 29.1.) Matson et al. (2003) have noted the importance of behavioral assessment and behavioral and psychosocial

Table 29.1 Screening and treatment monitoring instruments

Instrument	Description
Aberrant Behavior Checklist (ABC) (Aman et al., 1985)	Treater or carer-rated, 58-item scale measuring irritability, social withdrawal, stereotypic behavior, hyperactive/noncompliance, and inappropriate speech. Widely used with adults and children with intellectual disability
Brief Negative Symptom Scale (Kirkpatrick et al., 2010)	13-item scale to measure blunted affect, alogia, asociality, anhedonia, and avolition developed for clinical trials and experimental psychopathology studies
Brief Psychiatric Rating Scale (Overall & Gorham, 1962)	Clinician-rated 18 symptom scale based on interview and observations 1 (not present) to 7 (extremely severe). Widely used in schizophrenia spectrum disorder research
Brief Symptom Inventory (BSI) (Wieland, Wardenaar, Fontein, & Zitman, 2012)	Self or interviewer-administered 53-item instrument to identify symptoms and symptom severity. Scales include somatization, obsessive-compulsive, interpersonal sensitivity, depression, anxiety, hostility, phobic anxiety, paranoid ideation, psychoticism. Has been used with persons with borderline and mild ID
Clinical Assessment Interview for Negative Symptoms (CAINS) (Forbes et al., 2010)	23-item, seven-point interview to assess negative symptoms of schizophrenia – asociality, avolition, anhedonia (consummatory and anticipatory), affective flattening, and alogia
Diagnostic Assessment for the Severely Handicapped Scale I, II (DASH I, II) (Bamburg, Cherry, Matson, & Penn, 2001; Matson, Coe, Gardner, & Sovner, 1991)	Carer-rated, 84-item measure with 13 subscales (anxiety, depression, mania, DD/autism, schizophrenia, stereotypies, self-injury, elimination, eating, sleep, sexual, organic, and impulse control) designed to screen for mental illness in persons with severe and profound intellectual disability
Mood, Interest, Pleasure Questionnaire (MIPQ) (Ross & Oliver, 2003)	Carer-rated 25-item instrument with two subscales (mood and interest/pleasure) rated on a five-point Likert scale to measure affect in adults with severe and profound intellectual disability
Positive and Negative Syndrome Scale (PANSS) (Kay, Fiszbein, & Opler, 1987)	Clinical interview informed 30-item scale to identify general psychopathology and positive/negative symptoms of schizophrenia which are rated on a seven-point scale. This scale has been studied in persons with intellectual disability (Hatton et al., 2005)
Psychiatric Assessment Schedule for Adults with a Developmental Disability (PAS-ADD) (Prosser et al., 1998)	A comprehensive, semi-structured clinical interview to identify mental disorders in persons with intellectual disability or the general population. Available as a 25-item checklist for screening and child/adolescent version
Psychotic Symptom Rating Scales (PSYRATS) (Haddock, McCarron, Tarrier, & Faragher, 1999)	Multidimensional 17-item scale used to monitor hallucinations (11 items) and delusions (6 items) using a five-point scale. This scale has been studied in persons with intellectual disability (Hatton et al., 2005)
Scale for the Assessment of Negative Symptoms (SANS) (Andreasen, 1984)	25-item measure of the negative symptoms of schizophrenia with four domains – affective flattening or blunting, alogia, avolition-apathy, and anhedonia-asociality – rated on a six-point scale
Scale for the Assessment of Positive Symptoms (SAPS) (Andreasen, 1984)	34-item measure of the positive symptoms of schizophrenia with four domains – hallucinations, delusions, bizarre behavior, and positive formal thought disorder – rated on a six-point scale

intervention strategies before prescribing medication. Pharmacotherapy is best utilized in the presence of a substantiated mental disorder and the medication's purpose and efficacy profile should coincide with the symptoms (Matson et al., 2000).

Bipolar Disorder in Persons with Intellectual Disability

Bipolar disorder is an illness of mood instability with periods of mania or hypomania with or without depression. Persons with intellectual disability may be vulnerable to mood dysregulation due to sensory issues, trauma, limited coping strategies, and communication challenges. Mood dysregulation can be challenging to differentiate from bipolar disorder. The DSM-5 (2013) details diagnostic criteria for bipolar disorder and the *Diagnostic Manual – Intellectual Disability: A Textbook of Diagnosis of Mental Disorders in Persons with Intellectual Disability* (DM-ID-2) addresses the challenges clinicians encounter in diagnosing bipolar in dually diagnosed persons (Fletcher, Barnhill, & McCarthy, 2016). Treating persons dually diagnosed with intellectual disability and bipolar disorder requires attention to many elements of the mental status over time and can best be tracked using standardized instruments. (See Table 29.1 Screening and Treatment Monitoring Instruments.) The Diagnostic Assessment of the Severely Handicapped (DASH-II) mania subscale elements (agitation, irritability, restlessness, and decreased need for sleep) were correlated with mania in a study of nearly 700 adults in a residential setting (Sturmey, Laud, Cooper, Matson, & Fodstad, 2010). The Aberrant Behavior Checklist (ABC) and Mood Interest Pleasure Questionnaire (MIPQ) are also widely used to monitor mood (Aman, Singh, Stewart, & Field, 1985; Flynn et al., 2017; Ross & Oliver, 2003).

Antonacci and Attiah (2008) have reviewed the epidemiology of mood disorders in persons with intellectual disability noting that mood disorders are more prevalent than psychotic disorders or anxiety disorders. A prospective cohort study of 1023 adults with intellectual disability in Scotland found the 2-year incidence of a manic episode to be 1.1% and bipolar disorder to be 0.8% (Cooper et al., 2018).

Schizophrenia Spectrum Disorders in Persons with Intellectual Disability

The DSM-5 (2013) provides modern diagnostic criteria for schizophrenia spectrum disorders, and the *Diagnostic Manual – Intellectual Disability: A Textbook of Diagnosis of Mental Disorders in Persons with Intellectual Disability* (DM-ID-2) addresses the challenges clinicians encounter in diagnosing schizophrenia in dually diagnosed persons (Fletcher et al., 2016). The DM-ID-2 (2016) explores the presentation of positive symptoms (hallucinations, delusions, and disorganized speech and behavior) and negative symptoms (blunted affect, monotone speech, and avolition including difficulty with activities of daily living) of schizophrenia in persons with intellectual disability. The DM-ID-2 (2016) also notes that communication challenges and limited insight into behavior and/or thoughts are common in persons with intellectual disabilities. The treating clinician must understand an individual's baseline thinking, speech, and emotional regulation to avoid diagnostic overshadowing or masking. For example, a person with intellectual disability and autism may have blunted affect, monotone speech, and thinking with unusual organization at baseline; identifying a change from baseline and a clear presence of hallucinations or delusions would be necessary to diagnose schizophrenia and would have to be monitored carefully during treatment. Self-talk, also called private speech, is a common emotional regulation strategy for adults with intellectual disability and should not be confused with evidence of hallucinations (Dagnan & Rodgers, 2018). The Self-Talk Survey reported that 91% of children and adults with Down syndrome engaged in self-talk (Patti, Andiloro, & Gavin,

2008). Careful attention should be paid to the function of self-talk and the emotional response to the experience to differentiate normal self-talk from hallucinations (Levitas, Hurley, & Pary, 2001). Individuals typically experience self-talk as helpful. When responding to hallucinations, distress, worry, or fear are more likely. Movement disorders and other motor issues are common in persons with intellectual disability, raising the possibility of diagnostic masking or overshadowing (Boot et al., 2015; Hermans & Evenhuis, 2014; Maski, Jeste, & Spence, 2011). Again, treating professionals must understand an individual's baseline functioning and use caution in interpreting behavioral changes.

Persons with intellectual disabilities experience schizophrenic disorders at a higher prevalence, and when ill, suffer greater disability than persons who are not dually diagnosed (Bouras et al., 2004). A systematic review and meta-analysis of 25 studies ($n > 140{,}000$) reported an overall prevalence of schizophrenia of 3.55% in persons with intellectual disability. The prevalence in persons with mild intellectual disability was 5.55%, exceeding that of persons with moderate and severe disability, which were 4.21% and 0.89%, respectively (Aman et al., 2016). A systematic review of five articles describing symptoms of schizophrenia in persons with intellectual disability reported that dually diagnosed persons exhibit more negative symptoms, per rating scales, and may suffer a more severe form of schizophrenia (Welch, Lawrie, Muir, & Johnstone, 2011). Recent genetic studies have demonstrated associations between intellectual disability and schizophrenia (Singh et al., 2017; Soler et al., 2018; Thygesen et al., 2018). While these studies lend support to the neurodevelopmental hypothesis of schizophrenia, the contribution of epigenetics (the impact of lifestyle, the environment, and other factors on genetic expression) is an exciting area under increasing consideration (Bolhuis et al., 2019; Murray, Bhavsar, Tripoli, & Howes, 2017; Murray & Lewis, 1988; Weinberger, 2017).

Catatonia in Persons with Intellectual Disability

The DSM-5 (2013) defines catatonia as "a marked psychomotor disturbance that may involve decreased motor activity, decreased engagement, or excessive and peculiar motor activity" (p 119). Catatonia is classified as *associated with another mental disorder*, *due to another medical condition*, or *unspecified* if the history is limited or full criteria are not met. Catatonia due to a neurodevelopmental disorder would be coded with the disorder followed by the catatonia specifier (American Psychiatric Association & American Psychiatric Association. DSM-5 Task Force., 2013). (See Chap. 18 for a full description of catatonia.) Because symptoms of catatonia including limited speech, oppositionality, stereotypies, grimacing, echolalia, and agitation not influenced by external stimuli, commonly occur in persons with intellectual disability and neurodevelopmental disorders who do not have catatonia, the DM-ID-2 highlights the importance of differentiating baseline behaviors from the diagnostic criteria for catatonia (Fletcher et al., 2016). Video recordings of abnormal movements can be helpful for establishing a baseline and monitoring changes over time.

A meta-analysis of 74 studies found the prevalence of catatonia in general populations to be 9% (Solmi et al., 2017). Persons with neurodevelopmental disorders are at increased risk. A prospective study of catatonia in youth aged 10 to 18 found developmental disability in 31% (Consoli et al., 2012). Reviews have also reported co-occurring catatonia and autism (DeJong, Bunton, & Hare, 2014; Dhossche et al., 2015). There have been case reports of catatonia in Phelan-McDermid syndrome, a rare genetic disorder associated with intellectual disability, autism symptoms, motor delays, and epilepsy (Serret et al., 2015). Catatonia presenting without associated mental health or medical conditions has been reported in persons with Down syndrome (Ghaziuddin, Nassiri, & Miles, 2015; Jap & Ghaziuddin, 2011).

Genetic Disorders in Dually Diagnosed Persons

Persons with intellectual disability due to certain genetic disorders may be at increased risk of serious mental illness. Over 20% of adults with 22q11.2 deletion syndrome develop schizophrenia (Fung et al., 2015). A Swedish national sample of 860 persons with Klinefelter syndrome (KS) reported nearly four times the risk of schizophrenia or bipolar disorder in persons with the syndrome compared to controls (Cederlöf et al., 2014). Niemann-Pick type C, cerebrotendinous xanthomatosis, and Down and Prader-Willi syndromes also increase risk for serious mental illness (Alexander et al., 2016; Bonnot et al., 2016; Fraidakis, 2013; Patterson et al., 2012). Failure to diagnose genetic disorders is common; therefore, treating clinicians should have a high level of suspicion and consider genetic testing when symptoms of serious mental illness fail to respond to typical treatments, particularly if neurological symptoms are present (Fung et al., 2015; Geberhiwot et al., 2018; Groth, Gravholt, Skakkebæk, Høst, & Bojesen, 2013). Persons with genetic disorders may present with symptoms suggesting schizophrenia or bipolar disorder that may be better explained by the genetic disorder. In addition, multimorbidity is high in persons with genetic disorders, requiring special considerations in treatment planning. (See Table 29.2).

The metabolic disorders, Niemann-Pick type C and cerebrotendinous xanthomatosis, are caused by genetic mutations that produce the metabolic error characteristic of each and may require disorder-specific treatments (Bonnot et al., 2014). (See Table 29.2.) Both conditions are associated with dystonia and abnormal movements, complicating medication monitoring as psychotropic medications produce similar movements; however, psychiatric symptoms may improve with the chenodeoxycholic acid required to treat cerebrotendinous xanthomatosis (Fraidakis, 2013). Like cerebrotendinous xanthomatosis, 22q11.2, Down, and Klinefelter syndromes are associated with seizures and cardiac abnormalities and require coordination with the neurologist and cardiologist before treatment with psychotropic medications (Lin et al., 2008). Diabetes and/or thyroid disorders are common in persons with certain genetic syndromes and may necessitate referral to an endocrinologist (Tornese, Pellegrin, Barbi, & Ventura, 2019). In addition, the behaviors associated with a syndrome may also be a symptom of a psychiatric disorder. For example, Klinefelter syndrome has been associated with hypersexuality independent of bipolar disorder and may respond to an SSRI (Fisher et al., 2015; Okolie, Perampalam, Barker, & Nolan, 2017; Sinha, Jnanaprakasan, & Andrade, 2012). The multimorbidity of persons with intellectual disability due to genetic disorders and mental illness requires a multidisciplinary team approach for treatment success.

Pharmacotherapy for Schizophrenia Spectrum Disorders and Bipolar Disorder

When symptoms constituting serious mental illness lead to a psychiatric diagnosis in a person with an intellectual disability, a strategic treatment plan can be initiated. (See Fig. 29.1.) Osugo and Cooper (2016) have reported that there are few evidence-based interventions for people with mild intellectual disabilities and comorbid psychiatric illness, with the existing literature limited in quality and quantity. Many of the available studies do not use standardized measures for tracking efficacy or side effects, relying instead on global measures (Singh et al., 2010). The limited evidence base heightens the importance of careful treatment planning and care coordination. In many countries, the predominant service model for implementing interventions is the person-centered care team model of support and is applicable to community and residential settings. The care team may include direct care staff, psychologists, social workers, general medical providers, and specialists including psychiatrists, neurologists, cardiologists, and others. A member of the care team may make a referral for an assessment for psychotropic medication when challenging behaviors do not have a clear

Table 29.2 Genetic disorders associated with increased incidence of serious mental illness and comorbidities

Genetic disorder	Common comorbid conditions with implications for treatment	Treatment implications
22q11.2 deletion syndrome Autosomal dominant deletion of a portion of chromosome 22. Also called DiGeorge syndrome, velocardiofacial syndrome, conotruncal anomaly face syndrome, Opitz, and Cayler cardiofacial syndrome	Cleft palate Epilepsy Heart defects Hypotonia Thyroid disease	Consider specialist consultation – cardiology, neurology, endocrinology Medication monitoring may be complicated by poor muscle tone and oral issues
Cerebrotendinous xanthomatosis Autosomal recessive mutation if the CYP27A1 gene causing a deficiency in chenodeoxycholic acid leading to numerous metabolic abnormalities	Atypical parkinsonism Dementia Dystonia Epilepsy Heart disease Pain Spasticity	Psychiatric symptoms may respond to treatment with chenodeoxycholic acid (CDCA) Consider specialist consultation – cardiology, neurology Medication monitoring may be complicated by movement disorders
Down syndrome Trisomy 21	Dementia Diabetes Epilepsy Hearing loss Heart disease Hypothyroidism Hypotonia Sleep disorders	Consider specialist consultation – ENT, cardiology, neurology, endocrinology, sleep specialist Medication monitoring may be complicated by poor muscle tone and oral issues Medication monitoring may be complicated by poor muscle tone
Klinefelter syndrome Fetal developmental abnormality leading to extra X chromosome(s).	Diabetes Epilepsy Gynecomastia Heart disease Hypersexuality Metabolic syndrome	Consider specialist consultation – cardiology, neurology, endocrinology Hypersexuality may occur independent of bipolar disorder Gynecomastia and metabolic syndrome may be due to medication as well as the syndrome
Niemann-Pick type C Autosomal recessive mutation of the PC1 or NPC2 gene leading to errors in lipid metabolism	Dystonia Hearing loss Impaired gaze with associated blinking and unusual head movements Sleep disturbances Tremor	Treatment with Miglustat may be necessary to treat the metabolic error with psychotropic medication to address psychiatric symptoms that do not abate Consider specialist consultation – ENT, neurology, sleep specialist Medication monitoring may be complicated by dystonia, impaired gaze, and tremor
Prader-Willi Deletion of a portion of chromosome 15 or presence of an extra copy of chromosome 15	Diabetes Sleep disorders	Consider specialist consultation – endocrinology, sleep specialist

(Alexander et al., 2016; Belling et al., 2017; Bonnot et al., 2014; Bonnot et al., 2016; Fraidakis, 2013; Fung et al., 2015; Geberhiwot et al., 2018; Giannitelli et al., 2018; Kanakis & Nieschlag, 2018; Klünemann, Santosh, & Sedel, 2012; Larson, Whittington, Webb, & Holland, 2014; Patterson et al., 2012)

antecedent. Ideally, results from a functional behavioral analysis will be available. Prescribers review records, interview caregivers, and conduct a comprehensive evaluation including any necessary psychological or medical tests to render a diagnosis. Ample time should be allowed for the evaluation as queries will often need to be repeated, broken down to concrete concepts, and require probes progressing from open-ended to direct questions with choices. Collateral information from caregivers is critical in considering diagnostic options. For each diagnosis, clear

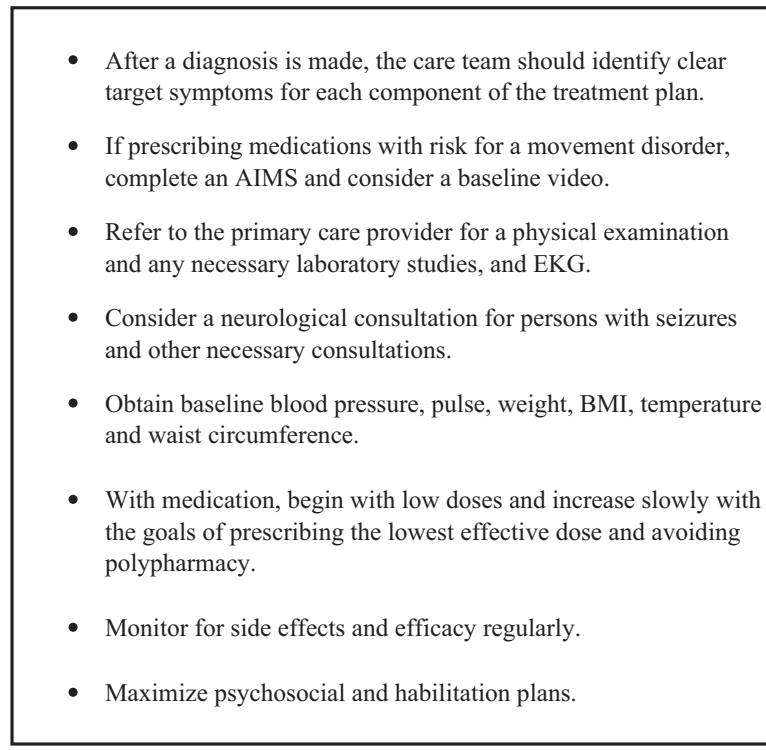

Fig. 29.1 Guideline for the treatment of dually diagnosed persons

target symptoms should be identified, with each addressed separately in the treatment plan. Medication may be prescribed when behavioral or psychosocial approaches require augmentation. The goal of pharmacotherapy is to alleviate the target psychiatric symptom in order to improve the quality of life for the individual and restore baseline activities of daily living. Medication as a sole intervention is rarely appropriate (Chien & Yip, 2013).

Prior to starting medication, a pretreatment workup is in order to exclude physical illness masquerading as mental illness or challenging behavior (Trollor, Salomon, & Franklin, 2016). Seizure disorders and pain related to constipation, GERD, musculoskeletal and other neurological disorders, and dental illnesses are more common in persons with intellectual disability than the general population (Cooper et al., 2015). Clearance from a neurologist may be necessary for persons with a seizure disorder or other neurological conditions. Some syndromes, including Down and 22q11 deletion syndrome, are associated with cardiac abnormalities and may require a cardiology consultation (Ko, 2015). A baseline ECG is warranted due to the concern of cardiac side effects. Due to concerns of potential metabolic derangement with medication, a baseline complete blood count and complete metabolic panel should be obtained and reviewed and any abnormalities addressed with a primary care practitioner. As thyroid disorders are more common in persons with intellectual disability and thyroid disorders can mimic mental illness, thyroid function testing should be obtained as well (Cooper et al., 2015). Furthermore, thyroid function can be directly affected by medication such as lithium and must always be monitored pretreatment and at regular intervals. Many psychotropic medications are known to cause abnormal movements as side effects necessitating an Abnormal Involuntary Movement Scale (AIMS) at baseline and every 6 months. Baseline vital signs are recorded as part of the physical examination and repeated at each appointment including blood pressure, pulse, weight, BMI (body mass index), temperature, and waist circumference (over concern of weight gain and metabolic

syndrome). A thorough understanding of an individual's comorbid medical conditions is critical to personalize the medication regimen to ensure optimal effect (Chien & Yip, 2013). Because of individual treatment responsiveness to medication pharmacokinetics, pharmacogenetic testing may be indicated in some persons (Bousman, Menke, & Müller, 2019).

The goal of person-centered and symptom-specific pharmacotherapy requires individual monitoring of adverse side effects and efficacy at regular intervals. Numerous standardized instruments are available for these purposes. (See Table 29.3.) Over time individuals may be treated with a variety of medications, requiring careful documentation in the medication history. Caregivers are valuable historians in constructing the medication history to allow prescribers to avoid represcribing medications that produced poor efficacy or increased an adverse side effect in the past. The individual, family members, caregivers, and members of a treatment team should be involved in the decision to initiate a pharmacological intervention and offered a full explanation of the potential benefits, risks, and alternatives to the proposed medication. An individual's capacity to give informed consent for medication treatment should be addressed in accordance with the established legal status of the individual in conjunction with family input as appropriate.

While the Food and Drug Administration and the Physician's Desk Reference publish general guidance for medication dosing, prescribers are admonished to "start low and go slow" when dosing persons with intellectual disabilities due to multimorbidity and possible neurophysiological vulnerability (Grier et al., 2018, p. S20). Patients

Table 29.3 Instruments used in medication monitoring

Instrument	Description
Abnormal Involuntary Movement Scale (AIMS) (Guy, 1976)	Clinician-rated 12-item scale designed to assess for tardive dyskinesia and track changes over time
Antipsychotic Non-Neurological Side Effects Rating Scale (ANNSERS) (Yusufi et al., 2005)	Patient or carer-rated 44-item screen for antipsychotic side effects that are *not* due to EPS (parkinsonism, akathisia, dystonia, tardive dyskinesia)
Antipsychotic Side-effect Checklist (ASC) (Weiden & Miller, 2001)	A clinician led 17-item interview used to screen for the presence of common antipsychotic side effects and, if present, subjective distress
Barnes Akathisia Rating Scale (BARS) (Barnes, 1989)	Clinician-rated restlessness is rated subjectively and objectively on a 0–3 scale with akathisia severity rated 0–5
Dyskinesia Identification System: Condensed User Scale (DISCUS) (Sprague, Kalachnik, & Slaw, 1989)	Clinician-rated 15-item screen for tardive dyskinesia
Liverpool University Neuroleptic Side Effect Rating Scale (LUNSERS) (Jung et al., 2005)	Patient or carer-rated 41-item screen for side effects of antipsychotic medications with each rated on a five-point scale
Matson Evaluation of Drug Side Effects Scale (MEDS) (Matson et al., 1998)	Carer-rated 93-item scale with 9 domains developed to assess for the side effects of psychotropic medications in persons with ID
Simpson-Angus Extrapyramidal Side Effects Scale (Simpson & Angus, 1970)	Clinician observation and physical evaluation is rated 10-item scale with items rated 0–4 to screen for drug induced parkinsonism
Systematic Monitoring of Adverse Events Related to Treatments (SMARTS) (Haddad et al., 2014)	A 12-item checklist designed to be completed by patients/caregiver before medication monitoring appointments to screen for side effects of antipsychotic medications
Udvalg for Kliniske Undersøgelser Side Effect Rating Scale (UKU-SERS) (Lingjaerde, Ahlfors, Bech, Dencker, & Elgen, 1987)	A 48-item semi-structured interview assessing side effects of psychotropic medications. The UKU-SERS-ID is a modified version adapted for persons with ID (Louise Tveter, Lise Bakken, Bramness, & Ivar Røssberg, 2014)

and caregivers should be instructed that medications require tapering and should not be stopped abruptly. Monotherapy is preferred, but if polypharmacy is necessary, medications should be started sequentially to allow side-effect and efficacy monitoring (Deb et al., 2009). As medication is added, psychosocial and habilitation plans should be reevaluated and optimized with the goal of minimizing medication use while maximizing psychological interventions.

There is limited professional guidance for the pharmacological treatment of severe psychopathology in persons with intellectual disability. The American Psychiatric Association (APA)*Practice Guideline for the Treatment of Persons with Schizophrenia* notes the increased risk of extrapyramidal side effects in persons with intellectual disability but is otherwise silent on the treatment of persons with intellectual disability (Lehman et al., 2004) as of the most recent update (Dixon, Perkins, & Calmes, 2010). The American Academy of Child and Adolescent Psychiatry (AACAP)*Practice Parameter for the Assessment and Treatment* of *Children and Adolescents with Schizophrenia* does address the antipsychotic treatment of youth with autism and pervasive developmental disability (McClellan & Stock, 2013). A Practice Parameter on *Intellectual Disability* and Clinical Practice Guideline on *Antipsychotic Medication* are currently under development by the AACAP (AACAP, 2018). The APA does not have a current guideline for bipolar disorder, and the AACAP practice parameter was last updated over a decade ago. The United Kingdom National Institute for Health and Care for Excellence (NICE) and the British Association for Psychopharmacology (BAP) have published guidelines for the treatment of schizophrenia and bipolar disorder with attention to medication side effects (Barnes & Schizophrenia Consensus Group of the British Association for, 2011; Excellence, 2014; Goodwin et al., 2016; National Collaborating Centre for Mental Health, 2018) The BAP guidelines do not address the needs of persons with ID. NICE guidelines address antipsychotic medication monitoring in persons with intellectual disability calling for baseline weight, waist circumference, pulse, blood pressure, fasting blood glucose, glycosylated hemoglobin (HbA1c), lipid profile, prolactin level, documentation of movement disorders, and assessment of nutritional status, diet, and level of physical activity with follow-up monitoring of vital signs and for extrapyramidal side effects (EPS) and/or metabolic syndrome. The NICE guideline specifies that an EKG should be considered on an individual basis (Excellence, 2014; National Institute for Clinical). *The Frith Prescribing Guidelines for People with Intellectual Disability* addresses the treatment of schizophrenia and bipolar disorder as well as general prescribing practices for children and adults with ID (Bhaumik, Gangadharan, Branford, & Barrett, 2015). In 2009, the World Psychiatric Association Section on Psychiatry of Intellectual Disability published a brief psychotropic medication prescribing guide for adults with intellectual disabilities (Deb et al., 2009). The University of Birmingham has developed an *LD Medication Guideline* available at https://www.birmingham.ac.uk/research/activity/ld-medication-guide/index.aspx.

Persons with intellectual disability are typically excluded from drug development trials and subsequent treatment efficacy studies, limiting the available information for the pharmacological management of dually diagnosed individuals. The reader is referred to de Leon, Greenlee, Barber, Sabaawi, and Singh (2009) for a review of the available research regarding the second-generation antipsychotic medications (aripiprazole, olanzapine, paliperidone, quetiapine, risperidone, and ziprasidone) and comprehensive considerations for their use. Antonacci and Attiah (2008) have reviewed the treatment of mood disorders in dually diagnosed persons noting that lithium carbonate and anticonvulsant mood stabilizers are recommended for use in the treatment of bipolar disorder.

Palumbo and McDougle (2018) analyzed the use of psychotropic medications for mental illness in persons with Down syndrome citing the typical antipsychotics, divalproex sodium, carbamazepine, and lithium as effective for treating symptoms of bipolar disorder. They noted that carbamazepine is not associated with weight gain

and can be used to treat bipolar disorder and significant mood lability. The treatment of bipolar disorder requires attention to the stage of illness, as mania, depression, and long-term maintenance may require distinct interventions (Baldessarini, Tondo, & Vázquez, 2019). Antidepressants are less effective in treating depression associated with bipolar disorder compared to major depressive episodes and must be used with caution as they may precipitate mania in persons with bipolar disorder (Baldessarini et al., 2019).

When symptoms of severe psychopathology do not respond to initial pharmacological interventions, a different medication in the same class may be initiated; however, polypharmacy, the prescribing multiple medications in the same class, should be avoided when possible (Masnoon, Shakib, Kalisch-Ellett, & Caughey, 2017). Residential living, psychiatric disorders, neurological illness, and gastrointestinal disorders are associated with polypharmacy in persons with intellectual disability (O'Dwyer, Peklar, McCallion, McCarron, & Henman, 2016). Care should be taken taper down a medication as the new medication is started to avoid side effects and a prescribing cascade. Prescribing cascade refers to the potentially dangerous practice that occurs when a medication adverse effect is attributed to a new condition and an additional medication is prescribed (Rochon & Gurwitz, 2017).

When two or more pharmacological interventions fail to ameliorate symptoms, clozapine, electroconvulsive therapy, or novel interventions such as transcranial magnetic stimulation may be considered (Noel, 2018). A review of 13 articles reporting on the use of clozapine by persons with intellectual disability found the data on the benefits for behavior or cognition to be inconclusive (Singh et al., 2010). Subsequently, a systematic review found no randomized controlled trials examining the effects of clozapine treating adults (Ayub, Saeed, Munshi, & Naeem, 2015). A Danish mirror image study of 405 persons with intellectual disability treated with clozapine reported a significant decrease in hospital admissions and total inpatient days with greater benefits for persons with moderate/severe intellectual disability compared to mild intellectual disability (Rohde, Hilker, Siskind, & Nielsen, 2018). Research on electroconvulsive therapy and transcranial magnetic stimulation is limited to case studies. If these options are pursued, the prescriber will need to carefully inform patients, caregivers, and the treatment team of the risks before proceeding. Capacity to consent to treatment should be considered and handled in accordance with state law. Consultation with a professional colleague should be sought when multiple treatments are unsuccessful or interventions with a scant evidence are considered.

The Treatment of Catatonia

When a change in mental status, motor behavior, or function is observed in a person with intellectual disability, careful assessment for symptoms of catatonia is indicated. Because catatonia can be life-threatening, when a clinician suspects catatonia, a prompt medical evaluation is indicated. A thorough description of baseline characteristics and abilities and a detailed account of the presentation and progression of catatonic symptoms are critical for the assessment and treatment of catatonia. All details should accompany the referral to medical care. The referral should also include a list of all medications or substances the individual is using. Medications, including lithium, antipsychotics, disulfiram, azithromycin, and GHB, have been associated with catatonic presentations, as have the medication-induced serotonin syndrome and neuroleptic malignant syndrome (Oldham, 2018). If possible, medications that may cause or worsen catatonia should be stopped. The presenting symptoms and those targeted for treatment must be accurately identified to monitor treatment response. Rating scales should be used to track symptoms over time and response to treatment (Sienaert, Rooseleer, & De Fruyt, 2011). The Bush-Francis Catatonia Rating Scale (BFCRS) is the most commonly used scale (Bush, Fink, Petrides, Dowling, & Francis, 1996). The prompt diagnosis and treatment of catatonia is necessary to prevent medical complications including dehydration, malnutrition, infection, organ failure, and malignant catatonia,

which may be fatal (Funayama, Takata, Koreki, Ogino, & Mimura, 2018; Park et al., 2017). In the medical setting, treatment begins with a thorough physical examination to identify possible medical conditions presenting as catatonia including infections, neurological conditions, and metabolic disorders. Treatment of underlying conditions contributing to the clinical presentation is critical. The need for laboratory studies and neuroimaging is guided by the clinical presentation, as there are not lab tests or scans that definitively diagnose catatonia. Stabilization of acute medical needs is a priority and may require treatment in the intensive care unit (Clinebell, Azzam, Gopalan, & Haskett, 2014; Oldham, 2018).

When catatonia is suspected, a lorazepam challenge test is performed. After carefully documenting symptoms, 1-2 mg of lorazepam is administered intravenously, and symptoms are reassessed after 5 minutes. If there is no symptom change in 30 minutes lorazepam administration may be repeated up to a total dose of 4–6 mg. In lieu of IV lorazepam, intramuscular or oral lorazepam may be administered, but improvement will be delayed (Fink & Taylor, 2006). A zolpidem challenge test has also been reported; however, the lorazepam challenge test is preferred (Thomas, Rascle, Mastain, Maron, & Vaiva, 1997). Improvement after a challenge test confirms the diagnosis of catatonia and treatment may begin.

Protocols and algorithms based on clinical experience guide the treatment of catatonia (Beach, Gomez-Bernal, Huffman, & Fricchione, 2017). A 2018 systematic review found insufficient data to propose an evidence-based treatment protocol for catatonia (Pelzer, van der Heijden, & den Boer, 2018). Medical stabilization continues to be the first priority during the initiation of treatment. Benzodiazepines are the most common treatment, with a combination of lorazepam and diazepam most commonly prescribed. Success has been reported with a wide range of benzodiazepines and other medications including NMDA-receptor antagonists and seizure medications (Beach et al., 2017). In persons with catatonia and Phelan-McDermid syndrome lithium resolved catatonia after typical interventions were ineffective (Serret et al., 2015). In persons with Down syndrome, catatonia responded to typical interventions (Ghaziuddin et al., 2015). Transcranial magnetic stimulation and transcranial direct current stimulation have been used to treat catatonia (da Silva, Fregni, & Brunoni, 2013; Trojak, Meille, Bonin, & Chauvet-Geliner, 2014). If initial treatment is not effective or contraindicated, electroconvulsive therapy (ECT) may be considered (Beach et al., 2017; Sienaert, Dhossche, Vancampfort, De Hert, & Gazdag, 2014). When medication does not resolve catatonic symptoms and the patient becomes unstable with a rising fever, racing heart, and rising blood pressure, malignant catatonia is a concern. Malignant catatonia is a life-threatening condition for which ECT should be considered (Beach et al., 2017) The use of ECT in persons with intellectual disability may raise concerns regarding consent to treatment and may require a court order (Zisselman & Jaffe, 2010). The medical treatment of catatonia carries a good prognosis, with most cases resolving; however, recurrence is possible (Pelzer et al., 2018).

Adverse Effects of Psychotropic Medications

Psychotropic medications are prescribed with the goal of maximizing potential benefits while minimizing potential risks as all medications may have adverse effects. This section will offer a high-level review noting some of the most serious risks. An individual's risk experiencing adverse effects is determined by many factors including age, gender, ethnicity, genetics, health status, use of alcohol or cigarettes, and medication-related factors. Persons with ID are at greater risk for these adverse effects due to multimorbidity and greater medication load due to high dosing, polypharmacy, and longer treatment (Ji & Findling, 2016; Matson & Mahan, 2010; O'Dwyer et al., 2016). Clinicians should provide ongoing education to patients and caregivers regarding adverse effects and conduct regular monitoring for these potentially serious events. Professional organizations offer guid-

ance for medication monitoring. Additional research and consensus is needed regarding adverse effects of psychotropic medications for persons with ID.

Adverse Effects of Antipsychotic Medications

Antipsychotic medications have a range of adverse effects and while some are common and less serious others may have grave consequences. Less serious side effects include drowsiness, dry mouth, nausea, and constipation. Guidance on timing of medication, avoiding driving or operating machinery when tired, and attention to diet and oral hygiene are usually sufficient to address these concerns. Some side effects that are easily managed in the general population may be more serious for persons with ID. For example, dizziness and urinary retention may occur. Rising from a sitting or lying position quickly may cause dizziness due to orthostatic blood pressure changes and can be addressed with the admonition to rise slowly. Persons with intellectual disability may require assistance to implement these directions and may be at greater risk for falls. In 2017, the US Food and Drug Administration added a fall warning to antipsychotic medication labels, noting the importance of education and repeated fall risk assessment (Stocks et al., 2017). For further reading on fall risk management, see the review by Phelan, Mahoney, Voit, and Stevens (2015). Persons with intellectual disability may not recognize a decreased frequency of urination and may not communicate the associated discomfort effectively.

Potentially serious adverse effects include a decreased seizure threshold, extrapyramidal symptoms (EPS), metabolic syndrome, cardiac abnormalities, eye changes, and neuroleptic malignant syndrome. While aripiprazole appears least likely to induce antipsychotic-related seizures, increased risk is noted with clozapine, chlorprothixene, thioridazine, and haloperidol (Wu, Wang, Yeh, & Liu, 2016). EPS includes tardive dyskinesia, dystonia, akathisia, parkinsonism, and bradykinesia. (See Table 29.4.) More severe intellectual disability, brain damage, and female sex are static risk factors for tardive dyskinesia; dynamic factors include higher doses of antipsychotic medication, greater duration of treatment, diabetes, smoking, and substance use (Matson, Fodstad, Neal, Dempsey, & Rivet, 2010; Solmi, Pigato, Kane, & Correll, 2018). A large cohort study found the rate of EPS in per-

Table 29.4 Serious adverse effects of antipsychotic medications defined

Adverse effect		What to look for
Extrapyramidal symptoms	Tardive dyskinesia	Involuntary, repetitive, sometime irreversible movements that often begin around the mouth and may progress to involve the truck or limbs
	Withdrawal dyskinesia	Involuntary, repetitive movements that emerge when a medication is decreased or stopped. Typically improves in 2–3 months
	Dystonia	A sustained involuntary muscle spasm that may affect any muscle group. Dystonia may be acute or chronic
	Akathisia	An uncomfortable restlessness that may result in rocking or pacing. In rare cases it may be associated with aggression
	Parkinsonism	Akinesia, the decreased voluntary control of muscles, and bradykinesia which result in reversible Parkinson-like symptoms including a masked facial expression, pill rolling tremor, and shuffling gait
	Bradykinesia	Abnormal slowness. There may be difficulty initiating and/or stopping actions
Metabolic syndrome		Three or more of the following - central obesity, elevated blood sugar, high blood pressure, high cholesterol level, and high triglyceride level- increase risk for heart disease, stroke, and diabetes
Neuroleptic malignant syndrome		Fever, muscle rigidity, mental status changes, sweating, and unstable blood pressure that characterize this life-threatening medical emergency

sons with intellectual disability 30% higher than persons without ID, with the incidence of neuroleptic malignant syndrome three times more common in persons with ID (Sheehan et al., 2017). The Healthy Aging in Intellectual Disability Study, including 866 individuals receiving antipsychotic medication, found metabolic syndrome in 44.7%, with 94% for whom this condition was previously undiagnosed (de Winter, Bastiaanse, Hilgenkamp, Evenhuis, & Echteld, 2012). A systematic review of the prevalence of the adverse effects of antipsychotic medications among adults in real-world clinical practice including 53 studies reported rates of metabolic syndrome in 23–50%, dyslipidemia in 15–53%, hypertension in 16–49%, and QTc prolongation/arrhythmia in 3–12% of patients (Young, Taylor, & Lawrie, 2014). Adverse effects including sedation, urinary problems, and difficulty swallowing were associated with a lower health-related quality of life in persons with ID which improved when medication was discontinued (Ramerman, Hoekstra, & de Kuijper, 2018a, 2018b). Antipsychotic medication may be associated with ocular side effects including retinopathy, increased risk of cataracts, and worsening of glaucoma, requiring biannual monitoring until age 40, when annual ophthalmological visits are indicated for persons receiving first-generation antipsychotic medications (Anthony, 2018). A systematic review including 19 studies reported that antipsychotic medications are associated with initial cognitive improvement; however, higher cumulative lifetime antipsychotic dose was related to poorer cognitive performance after 16.5 years of treatment (Husa et al., 2017; Karson, Duffy, Eramo, Nylander, & Offord, 2016).

Adverse Side Effects of Mood Stabilizers

Mood stabilizers, including lithium and select anticonvulsants, are associated with a number of rare, but serious adverse effects. In general, more serious effects are associated with higher dosages of medication resulting in the admonition to *start low and go slow* when prescribing mood stabilizers. In addition, many of the mood stabilizers are known to cause birth defects and should not be administered to woman who are pregnant or planning a pregnancy. Birth control should be discussed with women taking mood stabilizers. Lithium, valproic acid, and carbamazepine have a narrow range therapeutic index requiring regular blood work to monitor levels. Emotional and behavioral reactions to blood draws are more common in persons with ID and may require desensitization (Davit, Hundley, Bacic, & Hanson, 2011). Even at therapeutic levels, lithium is known to lower the seizure threshold and may cause diabetes insipidus, a rare condition characterized by polydipsia (excessive and compulsive drinking). Lithium requires monitoring for adverse effects on thyroid, electrolytes, and kidney function. As persons with autism and Down syndrome are at increased risk for hypothyroid conditions, careful monitoring is necessary (Alabaf et al., 2019; Pierce, LaFranchi, & Pinter, 2017). Persons with ID are also at increased risk for dehydration (Brameld, Spilsbury, Rosenwax, Leonard, & Semmens, 2018). Poor hydration may cause dangerous elevation of lithium in the blood which may be fatal if not detected and corrected. Symptoms of lithium toxicity include stomach pain, nausea, vomiting, weakness, tremors, and lethargy and in severe toxicity it can cause seizures, kidney failure, hypotension, and stupor. It is well established that lithium has wide-ranging cardiac effects (Mehta & Vannozzi, 2017) and that adults with ID experience a greater incidence of cardiovascular disease, so care must be taken to minimize the compounding effect of medications (Emerson, Hatton, Baines, & Robertson, 2016). Many neurodevelopmental disabilities including Down syndrome, Williams syndrome, fetal alcohol spectrum disorder, and certain microdeletion and duplication syndromes are associated with heart disease necessitating care when prescribing lithium. While serious adverse effects must be recognized and prevented when possible, common adverse effects of lithium such as gastrointestinal distress and tremor may reduce quality of life and progress to impair health or behavior.

Anticonvulsants including valproic acid, carbamazepine, oxycarbamazepine, and lamotrigine are prescribed for mood stabilization. Valproic acid and carbamazepine require regular blood work. Though rare, valproic acid has been associated with pancreatitis and hepatitis with increased risk associated with polypharmacy (Dols et al., 2013). Pancreatitis with associated abdominal pain may occur with therapeutic blood levels. Jaundice, confusion, or fever of unknown origin necessitates additional evaluation for hepatitis. Early identification of these adverse effects is critical to avoid complications which can lead to death. Hepatitis and a dangerous drop in the white blood cell count (agranulocytosis) may be caused by carbamazepine. A decrease in white blood cells is associated with frequent infections and slow-healing cuts and bruises. Lamotrigine and carbamazepine are associated with increased risk for Stevens-Johnson syndrome, a rare medical emergency. Stevens-Johnson syndrome typically begins with a fever and skin sensitivity which progresses to a painful red or purplish rash. Blisters appear on the skin or mucous membranes (mouth, nose, eyes, or genitals) leading to peeling skin with high risk of sepsis. The FDA recommends that persons of Asian descent be tested for the HLA-B∗1502 gene before treatment with carbamazepine due to the increased risk of Stevens-Johnson syndrome (Tangamornsuksan, Chaiyakunapruk, Somkrua, Lohitnavy, & Tassaneeyakul, 2013). Investigations of the impact of mood stabilizers on cognitive function are not conclusive (Dols et al., 2013). The care team must be well versed in the potential adverse effects to promote treatment compliance and quality of life.

Medication Monitoring Instruments

Regular monitoring for adverse effects is recommended by professional guidelines but less likely to occur in persons with ID. The 2015 Prescribing Observatory for Mental Health (POMH-UK) multicenter audit of psychotropic medication use among persons with intellectual disability in the United Kingdom found that 64%, 3618 individuals, were prescribed antipsychotic medication, approximately half for schizophrenia or an affective disorder (Paton, Bhatti, Purandare, Roy, & Barnes, 2016). The POMH-UK study (2016) also reviewed medication monitoring practices noting that less than half had documented EPS monitoring with even fewer screened for metabolic syndrome. Instruments commonly used to monitor side effects are noted in Table 29.3. The psychometric properties of side-effect assessment tools are reviewed in detail by van Strien, Keijsers, Derijks, and van Marum (2015) and Stomski, Morrison, and Meyer (2015). In addition to monitoring for abnormal movements with assessment tools, video recording is valuable for capturing a baseline and monitoring change over time.

Vignette: Ashley's 1-Month Follow-Up

Ashley's lab is normal, but vitals reveal she has gained 25 pounds. On weekends at her parent's home, she has eaten "constantly" and because she is not working, she is eating throughout the day at the group home. You notice that Ashley's clothes have stains and her hair is unkempt. Lynda acknowledges that Ashley needs reminders to shower and will put on dirty clothes after showering. In fact, Ashley needs prompts and reminders throughout the day. After reviewing medication side-effect profiles, Zyprexa is switched to Abilify to lessen the likelihood of weight gain. You explain the importance of exercising and proper diet to Ashley. Lynda reminds Ashley how she used to enjoy the dancercise program and Ashley agrees to try it again. You explain that neurocognitive decline is common with schizophrenia and Ashley agrees to meet with you for cognitive remediation therapy.

Psychosocial Treatments

The importance of psychosocial interventions for persons with serious mental illness and for persons with intellectual disability is widely recognized by leading mental health organizations (Gaebel, Riesbeck, & Wobrock, 2011). The APA (2010), the National Institute for Health and Care

Excellence (NICE) guidelines (2014, 2016), and the World Health Organization (2005) have issued recommendations for psychosocial interventions including family psychoeducation, assertive community treatment, cognitive behavioral therapy for psychosis, and cognitive remediation. Social skills and supportive employment are additional common psychosocial interventions for serious mental illness but will not be explored here as they are commonly part of habilitation programs. Psychosocial interventions seek to improve treatment compliance and outcomes while promoting patient autonomy. A meta-analysis of 48 randomized trials comparing psychosocial interventions for psychosis reported CBT-p was most beneficial for positive symptoms and social skills training more effective in reducing negative symptoms (Turner, van der Gaag, Karyotaki, & Cuijpers, 2014). Psychosocial interventions for serious mental illness including schizophrenia spectrum disorders and bipolar disorder will be introduced in this section along with information regarding the use of these interventions in dually diagnosed individuals. While research regarding the treatment of dually diagnosed persons is limited, failure to identify persons with intellectual disability as a variable in studies of seriously mentally ill populations is likely (Burge, 2009). For dually diagnosed persons with severe or profound intellectual disability, the evidence base for effective interventions is extremely limited (Vereenooghe et al., 2018). The concepts of dual diagnosis and multimorbidity should be explored in future studies of psychosocial interventions for persons with serious mental illness.

Psychoeducation and Family Psychoeducation (FPE)

Family psychoeducation (FPE) is recommended by professional guidelines including the APA and the NICE in the United Kingdom (Kuipers, Yesufu-Udechuku, Taylor, & Kendall, 2014; Lehman et al., 2004). FPE is an evidence-based intervention for persons with serious mental illness and their families that has been shown to improve quality of life and reduce relapse rates (Dixon et al., 2001; Lehman, Steinwachs,, & Co-Investigators of the, 1998). Elements of FPE include information about illness etiology, symptoms, and prognosis; introduction to goal setting and problem-solving skills; strengthening of family communication and supports; and development of crisis intervention and relapse prevention strategies. The material is delivered to multifamily groups or with individual families over at least 12 sessions (Harvey, 2018). Models of FPE address the needs of children and adults with schizophrenia and bipolar disorder and include behavioral family therapy, which focuses on strengthening specific skill deficits (Coker, Williams, Hayes, Hamann, & Harvey, 2016; Lucksted, McFarlane, Downing, & Dixon, 2012; Reinares et al., 2016).

FPE is a common intervention for addressing the needs of families and persons with neurodevelopmental disabilities (DaWalt, Greenberg, & Mailick, 2018; Dykens, 2015); however, few studies address FPE in dually diagnosed persons. Persons dually diagnosed with borderline intellectual functioning and schizophrenia have successfully participated in a general FPE group with gains in knowledge and improvement in treatment compliance similar to the general group (Pitschel-Walz, Bäuml, Froböse, Gsottschneider, & Jahn, 2009). A preliminary study including caregivers and persons dually diagnosed with schizophrenia and with mild ($n = 6$) or borderline ($n = 2$) intellectual disability found psychoeducation delivered in a group setting improved participants' understanding of psychosis, the importance of medication, and the role of stressors and individual factors in relapse (Crowley, Rose, Smith, Hobster, & Ansell, 2008). The Staying Well and Mind Matters psychoeducational programs address the needs of persons dually diagnosed with intellectual disability and serious mental illness in hospital settings (Ashworth, Jansen, Bullock, & Mooney, 2017; Douds, McKechanie, Simpson, & Murphy, 2014). The Staying Well program targeted relapse prevention, guiding persons with mild and moderate intellectual disability in developing a Staying Well Plan (Douds et al., 2014). The Mind

Matters program addressed wellness strategies in a forensic setting, noting improved social skills in participants (Ashworth et al., 2017). Modifications to the FPE format included frequent repetition, reinforcing concepts with visual images, and the participation of a speech and language therapist (Douds et al., 2014; Pitschel-Walz et al., 2009).

Assertive Community Treatment (ACT)

Assertive community treatment is a person-centered, multicomponent intervention to support persons with serious mental illness in the community. Studies of ACT in persons with serious mental illness have demonstrated success with decreased need for hospitalization, and improvements in treatment compliance and quality of life (Dixon, 2000). The original description of ACT included guidelines calling for an *assertive approach* defined as *strong encouragement and support* (p. 53) and skills building though social learning techniques (Test & Stein, 1976). Bond and Drake (2015) have identified the essential elements of ACT as community-based service delivery, small caseloads, service integration, and a holistic continuity of care. A systematic review and meta-analysis of 64 studies representing 7819 patients found ACT comparable to community mental health treatment, citing improvements in community mental health treatment over the past 40 years as an intervening variable (Burns, 2010). The Substance Abuse and Mental Health Services Administration *Toolkit* for ACT program implementation and evaluation does not address dually diagnosed individuals (SAMHSA, 2008).

Few studies have reported on the response of dually diagnosed persons to ACT. It is likely that dually diagnosed individuals are being served in general ACT programs. For example, the Canadian province of Ontario reported 9.3% (414 persons) of the ACT clients carried dual diagnoses (Burge, 2009). Unfortunately, no outcomes were reported by this study. A program similar to ACT, Intensive Personalized Support, demonstrated efficacy in a small study (n = 4) with significant improvement in global functioning, quality of life, insight into illness, processing skills, and social connectedness (Raftery et al., 2017). A larger, randomized controlled trial (n = 30) found no significant benefit of ACT over regular community treatment (Oliver et al., 2005). Prakash, Andrews, and Porter (2007) reported improved service engagement and decreases in substance use and anxiety among 19 persons with mild ID and mental illness. A Flexible ACT program for persons with borderline or mild intellectual disability and mental illness or challenging behaviors (n = 78) reports preliminary success as well as challenges with funding and the high level of complex needs experienced by their study population (Neijmeijer, Didden, Nijman, & Kroon, 2018). While anecdotal reports are positive, the scarcity of data regarding ACT of dually diagnosed persons prohibits conclusion regarding efficacy.

Cognitive Behavioral Therapy for Psychosis (CBT-p)

In the 1950s, Aaron Beck, the father of CBT, documented the use of a cognitive behavioral approach to support persons with schizophrenia in achieving and sustaining an improved quality of life (Beck, 1952). With the introduction of antipsychotic medications for psychosis in the mid-1950s, CBT-p was not systematically studied until the 1990s (Chadwick & Lowe, 1994; Garety, Kuipers, Fowler, Chamberlain, & Dunn, 1994). Since 2004, professional organizations have recommended CBT-p as an adjunct to medication treatment for schizophrenia. It has been suggested that CBT-p may delay or prevent psychotic illness although meta-analyses have challenged these findings (Davies et al., 2018; Stafford, Jackson, Mayo-Wilson, Morrison, & Kendall, 2013; van der Gaag, van den Berg, & Ising, 2019). There has been a recent trend to identify the efficacy of specific components of CBT-p and to focus studies on distinct symptoms or a specific CBT-p approach (Dunn et al., 2012; Lincoln & Peters, 2019; Tomlinson & Wright,

2018). Unlike medication treatment which seeks to eliminate symptoms, CBT-p assists the patient in developing a new perspective on psychotic symptoms and learning skills to limit their impact on day-to-day functioning. Case studies have reported successful CBT-p interventions in dually diagnosed persons (Chadwick & Lowe, 1994; Haddock, Lobban, Hatton, & Carson, 2004).

Multiple models of CBT-p have been investigated with common elements highlighting the importance of assessment, engagement, formulation, change strategies with homework, and relapse prevention (Morrison & Barratt, 2010). Although psychosis is not specifically addressed, Hassiotis et al. (2012) have published a detailed manual with modifications of CBT for persons with mild and moderate intellectual disability. Before the CBT assessment phase begins, an individual's capacity for CBT is evaluated. Capacities for communication, recognizing emotions, and distinguishing behaviors, thoughts, and feelings should be considered (Hassiotis et al., 2012). Oathamshaw and Haddock (2006) assessed 50 individuals dually diagnosed with intellectual disability and psychosis for capacity to participate in CBT, reporting that persons with mild and moderate intellectual disability with and without psychosis have similar capacities for CBT. Their study found particular challenges with perception of cognitions and cognitive mediation, the ability to associate events with subsequent thoughts or actions. For persons with the capacity for CBT-p, modifications may include additional time for assessment and engagement, adaptations to materials, use of manipulatives and visual aids, and caregiver involvement (Favrod, Linder, Pernier, & Chafloque, 2007; Haddock et al., 2004). As with any patient, clinicians should pay attention to communication style. Persons with intellectual disability may have developed coping mechanisms of agreeing with authority figures and may give the appearance of understanding to avoid embarrassment (Boardman, Bernal, & Hollins, 2014). Formulations will be more concrete and should be discussed with simple sentences, exploring each statement for full understanding before proceeding. Change strategies and homework may involve visual or text remainders and simple tasks such as attempting to record the voices to encourage discussion about alternative explanations for hallucinations (Morrison, 2017). Surley and Dagnan have compiled a comprehensive review of modifications of CBT for adults with intellectual disability using the Hurley framework (Hurley, Tomasulo, & Pfadt, 1998; Surley & Dagnan, 2018). Caregivers may provide important background information for formulations and assist with change strategies, homework, and implementation of a relapse plan; however, confidentiality and boundary issues will need to be considered. When implemented by a skilled clinician, CBT-p may be a useful psychosocial intervention for the dually diagnosed.

Cognitive Remediation Therapy (CRT)

The neurodevelopmental hypothesis of schizophrenia cites a common etiology for neurodevelopmental disorders and schizophrenia which may help explain the neurocognitive decline present before the onset of psychosis and persisting when psychosis is in remission (Murray & Lewis, 1988; Owen & O'Donovan, 2017). The benefits of CRT, an evidence-based intervention to address this decline, have been affirmed by multiple meta-analyses (Cella, Preti, Edwards, Dow, & Wykes, 2017; Reddy, Horan, Jahshan, & Green, 2014; Wykes, Huddy, Cellard, McGurk, & Czobor, 2011). The 2012 Cognitive Remediation Experts Workshop defined CRT as:

> an intervention targeting cognitive deficits using scientific principles of learning with the ultimate goal of improving functional outcomes. Its effectiveness is enhanced when provided in a context (formal or informal) that provides support and opportunity for extending everyday functioning (Wykes, 2018, p. 57–58).

CRT addresses the cognitive deficiencies associated with schizophrenia (attention, verbal memory, and executive functioning) through restorative and compensatory strategies with recognition that effective learning is mediated by cognitive ability, instruction, and motivation

(Medalia & Choi, 2009). Because approximately 30% of persons with 22q11.2 deletion syndrome (formerly known as DiGeorge, velocardiofacial, and Opitz syndrome) develop schizophrenia, adolescents with this genetic neurodevelopmental disorder have been treated with CRT. In a group of 21 adolescents with 22q11.2 deletion, Mariano, Tang, Kurtz, and Kates (2018) found that improvements in cognitive flexibility, executive functioning, reaction time, and working memory were evident 6 months posttreatment. Unfortunately, persons with intellectual disability and serious mental illness are often excluded from CRT studies. The benefits of CRT for working memory in persons with intellectual disability (without serious mental illness) has been explored. Kanthasamy, Lim, and Lee (2017) reported that 15 one-hour CRT group sessions addressing working memory in five women with mild or moderate intellectual disability decreased challenging behaviors in a residential setting; however, a meta-analysis of CRT for working memory found CRT promising but in need of additional study (Danielsson, Zottarel, Palmqvist, & Lanfranchi, 2015). As with other psychosocial interventions for dually diagnosed persons, CRT merits additional study.

Vignette: Conclusion

Six months later at her care team meeting, Ashley is well-groomed. She has lost weight due to medication change, less snacking, and regular exercise at the group home and long walks with her mother. She needs fewer reminders from others because she is using her CRT skill of setting alarms on her phone. Lynda helped her put emojis of tasks she needs to complete in the alarm note section. On returning to work, Ashley had trouble with her old routine so she was moved to a clothes hanging task; her movement in completing this task helps her stay alert and more focused.

The team reviews Ashley's progress and prognosis. The psychiatrist reports Abilify is working well for Ashley, but as expected, it has a greater impact on the positive symptoms of schizophrenia more than the negative. Ashley's parents are concerned that their daughter is "still not herself." As her father describes concerns, her mother recalls other families in their psychoeducational group have talked about letting go of expectations; adding "we are trying to enjoy who Ashley is now." She turns to Ashley, "I love our walks, thank you for helping me get in better shape." Ashley agrees to continue her medication, exercise, CRT, and monthly family psychoeducation.

Conclusion

It is well -documented that persons with intellectual disability are at increased risk for serious mental illness (Havercamp & Scott, 2015; Morgan, Leonard, Bourke, & Jablensky, 2008). To meet this treatment need, mental health professionals must understand the biopsychosocial complexities of intellectual disability and mental illness and be prepared to communicate effectively with patients and caregivers. Psychopharmacological and psychosocial interventions for schizophrenia spectrum disorders and bipolar disorder should be person-centered and maximize habilitation plans. Multimorbidity in the intellectually disabled population, including a fragile neurological system, demands cautious medication management. In accord with current epigenetic models, attention to healthy lifestyle factors such as diet, exercise, and sleep should not be overlooked as this can improve treatment outcome and minimize side effects of medications used to treat serious mental illness (Havercamp & Scott, 2015; Trollor et al., 2016). The limited evidence base for the treatment of severe psychopathology presents researchers and clinicians with the opportunity to contribute to an improved quality of life for the dually diagnosed.

References

AACAP. (2018). Parameters, Updates, and Guidelines. Retrieved from https://www.aacap.org/aacap/resources_for_primary_care/practice_parameters_and_resource_centers/practice_parameters.aspx

Alabaf, S., Gillberg, C., Lundström, S., Lichtenstein, P., Kerekes, N., Råstam, M., & Anckarsäter, H. (2019). Physical health in children with neurodevelopmental disorders. *Journal of Autism and Developmental Disorders, 49*(1), 83–95. Retrieved from https://doi.org/10.1007/s10803-018-3697-4

Alexander, M., Petri, H., Ding, Y., Wandel, C., Khwaja, O., & Foskett, N. (2016). Morbidity and medication in a large population of individuals with Down syndrome compared to the general population. *Developmental Medicine & Child Neurology, 58*(3), 246–254. Retrieved from https://doi.org/10.1111/dmcn.12868

Aman, H., Naeem, F., Farooq, S., & Ayub, M. (2016). Prevalence of nonaffective psychosis in intellectually disabled clients: systematic review and meta-analysis. *Psychiatric Genetics, 26*(4), 145–155. Retrieved from <Go to ISI>://WOS:000379377700001. https://doi.org/10.1097/ypg.0000000000000137

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

American Psychiatric Association, & American Psychiatric Association. DSM-5 Task Force. (2013). *Diagnostic and statistical manual of mental disorders : DSM-5* (5th ed.). Washington, D.C.: American Psychiatric Association.

Andreasen, N. (1984). *The scale for the assessment of negative symptoms (SANS) University of Iowa*. Iowa City, Iowa.

Anthony, S. A. (2018). Focus on eye care in schizophrenia. *Clinical and Experimental Optometry*. Retrieved from https://doi.org/10.1111/cxo.12826

Antonacci, D. J., & Attiah, N. (2008). Diagnosis and treatment of mood disorders in adults with developmental disabilities. *The Psychiatric Quarterly, 79*(3), 171–192. https://doi.org/10.1007/s11126-008-9079-x

Ashworth, S., Jansen, K., Bullock, L., & Mooney, P. (2017). Mind matters: A psychoeducation programme for individuals with intellectual disabilities and co-morbid diagnoses of mental disorder. *Journal of Intellectual Disabilities and Offending Behaviour, 8*(1), 34–40. Retrieved from https://doi.org/10.1108/JIDOB-07-2016-0011

Ayub, M., Saeed, K., Munshi, T. A., & Naeem, F. (2015). Clozapine for psychotic disorders in adults with intellectual disabilities. *Cochrane Database of Systematic Reviews, 9*. Retrieved from https://doi.org//10.1002/14651858.CD010625.pub2

Baldessarini, R. J., Tondo, L., & Vázquez, G. H. (2019). Pharmacological treatment of adult bipolar disorder. *Molecular Psychiatry, 24*(2), 198–217. Retrieved from https://doi.org/10.1038/s41380-018-0044-2

Bamburg, J. W., Cherry, K. E., Matson, J. L., & Penn, D. (2001). Assessment of schizophrenia in persons with severe and profound mental retardation using the Diagnostic Assessment for the Severely Handicapped-II (DASH-II). *Journal of Developmental and Physical Disabilities, 13*(4), 319–331. Retrieved from https://doi.org/10.1023/A:1012218611103

Barnes, T. R. E. (1989). A rating scale for drug-induced akathisia. *British Journal of Psychiatry, 154*(5), 672–676. Retrieved from https://www.cambridge.org/core/article/rating-scale-for-druginduced-akathisia/77334A34A80E801C6297640C63701866. https://doi.org/10.1192/bjp.154.5.672

Barnes, T. R. E., & Schizophrenia Consensus Group of the British Association for, P. (2011). Evidence-based guidelines for the pharmacological treatment of schizophrenia: Recommendations from the British Association for Psychopharmacology. *Journal of Psychopharmacology, 25*(5), 567–620.

Beach, S. R., Gomez-Bernal, F., Huffman, J. C., & Fricchione, G. L. (2017). Alternative treatment strategies for catatonia: A systematic review. *General Hospital Psychiatry, 48*, 1–19.

Beck, A. T. (1952). Successful outpatient psychotherapy of a chronic schizophrenic with a delusion based on borrowed guilt. *Psychiatry, 15*(3), 305–312.

Belling, K., Russo, F., Jensen, A. B., Dalgaard, M. D., Westergaard, D., Rajpert-De Meyts, E., ... Brunak, S. (2017). Klinefelter syndrome comorbidities linked to increased X chromosome gene dosage and altered protein interactome activity. *Human Molecular Genetics, 26*(7), 1219–1229. Retrieved from https://www.ncbi.nlm.nih.gov/pubmed/28369266. Retrieved from https://www.ncbi.nlm.nih.gov/pmc/PMC5390676/. https://doi.org/10.1093/hmg/ddx014

Bhaumik, S., Gangadharan, S. K., Branford, D., & Barrett, M. (2015). *The Frith prescribing guidelines for people with intellectual disability*. John Wiley & Sons, West Sussex, UK.

Boardman, L., Bernal, J., & Hollins, S. (2014). Communicating with people with intellectual disabilities: A guide for general psychiatrists. *Advances in Psychiatric Treatment, 20*(1), 27–36.

Bolhuis, K., Tiemeier, H., Jansen, P. R., Muetzel, R. L., Neumann, A., Hillegers, M. H. J., … Kushner, S. A. (2019). Interaction of schizophrenia polygenic risk and cortisol level on pre-adolescent brain structure. *Psychoneuroendocrinology, 101*, 295–303. Retrieved from http://www.sciencedirect.com/science/article/pii/S0306453018308862. https://doi.org/10.1016/j.psyneuen.2018.12.231

Bond, G. R., & Drake, R. E. (2015). The critical ingredients of assertive community treatment. *World Psychiatry: Official Journal of the World Psychiatric Association (WPA), 14*(2), 240–242. Retrieved from https://www.ncbi.nlm.nih.gov/pubmed/26043344. Retrieved from https://www.ncbi.nlm.nih.gov/pmc/PMC4471983/. https://doi.org/10.1002/wps.20234

Bonnot, O., Cohen, D., Thuilleaux, D., Consoli, A., Cabal, S., & Tauber, M. (2016). Psychotropic treatments in Prader-Willi syndrome: A critical review of published literature. *European Journal of Pediatrics, 175*(1), 9–18.

Bonnot, O., Klünemann, H. H., Sedel, F., Tordjman, S., Cohen, D., & Walterfang, M. (2014). Diagnostic and treatment implications of psychosis secondary to treatable metabolic disorders in adults: a systematic review. *Orphanet Journal of Rare Diseases, 9*, 65. Retrieved from. https://www.ncbi.nlm.nih.gov/pubmed/24775716. https://doi.org/10.1186/1750-1172-9-65

Boot, E., Butcher, N. J., van Amelsvoort, T. A. M. J., Lang, A. E., Marras, C., Pondal, M., … Bassett, A. S. (2015). Movement disorders and other motor abnormalities in adults with 22q11. 2 deletion syndrome. *American Journal of Medical Genetics Part A, 167*(3), 639–645.

Bouras, N., Martin, G., Leese, M., Vanstraelen, M., Holt, G., Thomas, C., … Boardman, J. (2004). Schizophrenia-spectrum psychoses in people with and without intellectual disability. *Journal of Intellectual and Disability Research, 48*(Pt 6), 548–555.. Retrieved from https://www.ncbi.nlm.nih.gov/pubmed/15312055. https://doi.org/10.1111/j.1365-2788.2004.00623.x

Bousman, C. A., Menke, A., & Müller, D. J. (2019). *Towards pharmacogenetic-based treatment in psychiatry*. Journal of Neural Transmission (Vienna). Retrieved from https://www.ncbi.nlm.nih.gov/pubmed/30673860. https://doi.org/10.1007/s00702-018-01968-9

Brameld, K., Spilsbury, K., Rosenwax, L., Leonard, H., & Semmens, J. (2018). Use of health services in the last year of life and cause of death in people with intellectual disability: a retrospective matched cohort study. *British Medical Journal open, 8*(2), e020268-e020268. Retrieved from https://www.ncbi.nlm.nih.gov/pubmed/29478966. Retrieved from https://www.ncbi.nlm.nih.gov/pmc/PMC5855242/. https://doi.org/10.1136/bmjopen-2017-020268

Burge, P. (2009). Assertive community treatment teams and adults with intellectual disabilities. *Journal on Developmental Disabilities, 15*(3), 96.

Burns, T. (2010). The rise and fall of assertive community treatment? *International Reviews in Psychiatry, 22*(2), 130–137. https://doi.org/10.3109/09540261003661841

Bush, G., Fink, M., Petrides, G., Dowling, F., & Francis, A. (1996). Catatonia. I. Rating scale and standardized examination. *Acta Psychiatrica Scandinavica, 93*(2), 129–136.

Cederlöf, M., Ohlsson Gotby, A., Larsson, H., Serlachius, E., Boman, M., Långström, N., … Lichtenstein, P. (2014). Klinefelter syndrome and risk of psychosis, autism and ADHD. *Journal of Psychiatric Research, 48*(1), 128–130. Retrieved from http://www.sciencedirect.com/science/article/pii/S0022395613003099. https://doi.org/10.1016/j.jpsychires.2013.10.001

Cella, M., Preti, A., Edwards, C., Dow, T., & Wykes, T. (2017). Cognitive remediation for negative symptoms of schizophrenia: A network meta-analysis. *Clinical Psychology Review, 52*, 43–51.

Chadwick, P. D. J., & Lowe, C. F. (1994). A cognitive approach to measuring and modifying delusions. *Behaviour Research and Therapy, 32*(3), 355–367. Retrieved from http://www.sciencedirect.com/science/article/pii/0005796794901333. https://doi.org/10.1016/0005-7967(94)90133-3

Chien, W. T., & Yip, A. L. (2013). Current approaches to treatments for schizophrenia spectrum disorders, part I: An overview and medical treatments. *Neuropsychiatric Disease and Treatment, 9*, 1311–1332. Retrieved from. https://www.ncbi.nlm.nih.gov/pubmed/24049446. https://doi.org/10.2147/NDT.S37485

Clinebell, K., Azzam, P. N., Gopalan, P., & Haskett, R. (2014). Guidelines for preventing common medical complications of catatonia: Case report and literature review. *The Journal of Clinical Psychiatry, 75*(6), 644–651.

Coker, F., Williams, A., Hayes, L., Hamann, J., & Harvey, C. (2016). Exploring the needs of diverse consumers experiencing mental illness and their families through family psychoeducation. *Journal of Mental Health, 25*(3), 197–203. https://doi.org/10.3109/09638237.2015.1057323

Consoli, A., Raffin, M., Laurent, C., Bodeau, N., Campion, D., Amoura, Z., … Cohen, D. (2012). Medical and developmental risk factors of catatonia in children and adolescents: A prospective case–control study. *Schizophrenia Research, 137*(1–3), 151–158.

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. *BioMed Central Family Practice, 16*(1), 110. Retrieved from https://doi.org/10.1186/s12875-015-0329-3

Cooper, S.-A., Smiley, E., Allan, L., & Morrison, J. (2018). Incidence of unipolar and bipolar depression, and mania in adults with intellectual disabilities: Prospective cohort study. *The British Journal of Psychiatry, 212*(5), 295–300. Retrieved from https://www.cambridge.org/core/article/incidence-of-unipolar-and-bipolar-depression-and-mania-in-adults-with-intellectual-disabilities-prospective-cohort-study/B5F4AE6461D1F33AE454257CC9D016D3. https://doi.org/10.1192/bjp.2018.12

Crowley, V., Rose, J., Smith, J., Hobster, K., & Ansell, E. (2008). Psycho-educational groups for people with a dual diagnosis of psychosis and mild intellectual disability: A preliminary study. *Journal of Intellectual Disability, 12*(1), 25–39. https://doi.org/10.1177/1744629507086606

Dagnan, D., & Rodgers, J. (2018). Exploring the emotion regulation strategies used by adults with intellectual disabilities AU – Littlewood, Mark. *International*

Journal of Developmental Disabilities, 64(3), 204–211. Retrieved from https://doi.org/10.1080/20473869.2018.1466510

Danielsson, H., Zottarel, V., Palmqvist, L., & Lanfranchi, S. (2015). The effectiveness of working memory training with individuals with intellectual disabilities–a meta-analytic review. *Frontiers in Psychology, 6*, 1230.

Davies, C., Cipriani, A., Ioannidis, J. P. A., Radua, J., Stahl, D., Provenzani, U., … Fusar-Poli, P. (2018). Lack of evidence to favor specific preventive interventions in psychosis: A network meta-analysis. *World Psychiatry, 17*(2), 196–209. Retrieved from https://doi.org/10.1002/wps.20526

Davit, C. J., Hundley, R. J., Bacic, J. D., & Hanson, E. M. (2011). A pilot study to improve venipuncture compliance in children and adolescents with autism spectrum disorders. *Journal of Developmental and Behavioral Pediatrics: JDBP, 32*(7), 521–525. Retrieved from https://www.ncbi.nlm.nih.gov/pubmed/21694630. Retrieved from https://www.ncbi.nlm.nih.gov/pmc/PMC3690561/. https://doi.org/10.1097/DBP.0b013e3182245b09

DaWalt, L. S., Greenberg, J. S., & Mailick, M. R. (2018). Transitioning together: A multi-family group psychoeducation program for adolescents with ASD and their parents. *Journal of Autism and Developmental Disorders, 48*(1), 251–263.

Da Silva, M. E., Fregni, F., & Brunoni, A. R. (2013). Transcranial direct current stimulation (tDCS) for catatonic schizophrenia: a case study. Schizophrenia research, 146(1–3), 374–375.

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 Developemental Disability, 30*(4), 613–669. https://doi.org/10.1016/j.ridd.2008.10.010

de Winter, C. F., Bastiaanse, L. P., Hilgenkamp, T. I. M., Evenhuis, H. M., & Echteld, M. A. (2012). Cardiovascular risk factors (diabetes, hypertension, hypercholesterolemia and metabolic syndrome) in older people with intellectual disability: Results of the HA-ID study. *Research in Developmental Disabilities, 33*(6), 1722–1731. Retrieved from http://www.sciencedirect.com/science/article/pii/S0891422212001023. https://doi.org/10.1016/j.ridd.2012.04.010

Deb, S., Kwok, H., Bertelli, M., Salvador-Carulla, L., Bradley, E., Torr, J., … Disability, G. D. G. (2009). International guide to prescribing psychotropic medication for the management of problem behaviours in adults with intellectual disabilities. *World Psychiatry, Retrieved from, 8*(3), 181–186. https://www.ncbi.nlm.nih.gov/pubmed/19812757

DeJong, H., Bunton, P., & Hare, D. J. (2014). A systematic review of interventions used to treat catatonic symptoms in people with autistic spectrum disorders. *Journal of Autism and Developmental Disorders, 44*(9), 2127–2136.

Dhossche, D. M., van der Steen, L. F., Shettar, S. M., Krefft, M., Frydecka, D., Adamowski, T., ... Forsyth, J. K. (2015). [Catatonia in autism spectrum disorders: Review and case-report] genetics of childhood-onset schizophrenia. In *Tijdschrift voor Psychiatrie* (Vol. 57, pp. 89–93). Netherlands.

Dixon, L. (2000). Assertive community treatment: Twenty-five years of gold. *Psychiatric Services, 51*(6), 759–765. Retrieved from https://doi.org/10.1176/appi.ps.51.6.759

Dixon, L., McFarlane, W. R., Lefley, H., Lucksted, A., Cohen, M., Falloon, I., … Sondheimer, D. (2001). Evidence-based practices for services to families of people with psychiatric disabilities. *Psychiatric Services, 52*(7), 903–910.

Dixon, L., Perkins, D., & Calmes, C. (2010). Guideline watch (September 2009): Practice guideline for the treatment of patients with schizophrenia. In *APA Practice Guidelines*: American Psychiatric Publishing

Dols, A., Sienaert, P., van Gerven, H., Schouws, S., Stevens, A., Kupka, R., & Stek, M. L. (2013). The prevalence and management of side effects of lithium and anticonvulsants as mood stabilizers in bipolar disorder from a clinical perspective: A review. *International Clinical Psychopharmacology, 28*(6). Retrieved from https://journals.lww.com/intclinpsychopharm/Fulltext/2013/11000/The_prevalence_and_management_of_side_effects_of.1.aspx

Douds, F., McKechanie, A., Simpson, Y., & Murphy, L. (2014). "Staying well": A psychoeducational group for people with an intellectual disability, co-morbid mental illness and offending behaviour. *Journal of Intellectual Disabilities and Offending Behaviour, 5*(1), 54–59.

Dunn, G., Fowler, D., Rollinson, R., Freeman, D., Kuipers, E., Smith, B., … Bebbington, P. (2012). Effective elements of cognitive behaviour therapy for psychosis: results of a novel type of subgroup analysis based on principal stratification. *Psychological Medicine, 42*(5), 1057–1068. Retrieved from https://www.ncbi.nlm.nih.gov/pubmed/21939591. Retrieved from https://www.ncbi.nlm.nih.gov/pmc/PMC3315767/. https://doi.org/10.1017/S0033291711001954

Dykens, E. M. (2015). Family adjustment and interventions in neurodevelopmental disorders. *Current Opinion in Psychiatry, 28*(2), 121–126. Retrieved from https://www.ncbi.nlm.nih.gov/pubmed/25594421. Retrieved from https://www.ncbi.nlm.nih.gov/pmc/PMC5348480/. https://doi.org/10.1097/YCO.0000000000000129

Emerson, E., Hatton, C., Baines, S., & Robertson, J. (2016). The physical health of British adults with intellectual disability: cross sectional study. *International Journal for Equity in Health, 15*, 11–11. Retrieved from https://www.ncbi.nlm.nih.gov/pubmed/26791808. Retrieved from https://www.ncbi.nlm.nih.gov/pmc/PMC4719222/. https://doi.org/10.1186/s12939-016-0296-x

Excellence, N. I. f. C. (2014). Psychosis and schizophrenia in adults: Prevention and management; National Clinical Practice Guidelines Number CG178.

Favrod, J., Linder, S., Pernier, S., & Chafloque, M. N. (2007). Cognitive and behavioural therapy of voices for with patients intellectual disability: two case reports. *Annals of General Psychiatry, 6*, 22–22. Retrieved from https://www.ncbi.nlm.nih.gov/pubmed/17705875. Retrieved from https://www.ncbi.nlm.nih.gov/pmc/PMC1994678/. https://doi.org/10.1186/1744-859X-6-22

Fink, M., & Taylor, M. A. (2006). *Catatonia: A clinician's guide to diagnosis and treatment.* Cambridge University Press. Cambridge, UK.

Fisher, A. D., Castellini, G., Casale, H., Fanni, E., Bandini, E., Campone, B., … Maggi, M. (2015). Hypersexuality, paraphilic behaviors, and gender dysphoria in individuals with Klinefelter's syndrome. *The Journal of Sexual Medicine, 12*(12), 2413–2424. Retrieved from http://www.sciencedirect.com/science/article/pii/S1743609516300157. https://doi.org/10.1111/jsm.13048

Fletcher, R., Barnhill, J., & McCarthy, S.-A. (2016). *Diagnostic Manual-Intellectual Disability (DM-ID): A clinical guide for diagnosis of mental disorders in persons with intellectual disability* (2nd ed.). Kingston, NY: NADD.

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. Retrieved from http://www.sciencedirect.com/science/article/pii/S0272735817301368. https://doi.org/10.1016/j.cpr.2017.08.006

Forbes, C., Blanchard, J. J., Bennett, M., Horan, W. P., Kring, A., & Gur, R. (2010). Initial development and preliminary validation of a new negative symptom measure: the Clinical Assessment Interview for Negative Symptoms (CAINS). *Schizophrenia research, 124*(1–3), 36–42. Retrieved from https://www.ncbi.nlm.nih.gov/pubmed/20869848. Retrieved from https://www.ncbi.nlm.nih.gov/pmc/PMC2981616/. https://doi.org/10.1016/j.schres.2010.08.039

Fraidakis, M. J. (2013). Psychiatric manifestations in cerebrotendinous xanthomatosis. *Translational Psychiatry, 3*(9), e302. Retrieved from https://www.ncbi.nlm.nih.gov/pubmed/24002088. Retrieved from https://www.ncbi.nlm.nih.gov/pmc/PMC3784765/. https://doi.org/10.1038/tp.2013.76

Funayama, M., Takata, T., Koreki, A., Ogino, S., & Mimura, M. (2018). Catatonic stupor in schizophrenic disorders and subsequent medical complications and mortality. *Psychosomatic Medicine, 80*(4), 370.

Fung, W. L. A., Butcher, N. J., Costain, G., Andrade, D. M., Boot, E., Chow, E. W. C., … Bassett, A. S. (2015). Practical guidelines for managing adults with 22q11.2 deletion syndrome. *Genetics in Medicine, 17*, 599. Retrieved from https://doi.org/10.1038/gim.2014.175

Gaebel, W., Riesbeck, M., & Wobrock, T. (2011). Schizophrenia guidelines across the world: A selective review and comparison. *International Review of Psychiatry, 23*(4), 379–387. Retrieved from http://search.ebscohost.com/login.aspx?direct=true&db=pbh&AN=66813180&site=ehost-live. https://doi.org/10.3109/09540261.2011.606801

Garety, P. A., Kuipers, L., Fowler, D., Chamberlain, F., & Dunn, G. (1994). Cognitive behavioural therapy for drug-resistant psychosis. *British Journal of Medical Psychology, 67*(3), 259–271.

Geberhiwot, T., Moro, A., Dardis, A., Ramaswami, U., Sirrs, S., Marfa, M. P., … on behalf of the International Niemann-Pick Disease, R. (2018). Consensus clinical management guidelines for Niemann-Pick disease type C. *Orphanet Journal of Rare Diseases, 13*(1), 50. Retrieved from https://doi.org/10.1186/s13023-018-0785-7

Geiss, M., Chamberlain, J., Weaver, T., McCormick, C., Raufer, A., Scoggins, L., … Edmonson, D. (2017). Diagnostic overshadowing of the psychiatric population in the emergency department: Physiological factors identified for an early warning system. *Journal of the American Psychiatric Nurses Association, 24*(4), 327–331. Retrieved from https://doi.org/10.1177/1078390317728775

Ghaziuddin, N., Nassiri, A., & Miles, J. H. (2015). Catatonia in Down syndrome; A treatable cause of regression. *Neuropsychiatric Disease and Treatment, 11*, 941–949. Retrieved from https://www.ncbi.nlm.nih.gov/pubmed/25897230. Retrieved from https://www.ncbi.nlm.nih.gov/pmc/PMC4396650/. https://doi.org/10.2147/NDT.S77307

Giannitelli, M., Consoli, A., Raffin, M., Jardri, R., Levinson, D. F., Cohen, D., & Laurent-Levinson, C. (2018). An overview of medical risk factors for childhood psychosis: Implications for research and treatment. *Schizophrenia Research, 192*, 39–49. Retrieved from http://www.sciencedirect.com/science/article/pii/S0920996417302700. https://doi.org/10.1016/j.schres.2017.05.011

Gijssen, R., Stirbu, I., Korevaar, J. C., Schellevis, F. G., Picavet, H. S. J., & Hoeymans, N. (2017). Time trends in prevalence of chronic diseases and multimorbidity not only due to aging: Data from general practices and health surveys. *Nederlands Tijdschrift voor Geneeskunde, 161*, D1429–D1429.

Goodwin, G. M., Haddad, P. M., Ferrier, I. N., Aronson, J. K., Barnes, T. R. H., Cipriani, A., … Grunze, H. (2016). Evidence-based guidelines for treating bipolar disorder: Revised third edition recommendations from the British Association for Psychopharmacology. *Journal of Psychopharmacology, 30*(6), 495–553.

Grier, E., Abells, D., Casson, I., Gemmill, M., Ladouceur, J., Lepp, A., … Sue, K. (2018). Managing complexity in care of patients with intellectual and developmental disabilities: Natural fit for the family physician as

an expert generalist. *Canadian Family Physician, 64*(Suppl 2), S15–S22. Retrieved from https://www.ncbi.nlm.nih.gov/pubmed/29650740

Groth, K. A., Gravholt, C. H., Skakkebæk, A., Høst, C., & Bojesen, A. (2013). Klinefelter syndrome—A clinical update. *The Journal of Clinical Endocrinology & Metabolism, 98*(1), 20–30. Retrieved from https://doi.org/10.1210/jc.2012-2382

Guy, W. (1976). Abnormal involuntary movement scale (AIMS). *Assessment Manual for Psychopharmacology, Revised*.

Haddad, P. M., Fleischhacker, W. W., Peuskens, J., Cavallaro, R., Lean, M. E., Morozova, M., ... Möller, H.-J. (2014). SMARTS (Systematic Monitoring of Adverse events Related to TreatmentS): The development of a pragmatic patient-completed checklist to assess antipsychotic drug side effects. *Therapeutic Advances in Psychopharmacology, 4*(1), 15–21. Retrieved from https://www.ncbi.nlm.nih.gov/pubmed/24490026. Retrieved from https://www.ncbi.nlm.nih.gov/pmc/PMC3896136/. https://doi.org/10.1177/2045125313510195

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. Retrieved from https://doi.org/10.1002/cpp.414

Haddock, G., McCarron, J., Tarrier, N., & Faragher, E. B. (1999). Scales to measure dimensions of hallucinations and delusions: the psychotic symptom rating scales (PSYRATS). *Psychological medicine, 29*(4), 879–889. Retrieved from http://europepmc.org/abstract/MED/10473315. Retrieved from https://doi.org/10.1017/S0033291799008661.

Harvey, C. (2018). Family psychoeducation for people living with schizophrenia and their families. *British Journal of Psychiatric Advances, 24*(1), 9–19. Retrieved from https://www.cambridge.org/core/article/family-psychoeducation-for-people-living-with-schizophrenia-and-their-families/1F624040803C69204CB936C7826185E3. https://doi.org/10.1192/bja.2017.4

Hassiotis, A., Serfaty, M., Azam, K., Strydom, A., Blizard, R., Romeo, R., ... King, M. B. (2012). A Manual of Cognitive Behaviour Therapy for People with Mild Learning Disabilities and Common Mental Disorders: A training guide to help professional therapists in treating people with communication and cognitive problems in CBT. In: Camden & Islington NHS Foundation Trust and University College London.

Hatton, C., Haddock, G., Taylor, J. L., Coldwell, J., Crossley, R., & Peckham, N. (2005). The reliability and validity of general psychotic rating scales with people with mild and moderate intellectual disabilities: An empirical investigation. *Journal of Intellectual Disability Research, 49*(Pt 7), 490–500. https://doi.org/10.1111/j.1365-2788.2005.00696.x

Havercamp, S. M., & Scott, H. M. (2015). National health surveillance of adults with disabilities, adults with intellectual and developmental disabilities, and adults with no disabilities. *Disability and Health Journal, 8*(2), 165–172. Retrieved from http://www.sciencedirect.com/science/article/pii/S1936657414001848. https://doi.org/10.1016/j.dhjo.2014.11.002

Hemm, C., Dagnan, D., & Meyer, T. D. (2015). Identifying training needs for mainstream healthcare professionals, to prepare them for working with individuals with intellectual disabilities: A systematic review. *Journal of Applied Research in Intellectual Disabilities, 28*(2), 98–110. Retrieved from https://doi.org/10.1111/jar.12117

Hermans, H., & Evenhuis, H. M. (2014). Multimorbidity in older adults with intellectual disabilities. *Research in Developmental Disabilities, 35*(4), 776–783. Retrieved from http://www.sciencedirect.com/science/article/pii/S0891422214000365. https://doi.org/10.1016/j.ridd.2014.01.022

Herrman, H., Saxena, S., & Moodie, R. (2005). Promoting mental health: concepts, emerging evidence, practice: a report of the World Health Organization, Department of Mental Health and Substance Abuse in collaboration with the Victorian Health Promotion Foundation and the University of Melbourne. World Health Organization.

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.

Husa, A. P., Moilanen, J., Murray, G. K., Marttila, R., Haapea, M., Rannikko, I., … Jääskeläinen, E. (2017). Lifetime antipsychotic medication and cognitive performance in schizophrenia at age 43 years in a general population birth cohort. *Psychiatry Research, 247*, 130–138. Retrieved from http://www.sciencedirect.com/science/article/pii/S0165178116305893. https://doi.org/10.1016/j.psychres.2016.10.085

Jap, S. N., & Ghaziuddin, N. (2011). Catatonia among adolescents with Down syndrome: A review and 2 case reports. *The Journal of Electroconvulsive Therapy, 27*(4). Retrieved from https://journals.lww.com/ectjournal/Fulltext/2011/12000/Catatonia_Among_Adolescents_With_Down_Syndrome__A.14.aspx

Ji, N. Y., & Findling, R. L. (2016). Pharmacotherapy for mental health problems in people with intellectual disability. *Current Opinion in Psychiatry, 29*(2), 103–125.

Jung, H.-Y., Kim, J.-H., Ahn, Y.-M., Kim, S.-C., Hwang, S. S., & Kim, Y.-S. (2005). Liverpool University Neuroleptic Side-Effect Rating Scale (LUNSERS) as a subjective measure of drug-induced parkinsonism and akathisia. *Human Psychopharmacology: Clinical and Experimental, 20*(1), 41–45. Retrieved from https://doi.org/10.1002/hup.655

Kanakis, G. A., & Nieschlag, E. (2018). Klinefelter syndrome: More than hypogonadism. *Metabolism, 86*, 135–144.

Kanthasamy, S., Lim, J. M., & Lee, J. J. W. (2017, January). The Usefulness of Group Cognitive Remediation in Young Adults With Mild to Moderate Intellectual

Disability: A Pilot Study. In Journal of Mental Health Research in Intellectual Disabilities (Vol. 10, pp. 215-216). 2-4 Park Square, Milton Park, Abingdon OX14 4RN, Oxon, England: Routledge Journals, Taylor & Francis LTD.

Karson, C., Duffy, R. A., Eramo, A., Nylander, A.-G., & Offord, S. J. (2016). Long-term outcomes of antipsychotic treatment in patients with first-episode schizophrenia: a systematic review. *Neuropsychiatric Disease and Treatment, 12*, 57–67. Retrieved from https://www.ncbi.nlm.nih.gov/pubmed/26792993. Retrieved from https://www.ncbi.nlm.nih.gov/pmc/PMC4708960/. https://doi.org/10.2147/NDT.S96392

Kay, S. R., Fiszbein, A., & Opler, L. A. (1987). The positive and negative syndrome scale (PANSS) for schizophrenia. *Schizophrenia Bulletin, 13*(2), 261–276.

Kirkpatrick, B., Strauss, G. P., Nguyen, L., Fischer, B. A., Daniel, D. G., Cienfuegos, A., & Marder, S. R. (2010). The brief negative symptom scale: Psychometric properties. *Schizophrenia Bulletin, 37*(2), 300–305. Retrieved from https://doi.org/10.1093/schbul/sbq059

Klünemann, H. H., Santosh, P. J., & Sedel, F. (2012). Treatable metabolic psychoses that go undetected: what Niemann-Pick type C can teach us. *International journal of psychiatry in clinical practice, 16*(3), 162–169.

Ko, J. M. (2015). Genetic Syndromes associated with Congenital Heart Disease. *Korean Circulation Journal, 45*(5), 357–361. Retrieved from https://www.ncbi.nlm.nih.gov/pubmed/26413101. Retrieved from https://www.ncbi.nlm.nih.gov/pmc/PMC4580692/. https://doi.org/10.4070/kcj.2015.45.5.357

Kraepelin, E., Barclay, R. M., & Robertson, G. M. (1919). Dementia praecox and paraphrenia.

Kuipers, E., Yesufu-Udechuku, A., Taylor, C., & Kendall, T. (2014). Management of psychosis and schizophrenia in adults: summary of updated NICE guidance. *British Medical Journal (Online),* 348.

Larson, F. V., Whittington, J., Webb, T., & Holland, A. J. (2014). A longitudinal follow-up study of people with Prader–Willi syndrome with psychosis and those at increased risk of developing psychosis due to genetic subtype. *Psychological Medicine, 44*(11), 2431–2435. Retrieved from https://www.cambridge.org/core/article/longitudinal-followup-study-of-people-with-praderwilli-syndrome-with-psychosis-and-those-at-increased-risk-of-developing-psychosis-due-to-genetic-subtype/63A7100963DDBDA22B3BAD33CC52B999. https://doi.org/10.1017/S0033291713002961

Lehman, A. F., Lieberman, J. A., Dixon, L. B., McGlashan, T. H., Miller, A. L., Perkins, D. O., … Altshuler, K. (2004). Practice guideline for the treatment of partients with schizophrenia. *American Journal of Psychiatry, 161*(2 SUPPL).

Lehman, A. F., Steinwachs, D. M., & Co-Investigators of the, P. P. (1998). Translating research into practice: The schizophrenia patient outcomes research team (PORT) treatment recommendations. *Schizophrenia Bulletin, 24*(1), 1–10.

Lehman, A. F., Lieberman, J., & Dixon, L. B. (2010). Treatment of Patients with Schizophrenia. American Psychiatric Association.

Levitas, A. S., Hurley, A. D., & Pary, R. (2001). The mental statys examination in patients with mental retardation and developmental disabilities. *Mental Health Aspects of Developmental Disabilities, 4*(1), 1–16.

Lin, A. E., Basson, C. T., Goldmuntz, E., Magoulas, P. L., McDermott, D. A., McDonald-McGinn, D. M., ... Pober, B. R. (2008). Adults with genetic syndromes and cardiovascular abnormalities: Clinical history and management. *Genetics in Medicine: Official Journal of the American College of Medical Genetics, 10*(7), 469–494. Retrieved from https://www.ncbi.nlm.nih.gov/pubmed/18580689. Retrieved from https://www.ncbi.nlm.nih.gov/pmc/PMC2671242/. https://doi.org/10.1097/GIM.0b013e3181772111

Lincoln, T. M., & Peters, E. (2019). A systematic review and discussion of symptom specific cognitive behavioural approaches to delusions and hallucinations. *Schizophrenia Research, 203*, 66–79. https://doi.org/10.1016/j.schres.2017.12.014

Lingjaerde, O., Ahlfors, U. G., Bech, P., Dencker, S. J., & Elgen, K. (1987). The UKU side effect rating scale: A new comprehensive rating scale for psychotropic drugs and a cross-sectional study of side effects in neuroleptic-treated patients. *Acta Psychiatrica Scandinavica, 76*, 1–100.

Louise Tveter, A., Lise Bakken, T., Bramness, G., & Ivar Røssberg, J. (2014). Adjustment of the UKU side effect rating scale for adults with intellectual disabilities. A pilot study. *Advances in Mental Health and Intellectual Disabilities, 8*(4), 260–267.

Lucksted, A., McFarlane, W., Downing, D., & Dixon, L. (2012). Recent developments in family psychoeducation as an evidence-based practice. *Journal of Marital and Family Therapy, 38*(1), 101–121. Retrieved from https://doi.org/10.1111/j.1752-0606.2011.00256.x

Manohar, H., Subramanian, K., Kandasamy, P., Penchilaiya, V., & Arun, A. (2016). Diagnostic masking and overshadowing in intellectual Disability—How structured evaluation helps. *Journal of Child and Adolescent Psychiatric Nursing, 29*(4), 171–176. Retrieved from https://doi.org/10.1111/jcap.12160

Mariano, M. A., Tang, K., Kurtz, M., & Kates, W. R. (2018). Examining the durability of a hybrid, remote and computer-based cognitive remediation intervention for adolescents with 22q11.2 deletion syndrome. *Early Intervention Psychiatry, 12*(4), 686–693. https://doi.org/10.1111/eip.12367

Maski, K. P., Jeste, S. S., & Spence, S. J. (2011). Common neurological co-morbidities in autism spectrum disorders. *Current Opinion in Pediatrics, 23*(6), 609–615. Retrieved from https://www.ncbi.nlm.nih.gov/pubmed/21970828. Retrieved from https://www.ncbi.nlm.nih.gov/pmc/PMC4229811/. https://doi.org/10.1097/MOP.0b013e32834c9282

Masnoon, N., Shakib, S., Kalisch-Ellett, L., & Caughey, G. E. (2017). What is polypharmacy? A systematic review of definitions. *BioMed Central Geriatrics, 17*(1), 230.

Matson, J. L., Bamburg, J. W., Mayville, E. A., Pinkston, J., Bielecki, J., Kuhn, D., … Logan, J. R. (2000). Psychopharmacology and mental retardation: A 10 year review (1990–1999). *Research in Developmental Disability, 21*(4), 263–296. Retrieved from https://www.ncbi.nlm.nih.gov/pubmed/10983783

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 Disability, 24*(3), 149–157. Retrieved from. https://www.ncbi.nlm.nih.gov/pubmed/12742384.

Matson, J. L., Coe, D. A., Gardner, W. I., & Sovner, R. (1991). A factor analytic study of the diagnostic assessment for the severely handicapped scale. *The Journal of Nervous and Mental Disease, 179*(9), 553–557. Retrieved from http://europepmc.org/abstract/MED/1833508

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 Disability, 31*(1), 108–116. Retrieved from. https://www.ncbi.nlm.nih.gov/pubmed/19720497. doi:https://doi.org/10.1016/j.ridd.2009.08.002

Matson, J. L., & Mahan, S. (2010). Antipsychotic drug side effects for persons with intellectual disability. *Research in Developmental Disability, 31*(6), 1570–1576. Retrieved from. https://www.ncbi.nlm.nih.gov/pubmed/20580203. doi: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.

Matson, J. L., & Shoemaker, M. E. (2011). Psychopathology and intellectual disability. *Current Opinion in Psychiatry, 24*(5), 367–371. Retrieved from. https://www.ncbi.nlm.nih.gov/pubmed/21150621. doi:https://doi.org/10.1097/YCO.0b013e3283422424

McCarron, M., Swinburne, J., Burke, E., McGlinchey, E., Carroll, R., & McCallion, P. (2013). Patterns of multimorbidity in an older population of persons with an intellectual disability: Results from the intellectual disability supplement to the Irish longitudinal study on aging (IDS-TILDA). *Research in Developmental Disabilities, 34*(1), 521–527. Retrieved from http://www.sciencedirect.com/science/article/pii/S0891422212001965. https://doi.org/10.1016/j.ridd.2012.07.029

McClellan, J., & Stock, S. (2013). Practice parameter for the assessment and treatment of children and adolescents with schizophrenia. *Journal of the American Academy of Child & Adolescent Psychiatry, 52*(9), 976–990. Retrieved from http://www.sciencedirect.com/science/article/pii/S0890856713001123. https://doi.org/10.1016/j.jaac.2013.02.008

Medalia, A., & Choi, J. (2009). Cognitive remediation in schizophrenia. *Neuropsychology Review, 19*(3), 353.

Mehta, N., & Vannozzi, R. (2017). Lithium-induced electrocardiographic changes: A complete review. *Clinical Cardiology, 40*(12), 1363–1367. Retrieved from https://doi.org/10.1002/clc.22822

Morgan, V. A., Leonard, H., Bourke, J., & Jablensky, A. (2008). Intellectual disability co-occurring with schizophrenia and other psychiatric illness: Population-based study. *British Journal of Psychiatry, 193*(5), 364–372. Retrieved from https://www.cambridge.org/core/article/intellectual-disability-cooccurring-with-schizophrenia-and-other-psychiatric-illness-populationbased-study/E994104AE84058AFB15C89484104B151. https://doi.org/10.1192/bjp.bp.107.044461

Morrison, A. P. (2017). A manualised treatment protocol to guide delivery of evidence-based cognitive therapy for people with distressing psychosis: Learning from clinical trials. *Psychosis, 9*(3), 271–281.

Morrison, A. P., & Barratt, S. (2010). What are the components of CBT for psychosis? A Delphi study. *Schizophrenia Bulletin, 36*(1), 136–142. Retrieved from https://www.ncbi.nlm.nih.gov/pubmed/19880824. Retrieved from https://www.ncbi.nlm.nih.gov/pmc/PMC2800146/. https://doi.org/10.1093/schbul/sbp118

Murray, R. M., Bhavsar, V., Tripoli, G., & Howes, O. (2017). 30 years on: How the neurodevelopmental hypothesis of schizophrenia morphed into the developmental risk factor model of psychosis. *Schizophrenia Bulletin, 43*(6), 1190–1196. Retrieved from https://doi.org/10.1093/schbul/sbx121

Murray, R. M., & Lewis, S. W. (1988). Is schizophrenia a neurodevelopmental disorder? *British Medical Journal (Clinical Research Ed.), 296*(6614), 63.

National Collaborating Centre for Mental, H. (2018). *Bipolar disorder: The NICE guideline on the assessment and management of bipolar disorder in adults, children and young people in primary and secondary care*.

National Guideline, A. (2016). National Institute for health and care excellence: Clinical guidelines. In *Mental Health Problems in People with Learning Disabilities: Prevention, Assessment and Management*. London, UK: National Institute for Health and Care Excellence (UK) Copyright (c) National Institute for Health and Care Excellence 2016.

Neijmeijer, L. J., Didden, R., Nijman, H. L. I., & Kroon, H. (2018). Assertive community treatment for people with mild intellectual disability or borderline intellectual functioning and mental health problems or challenging behavior: State of the art and implementation in the Netherlands. *Journal of Policy and Practice in Intellectual Disabilities, 15*(4), 329–342.

Noel, J. (2018). Recognition and treatment of mood dysregulation in adults with intellectual disability. *The Mental Health Clinician, 8*(6), 264–274. Retrieved from https://www.ncbi.nlm.nih.gov/pubmed/30397568. Retrieved from https://www.ncbi.nlm.nih.gov/pmc/PMC6213889/. https://doi.org/10.9740/mhc.2018.11.264

Oathamshaw, S. C., & Haddock, G. (2006). Do people with intellectual disabilities and psychosis have the cognitive skills required to undertake cognitive behavioural therapy? *Journal of Applied Research in Intellectual Disabilities, 19*(1), 35–46. Retrieved from https://doi.org/10.1111/j.1468-3148.2005.00284.x

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. *British Medical Journal Open, 6*(4), e010505.

Okolie, K., Perampalam, S., Barker, A., & Nolan, C. J. (2017). A case of Klinefelter syndrome with hypersexual desire. *Endocrinology, Diabetes & Metabolism Case Reports, 2017*, 17-0082. Retrieved from https://www.ncbi.nlm.nih.gov/pubmed/28883919. Retrieved from https://www.ncbi.nlm.nih.gov/pmc/PMC5581370/. https://doi.org/10.1530/EDM-17-0082

Oldham, M. A. (2018). The probability that catatonia in the hospital has a medical cause and the relative proportions of its causes: A systematic review. *Psychosomatics, 59*(4), 333–340.

Oliver, P. C., Piachaud, J., Tyrer, P., Regan, A., Dack, M., Alexander, R., … Rao, B. (2005). Randomized controlled trial of assertive community treatment in intellectual disability: The TACTILD study. *Journal of Intellectual Disability Research, 49*(7), 507–515. Retrieved from https://doi.org/10.1111/j.1365-2788.2005.00706.x

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. Retrieved from https://doi.org/10.1111/jir.12285

Overall, J. E., & Gorham, D. R. (1962). The brief psychiatric rating scale. *Psychological Reports, 10*(3), 799–812. Retrieved from https://doi.org/10.2466/pr0.1962.10.3.799

Owen, M. J., & O'Donovan, M. C. (2017). Schizophrenia and the neurodevelopmental continuum: Evidence from genomics. *World Psychiatry, 16*(3), 227–235. Retrieved from https://doi.org/10.1002/wps.20440

Palumbo, M. L., & McDougle, C. J. (2018). Pharmacotherapy of Down syndrome. *Expert Opinion in Pharmacotherapy*. https://doi.org/10.1080/14656566.2018.1529167

Park, J., Tan, J., Krzeminski, S., Hazeghazam, M., Bandlamuri, M., & Carlson, R. W. (2017). Malignant catatonia warrants early psychiatric-critical care collaborative management: Two cases and literature review. *Case reports in Critical Care, 2017*.

Paton, C., Bhatti, S., Purandare, K., Roy, A., & Barnes, T. (2016). Quality of prescribing of antipsychotic medication for people with intellectual disability under the care of UK mental health services: a cross-sectional audit of clinical practice. *British Medical Journal open, 6*(12), e013116. Retrieved from https://www.ncbi.nlm.nih.gov/pubmed/27920085. Retrieved from https://www.ncbi.nlm.nih.gov/pmc/PMC5168692/. https://doi.org/10.1136/bmjopen-2016-013116

Patterson, M. C., Hendriksz, C. J., Walterfang, M., Sedel, F., Vanier, M. T., & Wijburg, F. (2012). Recommendations for the diagnosis and management of Niemann–Pick disease type C: An update. *Molecular Genetics and Metabolism, 106*(3), 330–344. Retrieved from http://www.sciencedirect.com/science/article/pii/S1096719212001138. https://doi.org/10.1016/j.ymgme.2012.03.012

Patti, P., Andiloro, N., & Gavin, M. (2008). *Parent/carer ratings of self-talk behaviour in children and adults with Down syndrome in Canada and the United Kingdom*.

Pelzer, A. C. M., van der Heijden, F. M. M. A., & den Boer, E. (2018). Systematic review of catatonia treatment. *Neuropsychiatric Disease and Treatment, 14*, 317.

Phelan, E. A., Mahoney, J. E., Voit, J. C., & Stevens, J. A. (2015). Assessment and management of fall risk in primary care settings. *The Medical Clinics of North America, 99*(2), 281–293. Retrieved from https://www.ncbi.nlm.nih.gov/pubmed/25700584. Retrieved from https://www.ncbi.nlm.nih.gov/pmc/PMC4707663/. https://doi.org/10.1016/j.mcna.2014.11.004

Pierce, M. J., LaFranchi, S. H., & Pinter, J. D. (2017). Characterization of thyroid abnormalities in a large cohort of children with Down syndrome. *Hormone Research in Paediatrics, 87*(3), 170–178. Retrieved from https://www.ncbi.nlm.nih.gov/pubmed/28259872. Retrieved from https://www.ncbi.nlm.nih.gov/pmc/PMC5483988/. https://doi.org/10.1159/000457952

Pitschel-Walz, G., Bäuml, J., Froböse, T., Gsottschneider, A., & Jahn, T. (2009). Do individuals with schizophrenia and a borderline intellectual disability benefit from psychoeducational groups? *Journal of Intellectual Disabilities, 13*(4), 305–320. Retrieved from https://doi.org/10.1177/1744629509353237

Prakash, J., Andrews, T., & Porter, I. (2007). Service innovation: Assertive outreach teams for adults with learning disability. *Psychiatric Bulletin, 31*(4), 138–141.

Prosser, H., Moss, S., Costello, H., Simpson, N., Patel, P., & Rowe, S. (1998). 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. Retrieved from https://doi.org/10.1046/j.1365-2788.1998.00146.x

Raftery, M., Burke, K., Murray, N., O'Duinn, O., Murray, I., & Hallahan, B. (2017). An intensive personalised support approach to treating individuals with psychosis and co-morbid mild intellectual disability. *Irish Journal of Psychological*

Medicine, 34(2), 99–109. Retrieved from https://www.cambridge.org/core/article/an-intensive-personalised-support-approach-to-treating-individuals-with-psychosis-and-comorbid-mild-intellectual-disability/1BD7C8EACB6BB94723C48-6E12EB902C9. https://doi.org/10.1017/ipm.2016.19

Ramerman, L., Hoekstra, P. J., & de Kuijper, G. (2018a). Changes in health-related quality of life in people with intellectual disabilities who discontinue long-term used antipsychotic drugs for challenging behaviors. *The Journal of Clinical Pharmacology*. Retrieved from https://doi.org/10.1002/jcph.1311

Ramerman, L., Hoekstra, P. J., & de Kuijper, G. (2018b). 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. Retrieved from http://www.sciencedirect.com/science/article/pii/S0891422218300386. https://doi.org/10.1016/j.ridd.2018.02.011

Reddy, L. F., Horan, W. P., Jahshan, C., & Green, M. F. (2014). Cognitive remediation for schizophrenia: A review of recent findings. *Current Treatment Options in Psychiatry, 1*(2), 121–133. Retrieved from https://doi.org/10.1007/s40501-014-0011-8

Reinares, M., Bonnín, C. M., Hidalgo-Mazzei, D., Sánchez-Moreno, J., Colom, F., & Vieta, E. (2016). The role of family interventions in bipolar disorder: A systematic review. *Clinical Psychology Review, 43*, 47–57. Retrieved from http://www.sciencedirect.com/science/article/pii/S027273581530057X. https://doi.org/10.1016/j.cpr.2015.11.010

Rochon, P. A., & Gurwitz, J. H. (2017). The prescribing cascade revisited. *The Lancet, 389*(10081), 1778–1780.

Rohde, C., Hilker, R., Siskind, D., & Nielsen, J. (2018). Real-world effectiveness of clozapine for intellectual disability: Results from a mirror-image and a reverse-mirror-image study. *Journal of Psychopharmacology, 32*(11), 1197–1203. Retrieved from https://doi.org/10.1177/0269881118783322

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. *British Journal of Clinical Psychology, 42*(1), 81–93.

SAMHSA. (2008). *Assertive community treatment (ACT) evidence-based practices (EBP) kit*. Rockville, MD: Substance Abuse and Mental Health Services Association.

Serret, S., Thümmler, S., Dor, E., Vesperini, S., Santos, A., & Askenazy, F. (2015). Lithium as a rescue therapy for regression and catatonia features in two SHANK3 patients with autism spectrum disorder: Case reports. *BioMed Central Psychiatry, 15*(1), 107. Retrieved from https://doi.org/10.1186/s12888-015-0490-1

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 Medicial Journal Open, 7*(8), e017406. Retrieved from http://bmjopen.bmj.com/content/7/8/e017406.abstract. https://doi.org/10.1136/bmjopen-2017-017406

Sheehan, R., Strydom, A., Morant, N., Pappa, E., & Hassiotis, A. (2017). Psychotropic prescribing in people with intellectual disability and challenging behaviour. *British Medical Journal (Online), 358* https://doi.org/10.1136/bmj.j3896 (Published 18 August 2017) Cite this as: BMJ 2017;358:j3896.

Sienaert, P., Dhossche, D. M., Vancampfort, D., De Hert, M., & Gazdag, G. (2014). A clinical review of the treatment of catatonia. *Frontiers in Psychiatry, 5*, 181–181. Retrieved from https://www.ncbi.nlm.nih.gov/pubmed/25538636. Retrieved from https://www.ncbi.nlm.nih.gov/pmc/PMC4260674/. https://doi.org/10.3389/fpsyt.2014.00181

Sienaert, P., Rooseleer, J., & De Fruyt, J. (2011). Measuring catatonia: A systematic review of rating scales. *Journal of Affective Disorders, 135*(1–3), 1–9.

Simpson, G. M., & Angus, J. W. (1970). A rating scale for extrapyramidal side effects. *Acta Psychiatrica Scandinavica, 45*(S212), 11–19. Retrieved from https://doi.org/10.1111/j.1600-0447.1970.tb02066.x

Singh, A. N., Matson, J. L., Hill, B. D., Pella, R. D., Cooper, C. L., & Adkins, A. D. (2010). The use of clozapine among individuals with intellectual disability: A review. *Research in Developmental Disabilities, 31*(6), 1135–1141. Retrieved from http://www.sciencedirect.com/science/article/pii/S0891422210001599. https://doi.org/10.1016/j.ridd.2010.07.003

Singh, T., Walters, J. T. R., Johnstone, M., Curtis, D., Suvisaari, J., Torniainen, M., … Barrett, J. C. (2017). The contribution of rare variants to risk of schizophrenia in individuals with and without intellectual disability. *Nature Genetics, 49*, 1167. Retrieved from https://doi.org/10.1038/ng.3903

Sinha, P., Jnanaprakasan, P. P., & Andrade, C. (2012). Hyperactive sexual desire in Klinefelter syndrome: Treatment with sertraline. *Psychiatry and Clinical Neurosciences, 66*(6), 533–533. Retrieved from https://doi.org/10.1111/j.1440-1819.2012.02375.x

Soler, J., Fañanás, L., Parellada, M., Krebs, M.-O., Rouleau, G. A., & Fatjó-Vilas, M. (2018). Genetic variability in scaffolding proteins and risk for schizophrenia and autism-spectrum disorders: A systematic review. *Journal of Psychiatry & Neuroscience: JPN, 43*(4), 170066–170066.

Solmi, M., Pigato, G., Kane, J. M., & Correll, C. U. (2018). Clinical risk factors for the development of tardive dyskinesia. *Journal of the Neurological Sciences, 389*, 21–27. Retrieved from http://www.sciencedirect.com/science/article/pii/S0022510X18300704. https://doi.org/10.1016/j.jns.2018.02.012

Solmi, M., Pigato, G. G., Roiter, B., Guaglianone, A., Martini, L., Fornaro, M., … Correll, C. U. (2017). Prevalence of catatonia and its moderators in clinical samples: Results from a meta-analysis and meta-regression analysis. *Schizophrenia Bulletin, 44*(5),

1133–1150. Retrieved from https://doi.org/10.1093/schbul/sbx157

Sprague, R. L., Kalachnik, J. E., & Slaw, K. M. (1989). Psychometric properties of the dyskinesia identification system: Condensed user scale (DISCUS). *Mental Retardation, 27*(3), 141–148.

Stafford, M. R., Jackson, H., Mayo-Wilson, E., Morrison, A. P., & Kendall, T. (2013). Early interventions to prevent psychosis: Systematic review and meta-analysis. *British Medical Journal, 346*, f185. Retrieved from http://www.bmj.com/content/346/bmj.f185.abstract. https://doi.org/10.1136/bmj.f185

Stocks, S. J., Kontopantelis, E., Webb, R. T., Avery, A. J., Burns, A., & Ashcroft, D. M. (2017). Antipsychotic prescribing to patients diagnosed with dementia without a diagnosis of psychosis in the context of National Guidance and drug safety warnings: Longitudinal study in UK general practice. *Drug Safety, 40*(8), 679–692. Retrieved from https://doi.org/10.1007/s40264-017-0538-x

Stomski, N. J., Morrison, P., & Meyer, A. (2015). Antipsychotic medication side effect assessment tools: A systematic review. *Australian & New Zealand Journal of Psychiatry, 50*(5), 399–409. Retrieved from https://doi.org/10.1177/0004867415608244

Sturmey, P., Laud, R. B., Cooper, C. L., Matson, J. L., & Fodstad, J. C. (2010). Mania and behavioral equivalents: A preliminary study. *Research in Developmental Disabilities, 31*(5), 1008–1014. Retrieved from http://www.sciencedirect.com/science/article/pii/S089142221000096X. https://doi.org/10.1016/j.ridd.2010.04.017

Surley, L., & Dagnan, D. (2018). A review of the frequency and nature of adaptations to cognitive behavioural therapy for adults with intellectual disabilities. Journal of Applied Research in Intellectual Disabilities, *0*(0). Retrieved from https://doi.org/10.1111/jar.12534

Tangamornsuksan, W., Chaiyakunapruk, N., Somkrua, R., Lohitnavy, M., & Tassaneeyakul, W. (2013). Relationship between the hla-b∗1502 allele and carbamazepine-induced Stevens-Johnson syndrome and toxic epidermal necrolysis: A systematic review and meta-analysis. *Journal of the American Medical Association Dermatology, 149*(9), 1025–1032. Retrieved from https://doi.org/10.1001/jamadermatol.2013.4114

Test, M. A., & Stein, L. I. (1976). Practical guidelines for the community treatment of markedly impaired patients. *Community Mental Health Journal, 12*(1), 72–82.

Thomas, P., Rascle, C., Mastain, B., Maron, M., & Vaiva, G. (1997). Test for catatonia with zolpidem. *The Lancet, 349*(9053), 702.

Thygesen, J. H., Wolfe, K., McQuillin, A., Viñas-Jornet, M., Baena, N., Brison, N., ... Vogels, A. (2018). Neurodevelopmental risk copy number variants in adults with intellectual disabilities and comorbid psychiatric disorders. *The British Journal of Psychiatry, 212*(5), 287–294. Retrieved from https://www.cambridge.org/core/article/neurodevelopmental-risk-copy-number-variants-in-adults-with-intellectual-disabilities-and-comorbid-psychiatric-disorders/8AECCD39579852888E92030579A4BADC. https://doi.org/10.1192/bjp.2017.65

Tomlinson, M. F., & Wright, J. D. (2018). Identifying the "therapy targets" for treating the negative symptoms of psychosis using cognitive behavioral therapy. *Journal of Cognitive Psychotherapy, 32*(3), 203–220.

Tornese, G., Pellegrin, M. C., Barbi, E., & Ventura, A. (2019). Pediatric endocrinology through syndromes. *European Journal of Medical Genetics*. Retrieved from http://www.sciencedirect.com/science/article/pii/S1769721218303161. https://doi.org/10.1016/j.ejmg.2019.01.004

Trojak, B., Meille, V., Bonin, B., & Chauvet-Geliner, J.-C. (2014). Repetitive transcranial magnetic stimulation for the treatment of catatonia: An alternative treatment to electroconvulsive therapy? *The Journal of Neuropsychiatry and Clinical Neurosciences, 26*(2), E42–E43. Retrieved from https://doi.org/10.1176/appi.neuropsych.13050102

Trollor, J. N., Salomon, C., & Franklin, C. (2016). Prescribing psychotropic drugs to adults with an intellectual disability. *Austrailian Prescriber, 39*(4), 126–130. Retrieved from https://www.ncbi.nlm.nih.gov/pubmed/27756975. https://doi.org/10.18773/austprescr.2016.048

Turner, D. T., van der Gaag, M., Karyotaki, E., & Cuijpers, P. (2014). Psychological interventions for psychosis: A meta-analysis of comparative outcome studies. *American Journal of Psychiatry, 171*(5), 523–538.

Tyrer, P., Oliver-Africano, P. C., Ahmed, Z., Bouras, N., Cooray, S., Deb, S., … Reece, B. (2008). Risperidone, haloperidol, and placebo in the treatment of aggressive challenging behaviour in patients with intellectual disability: A randomised controlled trial. *The Lancet, 371*(9606), 57–63.

van der Gaag, M., van den Berg, D., & Ising, H. (2019). CBT in the prevention of psychosis and other severe mental disorders in patients with an at risk mental state: A review and proposed next steps. *Schizophrenia Research*, 203, 88–93.

van Strien, A. M., Keijsers, C. J. P. W., Derijks, H. J., & van Marum, R. J. (2015). Rating scales to measure side effects of antipsychotic medication: A systematic review. *Journal of Psychopharmacology, 29*(8), 857–866. Retrieved from https://doi.org/10.1177/0269881115593893

Vereenooghe, L., Flynn, S., Hastings, R. P., Adams, D., Chauhan, U., Cooper, S.-A., … Waite, J. (2018). Interventions for mental health problems in children and adults with severe intellectual disabilities: A systematic review. *British Medical Journal Open, 8*(6), e021911. Retrieved from http://bmjopen.bmj.com/content/8/6/e021911.abstract. https://doi.org/10.1136/bmjopen-2018-021911

Weiden, P. J., & Miller, A. L. (2001). Which side effects really matter? Screening for common and distressing side effects of antipsychotic medications. *Journal of Psychiatric Practice®, 7*(1), 41–47.

Weinberger, D. R. (2017). Future of days past: Neurodevelopment and schizophrenia. *Schizophrenia Bulletin, 43*(6), 1164–1168. Retrieved from https://doi.org/10.1093/schbul/sbx118

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. Retrieved from https://doi.org/10.1007/s10862-011-9224-y

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.

Wu, C.-S., Wang, S.-C., Yeh, I. J., & Liu, S.-K. (2016). Comparative risk of seizure with use of first-and second-generation antipsychotics in patients with schizophrenia and mood disorders. *The Journal of Clinical Psychiatry, 77*(5), e573–e579.

Wykes, T. (2018). Cognitive remediation–where are we now and what should we do next. *Journal of Psychopathology, 24*, 57–61.

Wykes, T., Huddy, V., Cellard, C., McGurk, S. R., & Czobor, P. (2011). A meta-analysis of cognitive remediation for schizophrenia: Methodology and effect sizes. *American Journal of Psychiatry, 168*(5), 472–485. https://doi.org/10.1176/appi.ajp.2010.10060855

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, 29*(4), 353–362. Retrieved from https://doi.org/10.1177/0269881114562090

Yusufi, B., Mukherjee, S., Aitchison, K., Dunn, G., Page, E., & Barnes, T. (2005). Reliability of the antipsychotic non-neurological side effects rating scale (ANNSERS). *Journal of Psychopharmacology, 19*(5), A10.

Zisselman, M. H., & Jaffe, R. L. (2010). ECT in the treatment of a patient with catatonia: Consent and complications. *American Journal of Psychiatry, 167*(2), 127–132. Retrieved from https://doi.org/10.1176/appi.ajp.2009.09050703

Treatment of Autism Spectrum Disorders in Dual Diagnosis

30

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Autism spectrum disorder (ASD) is a pervasive developmental disorder that is characterized by deficits in social communication and social interaction, in addition to restrictive or repetitive patterns of behavior, interests, and activities (American Psychiatric Association, 2013). ASD is a heterogeneous disorder; symptom severity, language abilities, and intellectual functioning vary greatly across diagnosed individuals. The Centers for Disease Control and Prevention (CDC) has recently estimated that 1 in 59 children in the USA is diagnosed with ASD (Baio et al., 2018). The CDC further reported findings that 31% of children with ASD were found to have IQ scores that may indicate intellectual disability (ID; 70 or below), 25% had borderline IQ scores (71–85), and 44% had average to above average IQ scores (above 85; Baio et al., 2018).

ID is known to co-occur with ASD (Lecavalier, Snow, & Norris, 2011; Matson & Shoemaker, 2009). As such, the latest edition of the *Diagnostic and Statistical Manual of Mental Disorders* (DSM-5) allows for the indication of accompanying intellectual impairment within ASD diagnoses (American Psychiatric Association, 2013). ID is characterized by impairments of adaptive functioning across three domains: conceptual (e.g., language, reading, writing, math, reasoning, knowledge, and memory skills), social (e.g., empathy, social judgment, interpersonal communication, and friendship skills), and practical (e.g., self-management in personal care, job responsibilities, money management, recreation, and organizing school and work tasks; American Psychiatric Association, 2013). ID is defined as approximately 2 standard deviations below the population average (i.e., an IQ score of about 70 or below), and severity is based on adaptive functioning.

Intellectual functioning has been found to affect the presentation of ASD symptoms. Lower IQ scores have been associated with greater ASD symptom severity (Ben-Itzchak, Lahat, Burgin, & Zachor, 2008) and higher rates of restricted or repetitive patterns of behaviors (Bishop, Richler, & Lord, 2006). IQ has also been found to be predictive of adaptive skills in individuals with ASD (Kanne et al., 2011). Given the different clinical presentation of ASD with co-occurring ID, it has been suggested that measurements of intellectual functioning may be useful in identifying subtypes of ASD (Lecavalier et al., 2011; Nowell, Goin-Kochel, McQuillin, & Mire, 2017), which may have implications on treatment needs and prognosis.

There are many treatments available for individuals with ASD; however, these treatments do not have the same level of empirical support (Vismara & Rogers, 2010). This chapter provides an overview of evidence-based treatments for

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individuals with ASD. While ASD treatment research includes participants with a wide range of intellectual functioning as demonstrated in pretreatment assessments (Reichow, 2012), very few studies report on co-occurring diagnoses of ID in their samples of participants. Though limited, existing research on the treatment of individuals with both ASD and ID diagnoses is also discussed.

Evidence-Based Treatments

Treatments for individuals with ASD can be categorized as either comprehensive or focused (Wong et al., 2013). Comprehensive treatment models target the core features of ASD as well as other developmental domains and are generally conducted over a long period of time (e.g., one or more years). Focused treatments are interventions that target a single skill or behavior and may be carried out over a shorter period of time (e.g., weeks or months). Focused treatments are often used in conjunction with other focused treatments and are frequently the components that make up comprehensive treatment models (Wong et al., 2013).

The treatments covered in this chapter were selected from existing systematic reviews of comprehensive (i.e., Maglione, Gans, Das, Timbie, & Kasari, 2012; Rogers & Vismara, 2008; Smith & Iadarola, 2015; Warren et al., 2011) and focused treatment literature (i.e., National Autism Center, 2015; Odom, Collet-Klingenberg, Rogers, & Hatton, 2010; Wong et al., 2013). While not exhaustive, this chapter covers treatments with the most rigorous empirical support.

Comprehensive Treatments

To identify comprehensive treatments that warranted inclusion in this chapter, the authors referred to systematic reviews on comprehensive treatments. While many systematic reviews focus on a single treatment philosophy (see Reichow, Hume, Barton, & Boyd, 2018 for a review of applied behavior analysis; see Ryberg, 2015 for a review on the Early Start Denver Model; see Verschuur, Didden, Lang, Sigafoos, & Huskens, 2014 for a review of pivotal response treatment), four articles were found to review multiple treatments independently (Maglione et al., 2012; Rogers & Vismara, 2008; Smith & Iadarola, 2015; Warren et al., 2011). Due to differing criteria used for study inclusion and strength of evidence, conclusions from these reviews vary. See Table 30.1 for a summary of these systematic reviews.

Four interventions emerged that met minimum inclusion criteria but varied on levels of

Table 30.1 Summary of comprehensive treatment systematic reviews

	Strength of evidence				
Article	ABA	ESDM	LEAP	PRT	TEACCH
Maglione et al. (2012)	Moderate	Moderate	–	–	Low
Rogers and Vismara (2008)	Well-established	–	–	Possibly efficacious	–
Smith and Iadarola (2015)	Well-established	Possibly efficacious	Possibly efficacious	Possibly efficacious for spoken communication (focused)	Experimental
Warren et al. (2011)[a]	Low	Insufficient	–	–	Insufficient

Note: ABA applied behavior analysis, *ESDM* Early Start Denver Model, *LEAP* Learning Experiences: An Alternative Program for Preschoolers and Parents, *PRT* pivotal response training, *TEACCH* Treatment and Education of Autistic and Communication-Handicapped Children

[a]A follow-up review conducted by Weitlauf et al. (2014) found substantially more studies of greater quality and determined early intensive behavioral and developmental intervention, which included ABA, ESDM, and LEAP, to have moderate strength of evidence for cognitive and language outcomes

strength of evidence: applied behavior analysis (ABA), pivotal response treatment (PRT), Learning Experiences: An Alternative Program for Preschoolers and Parents (LEAP), and the Early Start Denver Model (ESDM). Rogers and Vismara (2008) and a more recent follow-up review conducted by Smith and Iadarola (2015) found ABA to be well-established for the treatment of ASD. Rogers and Vismara (2008) found PRT to be a possibly efficacious comprehensive treatment, while Smith and Iadarola (2015) found it to be a possibly efficacious focused intervention for spoken communication. In addition, Smith and Iadarola (2015) found ESDM and LEAP to be possibly efficacious comprehensive interventions. While the Agency for Healthcare Research and Quality (AHRQ) found ABA to have low strength of evidence and ESDM to have insufficient evidence at the time of their review (Warren et al., 2011), a follow-up review conducted by the AHRQ identified substantially more studies of greater quality and they determined early intensive behavioral and developmental intervention (which included ABA, ESDM, and LEAP) to have moderate strength of evidence for cognitive and language outcomes (Weitlauf et al., 2014). Finally, Maglione et al. (2012) found both ABA and ESDM to have moderate strength of evidence. The treatment approach referred to as Treatment and Education of Autistic and Communication-Handicapped Children (TEACCH) was evaluated in a number of these systematic reviews; however, due to the insufficient strength of evidence found across the reviews (Maglione et al., 2012; Smith & Iadarola, 2015; Warren et al., 2011), it is not discussed further in the current chapter. ABA, PRT, LEAP, and ESDM are each discussed in turn.

Applied Behavior Analysis ABA-based treatment involves the application of principles and procedures of learning and motivation in order to alter behaviors of social significance (Cooper, Heron, & Heward, 2007; Granpeesheh, Tarbox, & Dixon, 2009). Treatment is typically delivered at a high intensity (e.g., 25–40 hr per week) over several years (Eldevik et al., 2009; Reichow, Barton, Boyd, & Hume, 2012). While ABA-based treatment is generally initiated in early development, treatment may be beneficial into a patient's adult years (Ivy & Schreck, 2016). Services may be conducted in the patient's home, school, clinic, community, or a combination of settings.

ABA-based treatment for ASD is recommended by the American Academy of Pediatrics (Myers & Johnson, 2007) and the US Surgeon General (U.S. Department of Health and Human Services, 1999). There are various ABA-based models available. While grounded on the same fundamental principles of learning and motivation, curricula, treatment intensity, patient age range, and treatment settings vary across different ABA-based treatment models (see Odom, Boyd, Hall, & Hume, 2010 for a review of different manualized comprehensive treatment models). The UCLA Young Autism Program (also known as the Lovaas model; Lovaas, 1987) is likely the most well-recognized ABA-based treatment model.

Comprehensive ABA-based treatment models focus on promoting skill acquisition across all developmental areas in which an individual displays deficits (e.g., language, academic skills, social skills, play skills, motor skills, adaptive skills, executive functions, cognition, etc.) and reducing challenging behaviors (e.g., restricted, repetitive behaviors; self-injury; etc.). Treatment programs are individualized to meet the specific needs of each patient and typically rely on the following components to alter behavior: (a) assessment of skill deficits and the function of challenging behaviors; (b) identification of treatment objectives; (c) evaluation of initial levels of skill performance and/or frequency of challenging behaviors (i.e., baseline); (d) selection and implementation of interventions that teach socially important skills and/or reduce challenging behaviors; (e) ongoing assessment of skill performance and/or frequency of challenging behaviors to determine the effectiveness of the intervention; and (f) modifications made as needed to maintain or increase the effectiveness of the intervention (Cooper et al., 2007;

Granpeesheh, Tarbox, Najdowski, & Kornack, 2014; Sulzer-Azaroff & Mayer, 1991).

ABA-based treatment is overseen by a clinical supervisor and delivered by a behavior technician. Supervisors are typically master's or doctoral-level licensed and/or certified clinicians (e.g., Board Certified Behavior Analyst, psychologist, marriage and family therapists, etc.). Supervisors have a variety of responsibilities, including assessing skill deficits, developing treatment plans, tracking progress, adjusting treatment goals, and providing training to caregivers to promote continuity of treatment strategies across settings. Supervisors also oversee behavior technicians, who are responsible for providing direct services to patients with ASD (Behavior Analyst Certification Board, 2014).

The first controlled trial evaluating ABA treatment for children with ASD was conducted by Lovaas (1987). Participants included 19 children receiving high-intensity treatment (i.e., 40 hr. per week) and 19 children receiving low-intensity treatment (i.e., 10 hr. per week). Findings revealed that 47% of children receiving high-intensity treatment achieved average-level IQ scores and were succeeding in mainstream education classrooms without additional support as compared to only 2% of children receiving low-intensity treatment. These findings that a large portion of children receiving high-intensity ABA achieve optimal outcomes have been replicated by other researchers (Cohen, Amerine-Dickens, & Smith, 2006; Howard, Sparkman, Cohen, Green, & Stanislaw, 2005; Sallows & Graupner, 2005). Furthermore, ABA-based treatment for individuals with ASD has been shown to have long-lasting effects across outcome measures of adaptive, language, social, and intellectual functioning (Magiati, Moss, Charman, & Howlin, 2011; McEachin, Smith, & Lovaas, 1993; Sallows & Graupner, 2005; Smith, Groen, & Wynn, 2000), as well as placement in mainstream education (Harris & Handleman, 2000).

A number of meta-analyses have been conducted on existing ABA-based treatment research (Eldevik et al., 2009; Makrygianni & Reed, 2010; Peters-Scheffer, Didden, Korzilius, & Sturmey, 2011; Reichow, 2012; Reichow et al., 2018; Reichow & Wolery, 2009; Virués-Ortega, 2010). Results of these meta-analyses revealed significant improvements in intellectual functioning, language, adaptive skills, social skills, and challenging behaviors; however, there is criticism with respect to the quality of evidence. Specifically, research studies have been criticized for small sample sizes and less than optimal study designs (Reichow et al., 2018).

In spite of these limitations, there is a strong consensus that ABA-based treatments for ASD are effective; however, research also reveals variance in individual treatment response (Eldevik et al., 2010; Howlin, Magiati, & Charman, 2009). Higher treatment intensity has been linked to greater treatment gains (Eldevik et al., 2010; Granpeesheh, Dixon, Tarbox, Kaplan, & Wilke, 2009;Linstead et al., 2016; Linstead et al., 2017; Makrygianni & Reed, 2010). For example, Linstead et al. (2016) demonstrated that the single variable of treatment intensity accounted for 60% of the variance in mastered learning objectives. Longer treatment duration (Linstead et al., 2016; Linstead et al., 2017; Makrygianni & Reed, 2010) and greater total intervention time (Virués-Ortega, 2010; Virués-Ortega, Rodríguez, & Yu, 2013) have also been linked to greater treatment gains. In addition to treatment-related variables, patient-related variables including younger age (Ben-Itzchak & Zachor, 2011; Eldevik, Hastings, Jahr, & Hughes, 2012; Flanagan, Perry, & Freeman, 2012; Granpeesheh, Dixon, et al., 2009; Makrygianni & Reed, 2010; Perry et al., 2011; Virués-Ortega et al., 2013), lower severity of ASD symptoms (Ben-Itzchak & Zachor, 2011; Eldevik et al., 2012; Perry et al., 2011; Remington et al., 2007), and greater intellectual functioning (Ben-Itzchak & Zachor, 2007; Eikeseth, Smith, Jahr, & Eldevik, 2002, 2007; Eldevik et al., 2010; Eldevik et al., 2012; Hayward, Eikeseth, Gale, & Morgan, 2009; Magiati et al., 2011; Magiati, Charman, & Howlin, 2007; Perry et al., 2011; Remington et al., 2007) have been associated with superior outcomes.

ABA-Based Treatment for ASD and Co-occurring ID Although ID commonly co-occurs with ASD, the impact of ABA-based

treatment on the outcomes of individuals with both diagnoses is not well researched. Only the latest edition of the DSM (DSM-5; American Psychiatric Association, 2013) addresses the differential diagnosis of ASD and ID within the ASD classification, which may explain why ASD treatment research does not often report on comorbid diagnoses in its samples of participants. Nevertheless, ABA treatment research has included participants with a range of intellectual functioning, which is commonly assessed pre- and posttreatment as an outcome measure. Although severity of ID is based on adaptive functioning and not merely IQ scores (American Psychiatric Association, 2013), IQ scores do provide some indication of the level of intellectual functioning of participants included in ABA research. The majority of ASD treatment research has included samples with mild (IQ scores between 55 and 70) to moderate (IQ scores between 40 and 55) deficits in intellectual functioning (see Reichow, 2012 for an overview of meta-analyses). For individuals with comorbid ASD and severe to profound intellectual impairments, ABA-based treatment research is limited.

While ABA treatment research has included participants with a range of intellectual functioning, very few studies report on the effectiveness of ABA treatment for individuals with both a diagnosis of ASD and ID. The limited studies that have included participants with both ASD and ID diagnoses also tended to have fewer treatment hours per week than other evaluations of ABA-based treatment for ASD (e.g., Cohen et al., 2006; Howard et al., 2005; Lovaas, 1987; Sallows & Graupner, 2005). Smith, Eikeseth, Klevstrand, and Lovaas (1997) evaluated outcomes of 11 children with ASD and severe ID diagnoses who were receiving intensive ABA-based treatment (i.e., 30 hr. per week) as compared to 10 children with ASD and severe ID who were receiving low-intensity treatment (i.e., 10 hr. or less). The high-intensity treatment group participants were found to make greater gains in IQ and expressive language as compared to participants in the low-intensity treatment group. While meaningful gains were made by the high-intensity treatment group, these participants were still found to have substantial deficits. Additional studies evaluating low-intensity ABA-based treatment for children with ASD and mild to severe ID have been conducted (Eldevik, Eikeseth, Jahr, & Smith, 2006; Peters-Scheffer, Didden, Mulders, & Korzilius, 2010). In comparison to control groups, participants receiving 12 hr. per week of ABA-based treatment were found to make modest but significant gains in intellectual functioning, receptive and expressive language, communication, and behavior (Eldevik et al., 2006), and participants receiving 6.5 hr. per week of treatment were shown to make gains in developmental age and adaptive skills (Peters-Scheffer et al., 2010). Across these studies, outcomes were not found to be as optimal as those reported in other studies (e.g., Cohen et al., 2006; Howard et al., 2005; Lovaas, 1987; Sallows & Graupner, 2005). It is difficult to isolate the cause of the poorer outcomes; that is, whether it is due to the presence of ID or due to participants not receiving intensive treatment. Despite outcomes not being as strong, participants were still found to make significant improvements.

While it has been demonstrated that individuals with ASD and co-occurring ID make significant progress in ABA-based treatments, the presence of intellectual disability likely moderates the impact of the treatment. Higher IQ has been linked to greater treatment outcomes, particularly for measures of language, adaptive skills, and intellectual functioning (Ben-Itzchak & Zachor, 2007; Eikeseth et al., 2002, 2007; Eldevik et al., 2010; Eldevik et al., 2012; Harris & Handleman, 2000; Hayward et al., 2009; Magiati et al., 2007; Magiati et al., 2011; Perry et al., 2011; Remington et al., 2007).

Pivotal Response Treatment PRT (Koegel & Koegel, 2019) is a treatment approach for individuals with ASD that is derived from ABA and involves targeting pivotal areas, including motivation to engage in social interactions, social initiations, and self-regulation of behavior. The aim of addressing these pivotal areas is to lead to improvements beyond the originally targeted skills (Koegel & Frea, 1993; Koegel, Koegel,

Shoshan, & Mcnerney, 1999). Another objective of PRT is to increase opportunities to learn in the natural environment (e.g., home, classroom, workplace). Caregiver participation and caregiver training are large components of PRT. The involvement of family members, peers, and teachers in treatment is also emphasized. PRT may be applied across developmental domains, including academic, behavioral, and social-communicative skills, and may be provided from early development through adulthood.

Some debate has occurred over whether PRT should be considered a comprehensive or focused treatment (Smith & Iadarola, 2015). Given that the founders of PRT specify that the intervention is comprehensive (Koegel & Koegel, 2019), the authors of the current chapter have classified it as such; however, there is debate as to whether there is enough empirical evidence to support PRT across a broad range of developmental outcomes. Since group studies on PRT have primarily centered on spoken communication outcomes, Smith and Iadarola (2015) classified the intervention as focused. Although PRT is covered in the comprehensive treatment section of this chapter, it should be noted that it has been identified as an evidence-based focused intervention for children with ASD (National Autism Center, 2015; Odom, Collet-Klingenberg, et al., 2010; Wong et al., 2013).

PRT has been evaluated in randomized, controlled trials as a focused intervention for spoken communication skills. Schreibman and Stahmer (2014) conducted a randomized, controlled trial that compared PRT to the Picture Exchange Communication System (PECS). Participants were found to make significant improvements in spoken language outcomes with no differences detected between groups. Mohammadzaheri, Koegel, Rezaee, and Rafiee (2014) conducted a randomized, controlled trial that evaluated the effectiveness of PRT as compared to structured ABA (e.g., discrete trial training) that did not include child choice (Koegel, O'Dell, & Koegel, 1987). PRT was found to have benefits over structured ABA in fostering communication skills. It should be noted that research comparing outcomes of comprehensive PRT and ABA-based treatment across a wide range of developmental domains has not yet been conducted.

Many research studies have evaluated the effectiveness of PRT. Verschuur et al. (2014) conducted a systematic review of research evaluating PRT for children with ASD. A total of 43 studies were included in the review. The majority of studies were found to have methodological concerns and were classified as providing suggestive evidence. From the studies classified as providing conclusive or preponderant evidence, PRT was found to produce gains in self-initiations, communication, language, and play skills, as well as reductions in challenging behaviors (Verschuur et al., 2014). More recently, a meta-analysis on single-subject PRT research was conducted by Bozkus-Genc and Yucesoy-Ozkan (2016). The findings of 34 single-subject studies were analyzed, and PRT was found to be fairly effective in teaching skills to children with ASD.

Learning Experiences: An Alternative Program for Preschoolers and Parents LEAP (Strain & Bovey, 2011; Strain & Hoyson, 2000) is an integrated preschool program for children with ASD that focuses on maximizing learning opportunities in the natural environment. Peers with typical development (TD) play an important role in the intervention. Peers with TD receive social skills training to help facilitate social interaction and social communication with their peers with ASD. In addition to peer-mediated intervention, caregiver training on behavior teaching strategies is a significant component of LEAP. As with the other comprehensive treatments, the curriculum is individualized to the student's needs, treatment decisions are data driven, and generalization of skills is emphasized. LEAP incorporates behavioral strategies, including errorless learning, time delay, incidental teaching, PRT, PECS, and positive behavior support.

LEAP has been evaluated in a randomized, controlled trial conducted by Strain and Bovey (2011). Participants were 288 students from 56 preschool classrooms. Classrooms were randomly assigned to either an experimental group

that received 2 years of LEAP training and coaching or a control group that received intervention manuals only. Posttreatment assessments revealed that the experimental group made significantly stronger gains according to measures of cognitive functioning, language, social skills, challenging behaviors, and symptoms of ASD. More recently, Boyd et al. (2014) conducted a quasi-experimental study that compared the outcomes of 198 preschool students receiving LEAP, TEACCH, or high-quality special education that did not follow a specific model. Participants were found to improve over time; however, no differences were detected between groups.

Early Start Denver Model ESDM (Rogers & Dawson, 2010) is a treatment model for children with ASD that combines developmental, relationship, and ABA-based approaches. Treatment is designed to be initiated between 1 and 3 years old and continue until 4–5 years old. Treatment may be delivered across a variety of settings, including center, preschool, and home. ESDM is delivered in the natural environment during play and daily routines. Caregiver training and caregiver involvement in treatment are important components. ESDM emphasizes intensive treatment (i.e., 20 hr. or more per week). The goal of treatment is to reduce symptoms of ASD and promote skill acquisition across developmental domains, including receptive communication, expressive communication, joint attention, imitation, social, play, cognitive, fine motor, gross motor, and self-care skills. Treatment is overseen by a professional who specializes in early childhood development (e.g., special education teacher, psychologist, speech and language pathologist, occupational therapist, behavior analyst) and is carried out one-to-one by a trained therapist or caregiver.

ESDM has been evaluated in multiple studies. Dawson et al. (2010) conducted a randomized, controlled trial in which 48 children with ASD were randomly assigned to receive ESDM or eclectic community-based interventions. After 2 years of intervention, participants in the ESDM group showed significantly greater improvements in measures of IQ, language, adaptive behavior, and diagnostic status. Recently, Rogers et al. (2019) conducted a randomized, controlled trial to extend the work of Dawson et al. (2010). Participants included 118 children with ASD, who were randomly assigned to receive either ESDM or community interventions. Participants were enrolled to receive treatment in one of three sites. After 27 months of intervention, participants receiving ESDM were found to make greater improvements in language outcomes than participants receiving community interventions for two of the three sites. No significant differences were detected between groups for the third site. Across all sites, stronger language outcomes were observed for the ESDM group; however, no significant between-group differences were found for developmental quotient, autism severity, or adaptive behavior outcomes. A systematic review of research evaluating the use of ESDM for children with ASD was conducted by Ryberg (2015). Eight articles were found to meet inclusion criteria, in which quality of evidence was found to range from low to high. Overall, ESDM was found to effectively improve cognition, language, and adaptive behavior outcomes in children with ASD (Ryberg, 2015).

Comprehensive Treatment Summary Four comprehensive treatment models were found to have some level of empirical support: ABA, PRT, LEAP, and ESDM. While these treatments vary in a number of ways (e.g., primary interventionist, treatment setting, patient age, curriculum, etc.), there are commonalities across these philosophies. All comprehensive treatment models described in this chapter are derived from or have integrated components of ABA, the most rigorously researched treatment for ASD (National Research Council, 2001; Vismara & Rogers, 2010). Given the overlap in philosophies, these treatments have a number of similarities. Intervention involves the use of behavioral techniques to teach new skills and reduce challenging behaviors. Furthermore, treatment is initiated in early development, and there is an emphasis on high intensity and/or the maximization of learning

opportunities. Treatment outcomes vary across individuals; not all individuals achieve optimal outcomes; however, treatment may be used to address functional skills to improve independence and quality of life. Ultimately, the goal of treatment is for the patient to achieve an average level of functioning across developmental domains.

Focused Interventions

Focused interventions are treatment strategies that target a single skill or behavior. They are often the components that make up comprehensive treatment models, like those discussed in the previous section. Oftentimes, focused interventions are combined during treatment. Interventions may be used in a variety of settings, including the home, clinic, community, and school. Using a given intervention across multiple settings may aid in generalization of skills (Stokes & Baer, 1977), which is a desired outcome of behavior reduction and skill acquisition interventions.

The focused interventions discussed in this chapter were selected from systematic reviews on focused treatments for ASD (National Autism Center, 2015; Odom, Collet-Klingenberg, et al., 2010; Wong et al., 2013). Unlike the comprehensive treatment literature, agreement was strong between the systematic reviews with respect to which focused interventions met evidence-based criteria. Clinical judgment was used to narrow down the interventions covered in this chapter to those that are most applicable to the population of individuals with both ASD and ID. Evidence supporting the use of interventions for individuals with comorbid ID is discussed when available; however, ASD treatment studies do not often specify whether participants have comorbid ID. The interventions discussed in this chapter are broken down into three categories: (1) behavior reduction interventions, (2) skill acquisition interventions, and (3) method of delivery. Each is discussed in turn.

Behavior Reduction Interventions Challenging behaviors are frequently exhibited by individuals with ASD (Jang, Dixon, Tarbox, & Granpeesheh, 2011). Jang et al. (2011) found 94% of their sample of children with ASD engage in some form of challenging behaviors, in which "repeated and unusual vocalizations" was the most commonly reported behavior. Challenging behaviors that are commonly targeted for reduction include self-injurious behavior, aggression, stereotypic behavior, tantrums, and property destruction.

Mere engagement in a challenging behavior does not necessarily mean that the behavior should be targeted for reduction. For example, tantrums may not be targeted for a 2-year-old who engages in the behavior twice a month as this is age appropriate. Additionally, the context of the behavior is extremely important to consider. For example, singing is a behavior that many people engage in at some point in time; however, some individuals engage in singing at an extreme rate and/or in socially inappropriate ways, in which case the behavior may be selected for reduction. When identifying behaviors to target for reduction, there are a number of factors to consider, centering on the behavior's "effects on the person's life" (Didden et al., 2012). Behaviors may be targeted because they interfere with learning, are dangerous to the individual or others, impact the individual's access to the community, and/or have social significance for the individual (Didden et al., 2012; Hurwitz & Minshawi, 2012; Lancioni, Singh, O'Reilly, Sigafoos, & Didden, 2012). After identification, the challenging behavior should be clearly defined in order to aid in communication and data collection (Hurwitz & Minshawi, 2012).

While it may not be readily apparent, challenging behaviors serve a function for the individual; identifying this role, or function, aids in treatment planning. For behavior reduction interventions, it is best practice to begin by identifying the function of the behavior through a functional assessment (Napolitano, Knapp, Speares, McAdam, & Brown, 2012). While terminology differs in the literature, functional assessment is an umbrella term that covers a variety of practices used to identify the function

of a behavior, including indirect assessments, direct assessments, and functional analyses or experimental functional analyses (see Chap. 13 of the current volume for a detailed discussion). Broadly speaking, a functional assessment involves the identification of the antecedents and consequences associated with the behavior to determine a hypothesized function (i.e., reinforcer received following engagement in the behavior; see Matson, 2012 for a detailed description of functional assessment). The use of functional assessments has been validated by a number of studies for individuals with ASD, across a wide range of challenging behaviors (Wong et al., 2013). Evidence suggests that there are four main functions of behavior: access to social reinforcement in the form of attention, access to social reinforcement in the form of tangibles, escape/avoidance of social input/demands, and automatic reinforcement (Lancioni et al., 2012). Following the completion of a functional assessment, intervention strategies are selected based on the hypothesized function of the behavior; research indicates that strategies that are matched to the function of the behavior are more successful than those selected arbitrarily (Napolitano et al., 2012). Whenever targeting a behavior for reduction, a replacement behavior, which serves the same function, should be identified to increase the individual's adaptive skills (Napolitano et al., 2012). Evidence-based strategies for reducing challenging behaviors are discussed below.

Antecedent-Based Interventions Antecedent-based interventions (ABIs) involve manipulations to the environment with the goal of reducing future occurrences of a challenging behavior. There are a number of ABI strategies, and the unifying factor behind these strategies is their timing: interventions are implemented before the challenging behavior occurs. Examples of strategies include providing the individual with choices, priming regarding expectations, using visual schedules, demand fading, and incorporating the individual's interest into activities. ABIs that match the function of the challenging behavior should be selected based on the results of a functional assessment. Effective instruction delivery (EID), errorless compliance training (ECT), and high probability command sequences (HPCS) are ABIs used to address behaviors with an escape function. EID involves gaining eye contact prior to giving a demand, praising eye contact, presenting the demand, and praising compliance (Radley & Dart, 2016). ECT involves starting with instructions to which an individual is likely to comply and gradually moving toward instructions to which the individual is not likely to comply (Radley & Dart, 2016). Similarly, HPCS involve presenting several demands that have a high probability of compliance and then presenting a demand that has a low probability of compliance (Radley & Dart, 2016).

Typically ABIs are used as part of a broader treatment plan with additional focused strategies. For instance, exercise may be used as an ABI to reduce challenging behaviors. Prior to the occurrence of a challenging behavior, the individual is directed to engage in a physical activity (e.g., jogging, walking, jumping jacks, yoga poses, leg stretches, arm curls, crab walks) for a fixed interval, typically at a predetermined time (e.g., before school, after lunch). Empirical support exists for the use of exercise as an ABI for reducing aggression, property destruction, and self-stimulatory behavior in children with ASD; additionally, gains in academic engagement and academic responding were noted when exercise was used before challenging behaviors occurred (Wong et al., 2013).

Research supports the use of ABIs to reduce challenging behaviors and improve social, communication, play, school readiness, academic, motor, and adaptive skills for individuals with ASD, from early development through early adulthood (Wong et al., 2013). The degree to which these findings may be generalized to individuals with both ASD and ID is not clearly documented. While the quantity of studies on ABI for individuals with ID is limited, existing evidence supports the use of several specific ABIs for this population. For example, Chan, Lambdin, Graham, Fragale, and Davis (2014) used a

picture-based activity schedule to teach adults with ID to use an iPad to engage in a leisure activity. Additionally, in a review of literature on ABIs used to promote compliance, Radley and Dart (2016) found support for the use of EID, ECT, and HPCS for individuals with ID.

Differential Reinforcement Differential reinforcement involves reinforcing a specific set of more appropriate behaviors while withholding reinforcement for the targeted challenging behavior (i.e., extinction). Reinforcement, which is discussed in greater detail later in the chapter, is a consequence of a behavior that makes the behavior more likely to occur. Extinction is a strategy used to reduce the occurrence of a challenging behavior by removing the reinforcement that was previously received for the behavior. Prior to implementing extinction, a functional assessment should be conducted to identify the consequences maintaining the challenging behavior, which would ultimately be withheld during an extinction protocol. For instance, if a functional assessment hypothesizes that the function of an individual's aggression is to gain attention, extinction would involve withholding all attention (e.g., reprimand, eye contact) in response to that individual's aggression. Extinction has been used to address a wide range of challenging behaviors, and evidence supports its use for individuals with ASD between the ages of 3 and 18 years (Wong et al., 2013); however, it is recommended that extinction be used in conjunction with reinforcement-based procedures, such as differential reinforcement, to mitigate potential side effects (e.g., extinction burst, increase in response amplitude, spontaneous recovery, aggression) and promote the use of appropriate behaviors (Cooper et al., 2007).

There are several categories of differential reinforcement, defined by the behaviors selected for reinforcement. Differential reinforcement of alternative behavior (DRA) involves the reinforcement of a specific behavior that has been identified as a more appropriate alternative to the challenging behavior. The alternative behavior typically serves the same function as the challenging behavior (as identified via a functional assessment). Examples of alternative behaviors include gaining attention by calling a person's name, requesting a break, and asking for access to a specific item. Functional communication training (FCT) is a specific form of DRA where the alternative behavior is a socially appropriate method of communicating. As with all forms of DRA, the inappropriate behavior is placed on extinction and the alternative method of communicating is reinforced. An example of FCT for an individual who engages in aggression with an attention-seeking function would be to teach the individual to tap the adult to gain his/her attention instead of engaging in aggression. FCT is considered an evidence-based practice for individuals with ASD, ages 3–18 years (Wong et al., 2013). Additionally, a review conducted by Heath, Ganz, Parker, Burke, and Ninci (2015) found FCT to be an evidence-based intervention for individuals with ASD and individuals with ID.

Other forms of differential reinforcement include differential reinforcement of incompatible behavior (DRI) and differential reinforcement of other behavior (DRO). DRI involves reinforcing a specific behavior that cannot occur at the same time as the challenging behavior (e.g., banging a drum instead of engaging in hand flapping). Alternatively to DRA and DRI, which involve identifying a specific behavior to be reinforced, DRO involves reinforcing anything other than the challenging behavior. For DRO, reinforcement is typically provided on a specified time schedule. For instance, reinforcement is provided at the end of a 5-minute time interval if no aggression occurred during that time frame. Differential reinforcement procedures have a strong evidence base for individuals with ASD, ages 3–22 years, across a range of challenging behaviors (Wong et al., 2013). Studies have also demonstrated the efficacy of differential reinforcement procedures with children (Vladescu & Kodak, 2010) and adults (Chowdhury & Benson, 2011) with ID.

Response Interruption and Redirection Response interruption and redirection (RIRD) entails

interrupting an occurrence of a challenging behavior with a demand or a sequence of demands to prompt engagement in a more appropriate response (Martinez & Betz, 2013). This strategy is typically used for vocal stereotypy that is maintained by automatic reinforcement as response blocking is difficult to implement for this topography class. Martinez and Betz (2013) reviewed eight studies that incorporated RIRD as an intervention for stereotypy for children with ASD. While decreases in levels of stereotypy were demonstrated across all the reviewed studies, several procedural variations of RIRD were identified that may impact the success of the intervention. In seven of the studies reviewed, three demands were presented following an occurrence of stereotypy, though one study found the presentation of one demand to be effective. Compliance with the demands without engagement in stereotypy was a requirement for six of the studies. Furthermore, six studies were found to use topographically matched demands, which are related to the type of stereotypy, one study included unmatched demands, and one study included both matched and unmatched demands. For vocal stereotypy, a matched demand would require a vocal response (e.g., asking "What color?"), while an unmatched demand may require a physical response (e.g., presenting the instruction "Clap hands"). While it may be more intuitive to present matched demands, both matched and unmatched demands have been found to be equally effective in reducing rates of the challenging behavior (Martinez & Betz, 2013). Evidence supports the use of RIRD to decrease the occurrence of challenging behaviors, specifically vocal stereotypy, in individuals with ASD from 3 to 22 years of age (Wong et al., 2013).

Self-Management Self-management is the ability to manage one's own behavior without requiring external support. Self-management includes self-identification (i.e., identifying the difference between one's appropriate and inappropriate behavior), self-monitoring (i.e., recording whether one has engaged in an appropriate or inappropriate behavior), self-evaluation (i.e., identifying whether or not one's behavior met a goal and merits reinforcement), and self-reinforcement (i.e., providing oneself with reinforcement for engaging in appropriate behaviors; Granpeesheh et al., 2014). Self-management is typically used to reduce challenging behaviors but can also be used to increase appropriate skills, such as social, communication, academic, play, and vocational skills.

The use of self-management strategies is validated for individuals with ASD from 3 to 22 years of age (Wong et al., 2013). Self-management has also been found to be an effective strategy for individuals with ID. Assistive technologies (Mechling, 2007) and visual modifications (e.g., picture cards, videos, flowcharts, written directions; Carr, Moore, & Anderson, 2014) have been implemented to facilitate self-management in individuals with ID. While verbal prompts are more commonly used to teach self-management, Carr et al. (2014) found the use of visual prompts to be highly effective in producing the target behavior change. In a review of the literature, Smith, Shepley, Alexander, and Ayres (2015) found self-instruction strategies with an emphasis on generalization to aid in the acquisition of multistep tasks for individuals with ID.

Skill Acquisition Interventions In addition to strategies aimed at reducing challenging behaviors, there is a wealth of evidence-based interventions available to increase appropriate behaviors. For organizational purposes, the skill acquisition interventions discussed in this chapter are broken down into three sections: (1) general skill acquisition interventions, (2) communication interventions, and (3) social interventions.

General Skill Acquisition Interventions

Discrete Trial Training Discrete trial training (DTT) is a method of teaching new behaviors in which a skill is broken down into discrete components and each component is taught in a series of trials. Trials are presented in rapid succession

and consist of an instruction, a response, a consequence, and a short intertrial interval (Smith, 2001). DTT involves the use of prompting and reinforcement, two treatment strategies that are discussed in greater detail later in this chapter. A prompt is provided, as needed, with or immediately after the instruction to support the individual to respond correctly. Error correction procedures are utilized when an individual responds incorrectly to teach the correct response. When an individual responds correctly, the individual is provided with reinforcement. DTT is typically conducted in a structured setting on a one-on-one basis. Distractions are reduced to help maintain the individual's attention on the target skill. While DTT supports rapid learning of new skills, a downside of this technique is limited generalization of skills to natural environments (e.g., school, playground) and natural cues (e.g., presence of toys instead of the direction to play with a toy; Smith, 2001;Sundberg & Partington, 1998; Weiss, 2001). Generalization should be assessed and additional protocols should be used to aid generalization, if needed, to mitigate this disadvantage.

While DTT is often used to teach simple, concrete skills like object labels, in recent years, complex skills like perspective taking and tacting situation-based emotions have been taught with this method (Gould, Tarbox, O'Hora, Noone, & Bergstrom, 2011; McHugh, Bobarnac, & Reed, 2011). Additionally, Jones, Feeley, and Takacs (2007) used DTT to effectively teach spontaneous responding to social cues (e.g., responding "bless you" when someone sneezes). Overall, DTT has been used to teach children with ASD a wide range of skills, including social, communication, behavior, joint attention, school readiness, academic, adaptive, safety, and vocational skills (Wong et al., 2013). While evidence for the use of DTT is strongest for the ASD population, emerging evidence supports the efficacy of the strategy across skills with individuals with comorbid ASD and ID (Peters-Scheffer et al., 2010).

Naturalistic Interventions Naturalistic interventions include a host of strategies aimed at increasing appropriate behaviors in settings where the behavior typically occurs. Various terms are used when referring to naturalistic interventions, including enhanced milieu teaching, natural environment training, natural language paradigm, and incidental teaching. Addressing skill acquisition in the natural environment is designed to increase generalization of the skill and harness the individual's motivation (Sundberg & Partington, 1998; Weiss, 2001). Naturalistic interventions involve instructing the individual during a natural activity using materials from the activity. For example, functions of objects may be targeted during craft time using the craft supplies (e.g., you draw with a crayon, you cut with scissors, you write on paper), rather than using pictures of the items in structured trials (i.e., DTT). One naturalistic strategy involves using naturally occurring reinforcers that are related to the task. For instance, a preferred outdoor activity (e.g., riding a bike) may be used as a reinforcer for tying shoes. Another strategy involves using the individual's interests during instruction. For example, colors may be targeted while the individual is eating a preferred snack (e.g., a strawberry is red, a banana is yellow).

Naturalistic interventions have a number of benefits. Koegel, Koegel, and Surratt (1992) demonstrated reduced rates of challenging behaviors during a naturalistic intervention when compared with an analogue condition (i.e., DTT). While naturalistic interventions typically result in greater generalization than structured interventions (e.g., DTT), learning can be slower; therefore, when using naturalistic interventions, it is critical to contrive teaching opportunities instead of merely waiting for learning opportunities to occur. Smith (2001) suggested that DTT and naturalistic interventions complement each other; skills initially taught using a DTT approach to achieve quicker mastery may be transferred to a naturalistic intervention approach to support generalization, independence, and initiation. Research supports the use of naturalistic interventions for individuals with ASD from infancy to young adulthood for a wide range of

behaviors, including social, communication, play, academic, and vocational skills (Wong et al., 2013).

Prompting Strategies A prompt is a cue used to aid the performance of a specific skill (Wong et al., 2013). There are a wide range of cues that may serve as prompts. Physical prompts involve providing direct support (e.g., hand over hand prompting while the individual is using a fork to eat). Echoic prompts may be used to provide a model for the individual to repeat (e.g., "say 'I want a cookie'"). Directive prompts provide a general or specific direction to the individual (e.g., "use your words," "what's next," "open the toothpaste cap"). Gestural prompts involve directing the individual's attention using motion (e.g., pointing to the trash can after directing the individual to throw away an item). Textual prompts involve writing out a phrase for the individual to say (e.g., writing out "Hi Susan" to prompt a greeting). Scripting is a prompting strategy that involves presenting a list of phrases, either textually or verbally, which is typically rehearsed in isolation before it is used in the intended setting. Visual prompts involve using pictorial representations to aid the performance of a skill (e.g., showing pictures of the steps involved in toothbrushing). Finally, modeling is a type of prompt that entails demonstrating the specific skill that the individual should imitate. Modeling can be used for a wide range of skills, ranging from simple (e.g., waving) to complex (e.g., engaging in a back-and-forth conversation).

Prompts are used to teach new skills. Overtime, prompts are faded out to support independent completion of the target skill. There are several options to fade prompts. Most-to-least prompting strategies involve initially providing the most intrusive prompt and gradually fading to a less intrusive prompt and then to independence. Least-to-most prompting involves initially providing a less intrusive prompt and progressing to a more intrusive prompt only if the less intrusive prompt is unsuccessful. Time delay is another method of fading prompts that involves inserting a delay between the instruction and the prompt; the delay can be systematically increased or remain the same. Prompting is a foundational strategy for a number of other behavioral treatments (e.g., DTT, PRT). Prompting strategies have been validated with individuals with ASD from infancy to 22 years of age for skills such as social, communication, adaptive, and academic (Wong et al., 2013). Evidence supports the use of prompting strategies for individuals with ID. Specifically, time delay has been shown to be effective in teaching academic skills to school-aged individuals with ID (Hudson, Browder, & Wood, 2013). Additionally, Sabielny and Cannella-Malone (2014) found both physical and physical plus directive prompting strategies to be effective in teaching daily living skills to adolescents with ID.

Reinforcement Reinforcement is a foundational component for the majority of the strategies described throughout this chapter and could fill entire volumes. There are two types of reinforcement that may be used to increase the occurrence of a target behavior: positive and negative (Cooper et al., 2007). Positive reinforcement entails the provision of a reinforcer following a specific behavior to increase the future occurrence of that behavior. A reinforcer is any stimulus that, when provided following a behavior, increases the likelihood that behavior will occur in the future. Reinforcers should not be confused with rewards or preferred items; the classification "reinforcer" is determined solely based on its impact on the individual's behavior. Reinforcers vary widely across individuals and may be edible (e.g., chips, candy), social (e.g., praise, hugs, tickles), an item or activity (e.g., toy, book, game), or currency that can be exchanged for other things (e.g., money, tokens). There are some considerations that impact the efficacy of positive reinforcement: (a) vary reinforcers to avoid satiation; (b) avoid states of deprivation that may be considered a restriction of rights (e.g., limiting access to water in certain circumstances); (c) provide reinforcement immediately following the target behavior to avoid reinforcing

an undesired behavior; and (d) move from contrived to naturally occurring reinforcers (e.g., shift from edibles to praise to avoid weight gain and teach the individual to contact natural reinforcement; Cooper et al., 2007).

Negative reinforcement involves the removal of an undesired stimulus (e.g., aversive event), contingent upon the occurrence of a behavior, that increases the future likelihood of that behavior. For instance, turning off the radio when an individual says, "Turn off the radio." There are ethical considerations in the implementation of negative reinforcement in regard to the presentation of an aversive event. The use of extremely aversive events may be considered unethical (Cooper et al., 2007). Furthermore, side effects, similar to those associated with punishment (e.g., avoidance behaviors, aggression), may be observed in response to aversive events. Reinforcement has been used in behavioral treatment protocols for individuals with ASD to address a variety of skills, such as working -memory (Baltruschat et al., 2011), inhibition of challenging behavior (Buckley & Newchok, 2006), task performance (Charlop-Christy & Haymes, 1998), liquid acceptance (Hagopian, Farrell, & Amari, 1996), safety skills (Hoch, Taylor, & Rodriguez, 2009), and question asking (Koegel, Camarata, Valdez-Menchaca, & Koegel, 1998) among other skills.

Task Analysis and Chaining Task analysis and chaining are used to teach new skills that can be broken into discrete steps. Task analysis is the process of breaking a skill down into smaller components. For example, a task analysis of putting on pants may include the following components: (1) grasp pants on waist band with the tag in the back, (2) step right foot into right pant leg, (3) push right foot through the bottom opening, (4) step left foot into left pant leg, (5) push left foot through the bottom opening, and (6) pull the waistband up to the waist. After a task analysis has been completed, instruction can begin on each step using chaining. There are several different chaining methods that differ depending on which step(s) are targeted first. For instance, forward chaining begins with targeting the first step in the sequence, while backward chaining begins with targeting the last step in the sequence. Task analysis and chaining have been validated for individuals with ASD, ages 3–14 years, across a variety of skills, including adaptive, motor, academic, social, and communication (Wong et al., 2013). Task analysis has also been identified as a promising strategy for teaching academic skills to school-aged children with ID (Hudson et al., 2013).

Video Modeling Video modeling involves teaching a new skill by showing a video of the correct performance of the target skill. Typically, the behavior is modeled by another person, such as an adult or peer; however, it is also possible to use self-video modeling, in which the video portrays the individual modeling the correct action (Delano, 2007). Studies support the use of video modeling across settings, including the classroom, home, and community (Delano, 2007). There is some evidence to suggest that video modeling is more effective than in vivo modeling, which is possibly attributed to increased attention via the reduction of distractions and the presentation of a focal point (Delano, 2007). Video modeling has been used to teach social, play, academic, and vocational skills to individuals with ASD up to 22 years of age (Wong et al., 2013). A recent review by Park, Bouck, and Duenas (2019) found video modeling to be effective for teaching daily living, academic, leisure, and job skills to individuals with ID; however, efficacy was enhanced by the use of additional strategies, such as prompting and error correction.

Communication Interventions

Augmentative and Alternative Communication Augmentative and alternative communication (AAC) refers to a variety of methods that supports forms of communication other than

spoken language. It is estimated that between 30% and 50% of children with ASD require the use of AAC devices due to a lack of functional communication (Light & McNaughton, 2012). There are no cognitive prerequisites required to use an AAC device, and there are a variety of systems available to meet individual needs (Light & McNaughton, 2012). Unaided AAC systems involve the individual using his or her body to communicate with others. This may include gestures (e.g., pointing to a desired item to request the item), facial expressions, and sign language (American Speech-Language-Hearing Association, n.d.). Aided systems involve the use of other items or devices to communicate, ranging from basic to high-tech (devices employing technology; American Speech-Language-Hearing Association, n.d.). Non-electronic aided AAC methods may include using a pencil and paper to write or using pictures to communicate (either by pointing or exchanging a picture card). PECS is a specific example that is described in detail later in this section. There are a variety of electronic devices developed specifically to aid communication. These devices have one or multiple buttons that, when pressed, play a prerecorded or synthesized voice output. A disadvantage of these devices is their relatively high cost to purchase and repair. The ubiquity and relatively low cost of tablets has led to a proliferation of speech-generating mobile applications. Proloquo2go (Collette, Brix, Brennan, DeRoma, & Muir, 2018) is one such application that allows for customization of pictures, voice output, and number of buttons among other features. Such applications have increased consumers' access to AAC technology at a decreased cost. There may also be a reduced level of social stigma associated with these applications in comparison to other AAC devices since tablets have become commonplace in school and community settings. Consistency across settings is important in the instruction and use of AAC devices; however, most individuals using AAC to communicate use a combination of methods. Research supports the use of AAC as a means of communication for individuals with ASD from 3 to 22 years of age (Wong et al., 2013). While the selection of a specific AAC system should be individualized, Ganz et al. (2012) found PECS and speech-generating devices to produce greater gains than other picture-based systems. Additionally, several studies have indicated that the use of AAC systems may aid verbal speech development in individuals with ASD and ID (Millar, Light, & Schlosser, 2006; Schlosser & Wendt, 2008).

Picture Exchange Communication System PECS targets communication skills using a sequence of six phases, in which requesting is a central focus (Bondy & Frost, 2001). The first phase involves teaching the individual to exchange a picture in a field of one to express wants and needs. The second phase includes teaching the individual to travel to the picture and then to the communicative partner to communicate. In the third phase, discrimination across pictures is targeted. The use of a sentence strip is taught in the fourth phase (i.e., placing an "I want" card on a sentence strip followed by a card representing a specific item/action and exchanging the sentence strip). In the fifth phase, the individual is taught to exchange PECS in response to the question, "What do you want?" In the final phase, the individual is taught to comment using pictures. PECS may be used as a form of FCT to reduce challenging behavior as well as increase an individual's ability to communicate his/her wants and needs. Evidence exists to support the use of PECS to increase communication skills, with an emphasis on requesting, in individuals with ASD and ID (Sulzer-Azaroff, Hoffman, Horton, Bondy, & Frost, 2009; Wong et al., 2013).

Social Interventions

Structured Play Groups and Peer-Mediated Instruction and Intervention Interventions may incorporate peers in order to teach skills and model appropriate behavior. Structured play groups are a type of group intervention that is

focused on a specific activity and takes place in a defined space. Peers with TD are often included to model appropriate social behavior; however, groups may also consist of peers with disabilities. The types of activities included in structured play groups are wide ranging. Several studies have reported on the effectiveness of LEGO® therapy, which involves children building collaboratively with LEGO sets in a group format (typically three children in a group; Owens, Granader, Humphrey, & Baron-Cohen, 2008). The children are required to follow LEGO club rules and identify their own solutions to problems when issues are highlighted by a therapist. In a study assessing the impact of LEGO therapy on treatment outcomes, Legoff and Sherman (2006) found that children with ASD who participated in LEGO therapy made greater gains on measures of socialization and social interaction after 3 years over children in a control group. Owens et al. (2008) compared the efficacy of LEGO® therapy with the Social Use of Language Programme (SULP) for children with ASD. SULP utilized stories, activities, and games to teach children social skills (e.g., eye contact, turn taking) in a group setting. While both groups showed significant decreases in challenging behavior over a control group, the LEGO therapy group demonstrated greater improvements on measures of social interaction (Owens et al., 2008). Evidence supports using structured play groups for individuals with ASD, ages 6–11, to improve social, communication, behavior, play, and academic skills (Wong et al., 2013).

Peer-mediated instruction and intervention (PMII) involves teaching an individual's peer(s) to implement an intervention. Peers with TD are taught strategies to interact with individuals with ASD. This intervention is generally utilized in a natural setting (e.g., school) and aids in the generalization of skills. PMII may be applied in both structured (e.g., teacher-led) and unstructured activities. Owen-DeSchryver, Carr, Cale, and Blakeley-Smith (2008) demonstrated that a peer training intervention improved social responses and initiations by children with ASD as well as initiations by the peers during lunch and recess. Loftin, Odom, and Lantz (2008) assessed the impact of a multicomponent social skills intervention on repetitive motor behaviors and socialization during school lunch and recess periods. Following intervention, which included peer training to respond positively to social initiations, instruction for children with ASD on social initiation, and the implementation of a self-monitoring program, participants were found to engage in fewer repetitive motor behaviors and have greater social initiations (Loftin et al., 2008). The number of peers involved in an intervention may vary from one to many. Carter, Cushing, Clark, and Kennedy (2005) found increased rates of socialization and engagement in the curriculum in students with disabilities who received support from two peers versus one peer. Class-wide peer instruction (where all children in the class receive instruction on how to support others) has also been used to improve reading fluency, reading comprehension, and social interactions (e.g., Kamps, Barbetta, Leonard, & Delquadri, 1994; Laushey & Heflin, 2000). Evidence supports the use of PMII to address social, communication, play, and academic skills in preschool- to high school-age individuals with ASD (Wong et al., 2013).

Social Narratives Social narratives (e.g., Social Stories™; Gray, 2000) are textual or visual stories used to prepare an individual for a particular situation by concretely describing appropriate behavior. While often used to depict social situations (e.g., how to respond when someone gives a gift), social narratives may be used to clearly and succinctly communicate information for a wide range of situations (e.g., crossing the street, maintaining appropriate behavior at the grocery store, responding to fire drills). The goal of a social narrative is to prepare the individual for the specified situation. This is accomplished by highlighting the relevant cues for the individual to attend to, as well as describing the expected response in the given situation. Social narratives are often written in first-person perspective to aid the individual in seeing themselves in the situa-

tion. Social narratives are brief, focused on the relevant information, and may include pictures, which are especially helpful for individuals who are unable to read. Social narratives may be read by the individual or may be read aloud to him/her. Evidence supports the use of social narratives with individuals with ASD from 3 to 18 years of age (Wong et al., 2013). Social Stories have been used to reduce challenging behaviors and increase appropriate behaviors in children with ASD (Chan & O'Reilly, 2008; Dodd, Hupp, Jewell, & Krohn, 2008). Emerging evidence exists for the use of social narratives with individuals with ASD and comorbid ID; Reynhout and Carter (2007) implemented a Social Story intervention to decrease a targeted challenging behavior for a child with ASD and moderate ID.

Method of Delivery

Strategies aimed at reducing challenging behaviors and increasing skill acquisition may be implemented through mediums other than a clinician. This section provides an overview of evidence-based delivery methods for ASD interventions.

Caregiver-Implemented Intervention and Training Caregiver training refers to any level of caregiver involvement in treatment, which may include a variety of different practices. According to Bearss, Burrell, Stewart, and Scahill (2015), caregiver training may be split into two broad categories: caregiver support and caregiver implementation. Caregiver support involves providing the caregiver with knowledge and resources; therefore, the child indirectly benefits. Conversely, caregiver implementation involves teaching the caregiver techniques and interventions to implement directly with their child. Caregiver-implemented interventions may be focused on reducing challenging behaviors and/or increasing appropriate skills. Both caregiver support and caregiver-implemented intervention have empirical support for individuals with ASD up to 11 years of age (Wong et al., 2013). Caregiver training has been shown to improve caregiver-child interactions, increase language comprehension, reduce ASD severity, decrease caregiver stress, reduce caregiver symptoms of depression, and increase caregiver self-efficacy (Frantz, Hansen, & Machalicek, 2018; Oono, Honey, & McConachie, 2013). Furthermore, caregiver-implemented interventions targeting language skills have been found to increase both receptive and expressive language in children with and without ID (Roberts & Kaiser, 2011). A caregiver's ability to effectively implement intervention may be impacted by a variety of factors (e.g., availability, education, stress level); as such, an individualized approach to caregiver involvement in treatment is suggested (Forehand & Kotchick, 2002).

Computer-Based Intervention Computer-based intervention (CBI) is software developed to deliver treatment using built-in mechanisms, such as instruction, feedback, and data collection. Several literature reviews have investigated the use of CBI to teach skills to children with ASD, such as academic skills (Knight, McKissick, & Saunders, 2013; Pennington, 2010), communication skills (Ramdoss et al., 2011), literacy skills (Ramdoss et al., 2011), reading comprehension (Khowaja & Salim, 2013), and social skills (Ramdoss et al., 2012). While CBI research has primarily focused on basic literacy skills, including matching, receptive identification, and spelling (Knight et al., 2013; Pennington, 2010), other technologies are emerging (e.g., virtual reality, robotics) that may be useful for targeting more complex skills. Virtual reality has emerged as a promising tool for teaching social, communication, daily living, and cognitive skills (den Brok & Sterkenburg, 2015; Mesa-Gresa, Gil-Gómez, Lozano-Quilis, & Gil-Gómez, 2018). There are concerns about prolonged screen time usage (Boone, Gordon-Larsen, Adair, & Popkin, 2007; Hale & Guan, 2015; Hardy, Denney-Wilson, Thrift, Okely, & Baur, 2010), which emphasizes the need for supervision and restricted use. Nevertheless, such interventions may be useful to augment portions of therapy, supplement ongoing interventions, and deliver treatment to individuals with limited access. While CBI in general

has been found to be evidence based (Odom, Collet-Klingenberg, et al., 2010; Wong et al., 2013), the success of a particular CBI software depends entirely on its ability to deliver intervention (Ramdoss, Lang, et al., 2011; Ramdoss, Mulloy, et al., 2011). There are a vast number of technologies developed and marketed to children with ASD, and very little have undergone empirical testing (Novack, Hong, Dixon, & Granpeesheh, 2018). Discretion must be taken when selecting individual products.

Focused Treatment Summary Many focused interventions have been validated for the treatment of individuals with ASD. The focused interventions discussed in this chapter may be classified by the behaviors they target (e.g., skill acquisition, behavior reduction) as well as the method of delivery (e.g., caregiver, computer based). Rarely are focused interventions used in isolation; instead, strategies are typically combined into a larger treatment package. While each intervention is presented as a discrete strategy in this chapter, there is some degree of overlap between strategies. Reinforcement, for instance, is foundational to many of the interventions described (e.g., differential reinforcement, PECS, self-management). Foundational ABA interventions, including prompting and reinforcement, have the greatest empirical support (Wong et al., 2013).

Conclusions

Given the variety of comprehensive treatment models and service providers available, it is important to consider the empirical support for comprehensive models as well as the focused interventions that make up those models. A number of empirically supported comprehensive and focused interventions have been found to be effective for the treatment of ASD. The underlying goal of both comprehensive and focused treatments is to increase appropriate behaviors and decrease challenging behaviors. With a vast number of interventions available, considerations should be made to individualize treatment to the individual; interventions should be selected that are appropriate for the target skill or challenging behavior, are suitable for the treatment setting (e.g., clinic, home, school), take into account the interests and learning style of the individual, and finally are empirically supported. While there are many evidence-based interventions for the treatment of ASD, there are gaps in the research that need to be addressed.

While there is growing support for a few comprehensive treatments (ABA, PRT, LEAP, ESDM), research comparing the differential effectiveness of these models is lacking. Oftentimes, ASD treatment research includes control groups made up of individuals receiving the same intervention at a lower dosage or with less oversight (Lovaas, 1987; Strain & Bovey, 2011) or individuals receiving an eclectic assortment of community-based interventions (Dawson et al., 2010; Eikeseth et al., 2002) rather than a single treatment philosophy administered at a consistent dosage. Meta-analyses may be another avenue to compare outcomes of different comprehensive treatment models as conducted by Tachibana et al. (2017). Randomized, controlled trials for behavioral, social-communication, and multimodal developmental interventions were identified and analyzed; however, the limited number of studies available curbed efforts to compare outcomes across interventions (Tachibana et al., 2017). Research and analyses comparing the effectiveness of different treatment models are warranted.

Future ASD treatment research should also consider subpopulations of ASD in their investigations. Evidence is emerging that suggests there are different subtypes of ASD (Beglinger & Smith, 2001), which may respond differently to treatment. One particular subpopulation that ought to be investigated further is individuals with ASD and co-occurring ID. Evidence suggests that intellectual functioning impacts treatment gains for individuals with ASD (Ben-Itzchak & Zachor, 2007; Harris & Handleman, 2000), and given the different clinical presentation, ASD with co-occurring ID may constitute a distinct subtype of ASD (Lecavalier et al., 2011; Nowell et al., 2017). Future ASD treatment research

should consider such ASD subpopulations and explore targeted treatment methods to maximize outcomes. In the meantime, evidence-based practices should be emphasized in the treatment of individuals with ASD and co-occurring ID.

References

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

American Speech-Language-Hearing Association. (n.d.). *Augmentative and alternative communication (AAC)*. Retrieved from https://www.asha.org/public/speech/disorders/aac/

Baio, J., Wiggins, L., Christensen, D. L., Maenner, M. J., Daniels, J., Warren, Z., … Dowling, N. F. (2018). Prevalence of autism spectrum disorder among children aged 8 years – Autism and Developmental Disabilities Monitoring Network, 11 sites, United States, 2014. *Surveillance Summaries, 67*, 1–23. https://doi.org/10.15585/mmwr.ss6706a1

Baltruschat, L., Hasselhorn, M., Tarbox, J., Dixon, D. R., Najdowski, A. C., Mullins, R. D., & Gould, E. R. (2011). Addressing working memory in children with autism through behavioral intervention. *Research in Autism Spectrum Disorders, 5*, 267–276. https://doi.org/10.1016/j.rasd.2010.04.008

Bearss, K., Burrell, T. L., Stewart, L., & Scahill, L. (2015). Parent training in autism spectrum disorder: What's in a name? *Clinical Child and Family Psychology Review, 18*, 170–182. https://doi.org/10.1007/s10567-015-0179-5

Beglinger, L. J., & Smith, T. H. (2001). A review of subtyping in autism and proposed dimensional classification model. *Journal of Autism and Developmental Disorders, 31*, 411–422. https://doi.org/10.1023/A:1010616719877

Behavior Analyst Certification Board, Inc. (2014). *Applied behavior analysis treatment of autism spectrum disorder: Practice guidelines for healthcare funders and managers* (2nd ed.). Retrieved from https://www.bacb.com/wp-content/uploads/2017/09/ABA_Guidelines_for_ASD.pdf

Ben-Itzchak, E., Lahat, E., Burgin, R., & Zachor, A. D. (2008). Cognitive, behavior and intervention outcome in young children with autism. *Research in Developmental Disabilities, 29*, 447–458. https://doi.org/10.1016/j.ridd.2007.08.003

Ben-Itzchak, E., & Zachor, D. A. (2007). The effects of intellectual functioning and autism severity on outcome of early behavioral intervention for children with autism. *Research in Developmental Disabilities, 28*, 287–303. https://doi.org/10.1016/j.ridd.2006.03.002

Ben-Itzchak, E., & Zachor, D. A. (2011). Who benefits from early intervention in autism spectrum disorders? *Research in Autism Spectrum Disorders, 5*, 345–350. https://doi.org/10.1016/j.rasd.2010.04.018

Bishop, S. L., Richler, J., & Lord, C. (2006). Association between restricted and repetitive behaviors and nonverbal IQ in children with autism spectrum disorders. *Child Neuropsychology, 12*, 247–267. https://doi.org/10.1080/09297040600630288

Bondy, A., & Frost, L. (2001). The picture exchange communication system. *Behavior Modification, 25*, 725–744. https://doi.org/10.1177/0145445501255004

Boone, J. E., Gordon-Larsen, P., Adair, L. S., & Popkin, B. M. (2007). Screen time and physical activity during adolescence: Longitudinal effects on obesity and young adulthood. *International Journal of Behavioral Nutrition and Physical Activity, 4*, 1–10. https://doi.org/10.1186/1479-5868-4-26

Boyd, B. A., Hume, K., McBee, M. T., Alessandri, M., Gutierrez, A., Johnson, L., … Odom, S. L. (2014). Comparative efficacy of LEAP, TEACCH and non-model-specific special education programs for preschoolers with autism spectrum disorders. *Journal of Autism and Developmental Disorders, 44*, 366–380. https://doi.org/10.1007/s10803-013-1877-9

Bozkus-Genc, G., & Yucesoy-Ozkan, S. (2016). Meta-analysis of pivotal response training for children with autism spectrum disorder. *Education and Training in Autism and Developmental Disabilities, 51*, 13–26.

Buckley, S. D., & Newchok, D. K. (2006). Analysis and treatment of problem behavior evoked by music. *Journal of Applied Behavior Analysis, 39*, 141–144. https://doi.org/10.1901/jaba.2006.120-04

Carr, M. E., Moore, D. W., & Anderson, A. (2014). Self-management interventions on students with autism: A meta-analysis of single-subject research. *Exceptional Children, 81*, 28–44. https://doi.org/10.1177/0014402914532235

Carter, E. W., Cushing, L. S., Clark, N. M., & Kennedy, C. H. (2005). Effects of peer support interventions on students' access to the general curriculum and social interactions. *Research and Practice for Persons with Severe Disabilities, 30*, 15–25. https://doi.org/10.2511/rpsd.30.1.15

Chan, J. M., Lambdin, L., Graham, K., Fragale, C., & Davis, T. (2014). A picture-based activity schedule intervention to teach adults with mild intellectual disability to use an iPad during a leisure activity. *Journal of Behavioral Education, 23*, 247–257. https://doi.org/10.1007/s10864-014-9194-8

Chan, J. M., & O'Reilly, M. F. (2008). A Social Stories™ intervention package for students with autism in inclusive classroom settings. *Journal of Applied Behavior Analysis, 41*, 405–409. https://doi.org/10.1901/jaba.2008.41-405

Charlop-Christy, M. H., & Haymes, L. K. (1998). Using objects of obsession as token reinforcers for children with autism. *Journal of Autism and Developmental*

Disorders, 28, 189–198. https://doi.org/10.1023/A:1026061220171

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

Cohen, H., Amerine-Dickens, M., & Smith, T. (2006). Early intensive behavioral treatment: Replication of the UCLA model in a community setting. *Journal of Developmental & Behavioral Pediatrics, 27*, S145–S155.

Collette, D., Brix, A., Brennan, P., DeRoma, N., & Muir, B. C. (2018). Proloquo2go enhances classroom performance in children with autism spectrum disorder. *OTJR: Occupation, Participation and Health, 39*(3), 143–150. https://doi.org/10.1177/1539449218799451

Cooper, J. O., Heron, T. E., & Heward, W. L. (2007). *Applied behavior analysis* (2nd ed.). Upper Saddle River, NJ: Pearson.

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*, e17–e23. https://doi.org/10.1542/peds.2009-0958

Delano, M. E. (2007). Video modeling interventions for individuals with autism. *Remedial and Special Education, 28*, 33–42. https://doi.org/10.1177/07419325070280010401

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*, 1–10. https://doi.org/10.3109/17483107.2014.921248

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, NY: Springer. https://doi.org/10.1007/978-1-4614-3037-7_3

Dodd, S., Hupp, S. D., Jewell, J. D., & Krohn, E. (2008). Using parents and siblings during a Social Story intervention for two children diagnosed with PDD-NOS. *Journal of Developmental and Physical Disabilities, 20*, 217–229. https://doi.org/10.1007/s10882-007-9090-4

Eikeseth, S., Smith, T., Jahr, E., & Eldevik, S. (2002). Intensive behavioral treatment at school for 4- to 7-year-old children with autism: A 1-year comparison controlled study. *Behavior Modification, 26*, 49–68. https://doi.org/10.1177/0145445502026001004

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

Eldevik, S., Eikeseth, S., Jahr, E., & Smith, T. (2006). Effects of low intensity behavioral treatment for children with autism and mental retardation. *Journal of Autism and Developmental Disorders, 36*, 211–224. https://doi.org/10.1007/s10803-005-0058-x

Eldevik, S., Hastings, R. P., Hughes, J. C., Jahr, E., Eikeseth, S., & Cross, S. (2009). Meta-Analysis of early intensive behavioral intervention for children with autism. *Journal of Clinical Child & Adolescent Psychology, 38*, 439–450. https://doi.org/10.1080/15374410902851739

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., Hastings, R. P., Jahr, E., & Hughes, J. C. (2012). Outcomes of behavioral intervention for children with autism in mainstream preschool settings. *Journal of Autism and Developmental Disorders, 42*, 210–220. https://doi.org/10.1007/s10803-011-1234-9

Flanagan, H. E., Perry, A., & Freeman, N. L. (2012). Effectiveness of large-scale community-based intensive behavioral intervention: A waitlist comparison study exploring outcomes and predictors. *Research in Autism Spectrum Disorders, 6*, 673–682. https://doi.org/10.1016/j.rasd.2011.09.011

Forehand, R., & Kotchick, B. A. (2002). Behavioral parent training: Current challenges and potential solutions. *Journal of Child and Family Studies, 11*, 377–384. https://doi.org/10.1023/A:1020913422609

Frantz, R., Hansen, S. G., & Machalicek, W. (2018). Interventions to promote well-being in parents of children with autism: A systematic review. *Review Journal of Autism and Developmental Disorders, 5*, 58–77. https://doi.org/10.1007/s40489-017-0123-3

Ganz, J. B., Earles-Vollrath, T. L., Heath, A. K., Parker, R. I., Rispoli, M. J., & Duran, J. B. (2012). A meta-analysis of single case research studies on aided augmentative and alternative communication systems with individuals with autism spectrum disorders. *Journal of Autism and Developmental Disorders, 42*, 60–74. https://doi.org/10.1007/s10803-011-1212-2

Gould, E., Tarbox, J., O'Hora, D., Noone, S., & Bergstrom, R. (2011). Teaching children with autism a basic component skill of perspective-taking. *Behavioral Interventions, 26*, 50–66. https://doi.org/10.1002/bin.320

Granpeesheh, D., Dixon, D. R., Tarbox, J., Kaplan, A. M., & Wilke, A. E. (2009). The effects of age and treatment intensity on behavioral intervention outcomes for children with autism spectrum disorders. *Research in Autism Spectrum Disorders, 3*, 1014–1022. https://doi.org/10.1016/j.rasd.2009.06.007

Granpeesheh, D., Tarbox, J., & Dixon, D. R. (2009). Applied behavior analytic interventions for children

with autism: A description and review of treatment research. *Annals of Clinical Psychiatry, 21*, 162–173.

Granpeesheh, D., Tarbox, J., Najdowski, A. C., & Kornack, J. (Eds.). (2014). *Evidence-based treatment for children with autism: The CARD model*. Waltham, MA: Elsevier.

Gray, C. (2000). *The new Social Story book*. Arlington, TX: Jenison Public Schools.

Hagopian, L. P., Farrell, D. A., & Amari, A. (1996). Treating total liquid refusal with backward chaining and fading. *Journal of Applied Behavior Analysis, 29*, 573–575. https://doi.org/10.1901/jaba.1996.29-573

Hale, L., & Guan, S. (2015). Screen time and sleep among school-aged children and adolescents: A systematic literature review. *Sleep Medicine Reviews, 21*, 50–58. https://doi.org/10.1016/j.smrv.2014.07.007

Hardy, L. L., Denney-Wilson, E., Thrift, A. P., Okely, A. D., & Baur, L. A. (2010). Screen time and metabolic risk factors among adolescents. *Archives of Pediatrics and Adolescent Medicine, 164*, 643–649. https://doi.org/10.1001/archpediatrics.2010.88

Harris, S. L., & Handleman, J. S. (2000). Age and IQ at intake as predictors of placement for young children with autism: A four- to six-year follow-up. *Journal of Autism and Developmental Disorders, 30*, 137–142. https://doi.org/10.1023/A:1005459606120

Hayward, D., Eikeseth, S., Gale, C., & Morgan, S. (2009). Assessing progress during treatment for young children with autism receiving intensive behavioral interventions. *Autism, 13*, 613–633. https://doi.org/10.1177/1362361309340029

Heath, A. K., Ganz, J. B., 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

Hoch, H., Taylor, B. A., & Rodriguez, A. (2009). Teaching teenagers with autism to answer cell phones and seek assistance when lost. *Behavior Analysis in Practice, 2*, 14–20. https://doi.org/10.1007/BF03391733

Howard, J. S., Sparkman, C. R., Cohen, H. G., Green, G., & Stanislaw, H. (2005). A comparison of intensive behavior analytic and eclectic treatments for young children with autism. *Research in Developmental Disabilities, 26*, 359–383. https://doi.org/10.1016/j.ridd.2004.09.005

Howlin, P., Magiati, I., & Charman, T. (2009). Systematic review of early intensive behavioral interventions for children with autism. *American Journal on Intellectual and Developmental Disabilities, 114*, 23–41. https://doi.org/10.1352/2009.114:23;nd41

Hudson, M. E., Browder, D. M., & Wood, L. A. (2013). Review of experimental research on academic learning by students with moderate and severe intellectual disability in general education. *Research and Practice for Persons with Severe Disabilities, 38*, 17–29. https://doi.org/10.2511/027494813807046926

Hurwitz, S., & Minshawi, N. F. (2012). Methods of defining and observing behaviors. In J. L. Matson (Ed.), *Functional assessment for challenging behaviors* (pp. 91–103). New York, NY: Springer. https://doi.org/10.1007/978-1-4614-3037-7_6

Ivy, J. W., & Schreck, K. A. (2016). The efficacy of ABA for individuals with autism across the Lifespan. *Current Developmental Disorders Reports, 3*, 57–66. https://doi.org/10.1007/s40474-016-0070-1

Jang, J., Dixon, D. R., Tarbox, J., & Granpeesheh, D. (2011). Symptom severity and challenging behavior in children with ASD. *Research in Autism Spectrum Disorders, 5*, 1028–1032. https://doi.org/10.1016/j.rasd.2010.11.008

Jones, E. A., Feeley, K. M., & Takacs, J. (2007). Teaching spontaneous responses to young children with autism. *Journal of Applied Behavior Analysis, 40*, 565–570. https://doi.org/10.1901/jaba.2007.40-565

Kamps, D. M., Barbetta, P. M., Leonard, B. R., & Delquadri, J. (1994). Classwide peer tutoring: An integration strategy to improve reading skills and promote peer interactions among students with autism and general education peers. *Journal of Applied Behavior Analysis, 27*, 49–61. https://doi.org/10.1901/jaba.1994.27-49

Kanne, S. M., Gerber, A. J., Quirmbach, L. M., Sparrow, S. S., Cicchetti, D. V., & Saulnier, C. A. (2011). The role of adaptive behavior in autism spectrum disorders: Implications for functional outcome. *Journal of Autism and Developmental Disorders, 41*, 1007–1018. https://doi.org/10.1007/s10803-010-1126-4

Khowaja, K., & Salim, S. S. (2013). A systematic review of strategies and computer-based intervention (CBI) for reading comprehension of children with autism. *Research in Autism Spectrum Disorders, 7*, 1111–1121. https://doi.org/10.1016/j.rasd.2013.05.009

Knight, V., McKissick, B. R., & Saunders, A. (2013). A review of technology-based interventions to teach academic skills to students with autism spectrum disorder. *Journal of Autism and Developmental Disorders, 43*, 2628–2648. https://doi.org/10.1007/s10803-013-1814-y

Koegel, L. K., Camarata, S. M., Valdez-Menchaca, M., & Koegel, R. L. (1998). Setting generalization of question-asking by children with autism. *American Journal on Mental Retardation, 102*, 346–357.

Koegel, L. K., Koegel, R. L., Shoshan, Y., & Mcnerney, E. (1999). Pivotal response intervention II: Preliminary long-term outcome data. *Journal of the Association for Persons with Severe Handicaps, 24*, 186–198. https://doi.org/10.2511/rpsd.24.3.186

Koegel, R. L., & Frea, W. D. (1993). Treatment of social behavior in autism through the modification of pivotal social skills. *Journal of Applied Behavior Analysis, 26*, 369–377. https://doi.org/10.1901/jaba.1993.26-369

Koegel, R. L., & Koegel, L. K. (2019). *Pivotal response treatment for autism spectrum disorders* (2nd ed.). Baltimore, MA: Paul H. Brookes Publishing.

Koegel, R. L., Koegel, L. K., & Surratt, A. (1992). Language intervention and disruptive behavior in preschool children with autism. *Journal of Autism and Developmental Disorders, 22*, 141–153. https://doi.org/10.1007/BF01058147

Koegel, R. L., O'Dell, M. C., & Koegel, L. K. (1987). A natural language teaching paradigm for nonverbal autistic children. *Journal of Autism and Developmental Disorders, 17*, 187–200. https://doi.org/10.1007/BF01495055

Lancioni, G. E., Singh, N. N., O'Reilly, M. F., Sigafoos, J., & Didden, R. (2012). Function of challenging behaviors. In J. L. Matson (Ed.), *Functional assessment for challenging behaviors* (pp. 45–64). New York, NY: Springer. https://doi.org/10.1007/978-1-4614-3037-7_4

Laushey, K. M., & Heflin, L. J. (2000). Enhancing social skills of kindergarten children with autism through the training of multiple peers as tutors. *Journal of Autism and Developmental Disorders, 30*, 183–193. https://doi.org/10.1023/A:1005558101038

Lecavalier, L., Snow, A. V., & Norris, M. (2011). Autism spectrum disorder and intellectual disability. In J. L. Matson & P. Sturmey (Eds.), *International handbook of autism and pervasive developmental disorders* (pp. 37–51). New York, NY: Springer. https://doi.org/10.1007/978-1-4419-8065-6_4

Legoff, D. B., & Sherman, M. (2006). Long-term outcome of social skills intervention based on interactive LEGO© play. *Autism, 10*, 317–329. https://doi.org/10.1177/1362361306064403

Light, J., & McNaughton, D. (2012). The changing face of augmentative and alternative communication: Past, present, and future challenges. *Augmentative and Alternative Communication, 28*, 197–204. https://doi.org/10.3109/07434618.2012.737024

Linstead, E., Dixon, D. R., French, R., Granpeesheh, D., Adams, H., German, R., … Kornack, J. (2016). Intensity and learning outcomes in the treatment of children with autism spectrum disorder. *Behavior Modification, 41*, 229–252. https://doi.org/10.1177/0145445516667059

Linstead, E., Dixon, D. R., Hong, E., Burns, C. O., French, R., Novack, M. N., & Granpeesheh, D. (2017). An evaluation of the effects of intensity and duration on outcomes across treatment domains for children with autism spectrum disorder. *Translational Psychiatry, 7*, 1–6. https://doi.org/10.1038/tp.2017.207

Loftin, R. L., Odom, S. L., & Lantz, J. F. (2008). Social interaction and repetitive motor behaviors. *Journal of Autism and Developmental Disorders, 38*, 1124–1135. https://doi.org/10.1007/s10803-007-0499-5

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. https://doi.org/10.1037/0022-006X.55.1.3

Magiati, I., Charman, T., & Howlin, P. (2007). A two-year prospective follow-up study of community-based early intensive behavioural intervention and specialist nursery provision for children with autism spectrum disorders. *Journal of Child Psychology and Psychiatry, 48*, 803–812. https://doi.org/10.1111/j.1469-7610.2007.01756.x

Magiati, I., Moss, J., Charman, T., & Howlin, P. (2011). Patterns of change in children with autism spectrum disorders who received community based comprehensive interventions in their pre-school years: A seven year follow-up study. *Research in Autism Spectrum Disorder, 5*, 1016–1027. https://doi.org/10.1016/j.rasd.2010.11.007

Maglione, M. A., Gans, D., Das, L., Timbie, J., & Kasari, C. (2012). Nonmedical interventions for children with ASD: Recommended guidelines and further research needs. *Pediatrics, 130*, S169–S178. https://doi.org/10.1542/peds.2012-0900O

Makrygianni, M. K., & Reed, P. (2010). A meta-analytic review of the effectiveness of behavioural early intervention programs for children with autism spectrum disorders. *Research in Autism Spectrum Disorders, 4*, 577–593. https://doi.org/10.1016/j.rasd.2010.01.014

Martinez, C. K., & Betz, A. M. (2013). Response interruption and redirection: Current research trends and clinical application. *Journal of Applied Behavior Analysis, 46*, 549–554. https://doi.org/10.1002/jaba.38

Matson, J. L. (Ed.). (2012). *Functional assessment for challenging behaviors*. New York, NY: Springer.

Matson, J. L., & Shoemaker, M. (2009). Intellectual disability and its relationship to autism spectrum disorders. *Research in Developmental Disabilities, 30*, 1107–1114. https://doi.org/10.1016/j.ridd.2009.06.003

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

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*, 1423–1428. https://doi.org/10.1007/s10803-010-1152-2

Mechling, L. C. (2007). Assistive technology as a self-management tool for prompting students with intellectual disabilities to initiate and complete daily tasks: A literature review. *Education and Training in Developmental Disabilities, 42*, 252.

Mesa-Gresa, P., Gil-Gómez, H., Lozano-Quilis, J. A., & Gil-Gómez, J. A. (2018). Effectiveness of virtual reality for children and adolescents with autism spectrum disorder: An evidence-based systematic review. *Sensors, 18*, 1–15. https://doi.org/10.3390/s18082486

Millar, D. C., Light, J. C., & Schlosser, R. W. (2006). The impact of augmentative and alternative communication intervention on the speech production of individuals with developmental disabilities: A research review. *Journal of Speech, Language, and Hearing Research, 49*, 248–264. https://doi.org/10.1044/1092-4388(2006/021)

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*, 2769–2777. https://doi.org/10.1007/s10803-014-2137-3

Myers, S. M., & Johnson, C. P. (2007). Management of children with autism spectrum disorders. *Pediatrics, 120*, 1162–1182. https://doi.org/10.1542/peds.2007-2362

Napolitano, D. A., Knapp, V. M., Speares, E., McAdam, D. B., & Brown, H. (2012). The role of functional assessment in treatment planning. In J. L. Matson (Ed.), *Functional assessment for challenging behaviors* (pp. 195–211). New York, NY: Springer. https://doi.org/10.1007/978-1-4614-3037-7_12

National Autism Center. (2015). *Findings and conclusions: National standards project, phase 2*. Randolph, MA: Author.

National Research Council. (2001). *Educating children with autism*. Washington, DC: National Academy Press.

Novack, M. N., Hong, E., Dixon, D. R., & Granpeesheh, D. (2018). An evaluation of a mobile application designed to teach receptive language skills to children with autism spectrum disorder. *Behavior Analysis in Practice, 12*, 66–77. https://doi.org/10.1007/s40617-018-00312-7

Nowell, K. P., Goin-Kochel, R., McQuillin, S., & Mire, S. S. (2017). Intellectual functioning and autism spectrum disorder: Can profiles inform identification of subpopulations? *Review Journal of Autism and Developmental Disorders, 4*, 339–349. https://doi.org/10.1007/s40489-017-0118-0

Odom, S. L., Boyd, B. A., Hall, L. J., & Hume, K. (2010). Evaluation of comprehensive treatment models for individuals with autism spectrum disorders. *Journal of Autism and Developmental Disorders, 40*, 425–436. https://doi.org/10.1007/s10803-009-0825-1

Odom, S. L., Collet-Klingenberg, L., Rogers, S. J., & Hatton, D. D. (2010). Evidence-based practices in interventions for children and youth with autism spectrum disorders. *Preventing School Failure, 54*, 275–282. https://doi.org/10.1080/10459881003785506

Oono, I. P., Honey, E. J., & McConachie, H. (2013). Parent-mediated early intervention for young children with autism spectrum disorders (ASD). *Evidence-Based Child Health: A Cochrane Review Journal, 8*, 2380–2479. https://doi.org/10.1002/14651858.CD009774.pub2

Owen-DeSchryver, J. S., Carr, E. G., Cale, S. I., & Blakeley-Smith, A. (2008). Promoting social interactions between students with autism spectrum disorders and their peers in inclusive school settings. *Focus on Autism and Other Developmental Disabilities, 23*, 15–28. https://doi.org/10.1177/1088357608314370

Owens, G., Granader, Y., Humphrey, A., & Baron-Cohen, S. (2008). LEGO® therapy and the Social Use of Language Programme: An evaluation of two social skills interventions for children with high functioning autism and Asperger syndrome. *Journal of Autism and Developmental Disorders, 38*, 1944–1957. https://doi.org/10.1007/s10803-008-0590-6

Park, J., Bouck, E., & Duenas, A. (2019). The effect of video modeling and video prompting interventions on individuals with intellectual disability: A systematic literature review. *Journal of Special Education Technology, 34*, 3–16. https://doi.org/10.1177/0162643418780464

Pennington, R. C. (2010). Computer-assisted instruction for teaching academic skills to students with autism spectrum disorders: A review of literature. *Focus on Autism and Other Developmental Disabilities, 25*, 239–248. https://doi.org/10.1177/1088357610378291

Perry, A., Cummings, A., Geier, J. D., Freeman, N. L., Hughes, S., Managhan, T., … Williams, J. (2011). Predictors of outcome for children receiving intensive behavioral intervention in a large, community-based program. *Research in Autism Spectrum Disorders, 5*, 592–603. https://doi.org/10.1016/j.rasd.2010.07.003

Peters-Scheffer, N., Didden, R., Korzilius, H., & Sturmey, P. (2011). A meta-analytic study on the effectiveness of comprehensive ABA-based early intervention programs for children with autism spectrum disorders. *Research in Autism Spectrum Disorders, 5*, 60–69. https://doi.org/10.1016/j.rasd.2010.03.011

Peters-Scheffer, N., Didden, R., Mulders, M., & Korzilius, 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

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*, 39–54. https://doi.org/10.1007/s10567-015-0197-3

Ramdoss, S., Lang, R., Mulloy, A., Franco, J., O'Reilly, M., Didden, R., & Lancioni, G. (2011). Use of computer-based intervention to teach communication skills to children with autism spectrum disorders: A systematic review. *Journal of Behavioral Education, 20*, 55–76. https://doi.org/10.1007/s10864-010-9112-7

Ramdoss, S., Machalicek, W., Rispoli, M., Mulloy, A., Lang, R., & O'Reilly, M. (2012). Computer-based interventions to improve social and emotional skills in individuals with autism spectrum disorders: A systematic review. *Developmental Neurorehabilitation, 15*, 119–135. https://doi.org/10.3109/17518423.2011.651655

Ramdoss, S., Mulloy, A., Lang, R., O'Reilly, M., Sigafoos, J., Lancioni, G., & El Zein, F. (2011). Use of computer-based interventions to improve literacy skills in students with autism spectrum disorders: A systematic review. *Research in Autism Spectrum Disorders, 5*, 1306–1318. https://doi.org/10.1016/j.rasd.2011.03.004

Reichow, B. (2012). Overview of meta-analyses on early intensive behavioral intervention for young children with autism spectrum disorders. *Journal of Autism*

and Developmental Disorders, 42, 512–520. https://doi.org/10.1007/s10803-011-1218-9

Reichow, B., Barton, E. E., Boyd, B. A., & Hume, K. (2012). Early intensive behavioral intervention (EIBI) for young children with autism spectrum disorders (ASD). *Cochrane Database of Systematic Reviews, 10*. https://doi.org/10.1002/14651858.CD009260.pub2

Reichow, B., Hume, K., Barton, E. E., & Boyd, B. A. (2018). Early intensive behavioral intervention (EIBI) for young children with autism spectrum disorders (ASD). *Cochrane Database of Systematic Reviews, 5*, 1–63. https://doi.org/10.1002/14651858.CD009260.pub3

Reichow, B., & Wolery, M. (2009). Comprehensive synthesis of early intensive behavioral interventions for young children with autism based on the UCLA Young Autism Project model. *Journal of Autism and Developmental Disorders, 39*, 23–41. https://doi.org/10.1007/s10803-008-0596-0

Remington, B., Hastings, R. P., Kovshoff, H., degli Espinosa, F., Jahr, E., Brown, T., … Ward, N. (2007). Early intensive behavioral intervention: Outcomes for children with autism and their parents after two years. *American Journal on Mental Retardation, 112*, 418–438. https://doi.org/10.1352/0895-8017(2007)112[418:EIBIOF]2.0.CO;2

Reynhout, G., & Carter, M. (2007). Social Story™ efficacy with a child with autism spectrum disorder and moderate intellectual disability. *Focus on Autism and Other Developmental Disabilities, 22*, 173–181. https://doi.org/10.1177/10883576070220030401

Roberts, M. Y., & Kaiser, A. P. (2011). The effectiveness of parent-implemented language interventions: A meta-analysis. *American Journal of Speech-Language Pathology, 20*, 180–199. https://doi.org/10.1044/1058-0360(2011/10-0055)

Rogers, S. J., & Dawson, G. (2010). *Early Start Denver Model for young children with autism: Promoting language, learning, and engagement*. New York, NY: Guilford Press.

Rogers, S. J., Estes, A., Lord, C., Munson, J., Rocha, M., Winter, J., … Talbot, M. (2019). A multisite randomized controlled two-phase trial of the Early Start Denver Model compared to treatment as usual. *Journal of the American Academy of Child & Adolescent Psychiatry*. https://doi.org/10.1016/j.jaac.2019.01.004

Rogers, S. J., & Vismara, L. A. (2008). Evidence-Based comprehensive treatments for early autism. *Journal of Clinical Child & Adolescent Psychology, 37*, 8–38. https://doi.org/10.1080/15374410701817808

Ryberg, K. H. (2015). Evidence for the implementation of the Early Start Denver Model for young children with autism spectrum disorder. *Journal of the American Psychiatric Nurses Association, 21*, 327–337. https://doi.org/10.1177/1078390315608165

Sabielny, L. M., & Cannella-Malone, H. I. (2014). Comparison of prompting strategies on the acquisition of daily living skills. *Education and Training in Autism and Developmental Disabilities, 49*, 145–152.

Sallows, G. O., & Graupner, T. D. (2005). Intensive behavioral treatment for children with autism: Four-year outcome and predictors. *American Journal on Mental Retardation, 110*, 417–438. https://doi.org/10.1352/0895-8017(2005)110[417:IBTFCW]2.0.CO;2

Schlosser, R. W., & Wendt, O. (2008). Effects of augmentative and alternative communication intervention on speech production in children with autism: A systematic review. *American Journal of Speech-Language Pathology, 17*, 212–230. https://doi.org/10.1044/1058-0360(2008/021)

Schreibman, L., & Stahmer, A. C. (2014). A randomized trial comparison of the effects of verbal and pictorial naturalistic communication strategies on spoken language for young children with autism. *Journal of Autism and Developmental Disorders, 44*, 1244–1251. https://doi.org/10.1007/s10803-013-1972-y

Smith, K. A., Shepley, S. B., Alexander, J. L., & Ayres, K. M. (2015). The independent use of self-instructions for the acquisition of untrained multi-step tasks for individuals with an intellectual disability: A review of the literature. *Research in Developmental Disabilities, 40*, 19–30. https://doi.org/10.1016/j.ridd.2015.01.010

Smith, T. (2001). Discrete trial training in the treatment of autism. *Focus on Autism and Other Developmental Disabilities, 16*, 86–92. https://doi.org/10.1177/108835760101600204

Smith, T., Eikeseth, S., Klevstrand, M., & Lovaas, O. I. (1997). Intensive behavioral treatment for preschoolers with severe mental retardation and pervasive developmental disorder. *American Journal on Mental Retardation, 102*, 238–249. https://doi.org/10.1352/0895-8017(1997)102<0238:IBTFPW>2.0.CO;2

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. https://doi.org/10.1352/0895-8017(2000)105<0269:RTOIEI>2.0.CO;2

Smith, T., & Iadarola, S. (2015). Evidence base update for autism spectrum disorder. *Journal of Clinical Child & Adolescent Psychology, 44*, 897–922. https://doi.org/10.1080/15374416.2015.1077448

Stokes, T. F., & Baer, D. M. (1977). An implicit technology of generalization. *Journal of Applied Behavior Analysis, 10*, 349–367. https://doi.org/10.1901/jaba.1977.10-349

Strain, P. S., & Bovey, E. H. (2011). Randomized, controlled trial of the LEAP model of early intervention for young children with autism spectrum disorders. *Topics in Early Childhood Special Education, 31*, 133–154. https://doi.org/10.1177/0271121411408740

Strain, P. S., & Hoyson, M. (2000). The need for longitudinal, intensive social skill intervention: LEAP follow-up outcomes for children with autism. *Topics in Early*

Childhood Special Education, 20, 116–122. https://doi.org/10.1177/027112140002000207

Sulzer-Azaroff, B., Hoffman, A. O., Horton, C. B., Bondy, A., & Frost, L. (2009). The Picture Exchange Communication System (PECS): What do the data Say? *Focus on Autism and Other Developmental Disabilities, 24*, 89–103. https://doi.org/10.1177/1088357609332743

Sulzer-Azaroff, B., & Mayer, G. R. (1991). *Behavior analysis for lasting change*. New York, NY: Holt, Rinehart & Winston.

Sundberg, M. L., & Partington, J. W. (1998). *Teaching language to children with autism or other developmental disabilities*. Pleasant Hill, CA: Behavior Analysts.

Tachibana, Y., Miyazaki, C., Ota, E., Mori, R., Hwang, Y., Kobayashi, E., … Kamio, Y. (2017). A systematic review and meta-analysis of comprehensive interventions for pre-school children with autism spectrum disorder (ASD). *PLoS One, 12*, 1–28. https://doi.org/10.1371/journal.pone.0186502

U.S. Department of Health and Human Services. (1999). *Mental health: A report of the Surgeon General*. Rockville, MD: U.S. Department of Health and Human Services, Substance Abuse and Mental Health Services Administration, Center for Mental Health Services, National Institutes of Health, National Institute of Mental Health.

Verschuur, R., Didden, R., Lang, R., Sigafoos, J., & Huskens, B. (2014). Pivotal response treatment for children with autism spectrum disorders: A systematic review. *Review Journal of Autism and Developmental Disorders, 1*, 34–61. https://doi.org/10.1007/s40489-013-0008-z

Virués-Ortega, J. (2010). Applied behavior analytic intervention for autism in early childhood: Meta-analysis, meta-regression and dose-response meta-analysis of multiple outcomes. *Clinical Psychology Review, 30*, 387–399. https://doi.org/10.1016/j.cpr.2010.01.008

Virués-Ortega, J., Rodríguez, V., & Yu, C. T. (2013). Prediction of treatment outcomes and longitudinal analysis in children with autism undergoing intensive behavioral intervention. *International Journal of Clinical and Health Psychology, 13*, 91–100. https://doi.org/10.1016/S1697-2600(13)70012-7

Vismara, L. A., & Rogers, S. J. (2010). Behavioral treatments in autism spectrum disorder: What do we know? *Annual Review of Clinical Psychology, 6*, 447–468. https://doi.org/10.1146/annurev.clinpsy.121208.131151

Vladescu, J. C., & Kodak, T. (2010). A review of recent studies on differential reinforcement during skill acquisition in early intervention. *Journal of Applied Behavior Analysis, 43*, 351–355. https://doi.org/10.1901/jaba.2010.43-351

Warren, Z., Veenstra-Vanderweele, J., Stone, W., Bruzek, J. L., Nahmias, A. S., Foss-Feig, J. H., … McPheeters, M. L. (2011). *Therapies for children with autism spectrum disorders* (AHRQ Publication No. 11-EHC029-EF). Rockville, MD: Agency for Healthcare Research and Quality.

Weiss, M. J. (2001). Expanding ABA intervention in intensive programs for children with autism: The inclusion of natural environment training and fluency based instruction. *The Behavior Analyst Today, 2*, 182–186. https://doi.org/10.1037/h0099946

Weitlauf, A. S., McPheeters, M. L., Peters, B., Sathe, N., Travis, R., Aiello, R., ... Warren, Z. (2014). *Therapies for children with autism spectrum disorder: Behavioral interventions update* (AHRQ Publication No. 14-EHC036-EF). Rockville, MD: Agency for Healthcare Research and Quality.

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, NC: The University of North Carolina, Frank Porter Graham Child Development Institute, Autism Evidence-Based Practice Review Group. Retrieved from http://autismpdc.fpg.unc.edu/sites/autismpdc.fpg.unc.edu/files/2014-EBP-Report.pdf

Treatment of ADHD in Individuals With and Without Intellectual Disabilities

31

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Attention-deficit/hyperactivity disorder (ADHD) and intellectual disability (ID) are neurodevelopmental disorders characterized by deficits in academic, social, and vocational functioning (American Psychiatric Association, 2013; Swanson et al., 1998). Attention-deficit/hyperactivity disorder (ADHD) is accompanied by persistent symptoms of inattention, impulsivity, and hyperactivity that originate in childhood and are associated with long-term impairment across several domains (American Psychiatric Association, 2013; Barkley, 2015). Individuals with intellectual disabilities vary in the severity of their adaptive behavior and cognitive impairments, and onset typically occurs within the developmental period prior to age 18 (American Association on Intellectual and Developmental Disabilities, 2018; American Psychiatric Association, 2013). Although neurodevelopmental deficits may vary among the disorders, they typically include a range of specific (e.g., executive functioning deficits) and global impairments (e.g., social skills or intelligence; American Psychiatric Association, 2013).

Neurodevelopmental disorders often co-occur with other psychological symptoms, and comorbidity is thought to be the norm rather than the exception (American Psychiatric Association, 2013; Kutcher et al., 2004). Research examining the comorbidity of ADHD in children with intellectual disabilities report prevalence rates between 6% (Capone, Goyal, Ares, & Lannigan, 2006; Dykens, 2007) and 80% (Frazier et al., 2001). The large range of prevalence rates across studies is likely due to the variability of the sample, and includes children with developmental delays, genetic disorders, sub-average intelligence, pervasive developmental disorders, and complications due to prenatal exposure to ethanol (Barkley, 2015).

Prevalence

ADHD is more prevalent in individuals diagnosed with ID than in the general population (Dekker & Koot, 2003; Hastings, Beck, Daley, & Hill, 2005; La Malfa, Lassi, Bertelli, Pallanti, & Albertini, 2008). Further, the prevalence of ADHD is positively correlated with the severity of cognitive impairment (Voigt, Barbaresi, Colligan, Weaver, & Katusic, 2006). The reported prevalence of ADHD in children with ID ranges from 18 to 40%, which is much higher than the 7–10% reported in the general population (Epstein, Cullinan, & Polloway, 1986; Koller, Richardson, Katz, & McLaren, 1983; Pearson & Aman, 1994).

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Despite the high prevalence rates, research indicates that ADHD remains underdiagnosed in individuals with ID (Fisher, Burd, Kuna, & Berg, 1985). Clinicians tend to emphasize the client's intellectual disability and underemphasize psychopathological symptoms, leading to undiagnosed ADHD in the ID population (Jopp & Keys, 2001; Mason & Scior, 2004; White et al., 1995). Additionally, the comorbidity of ADHD and ID may be overshadowed by the frequency of behavior problems exhibited by these individuals, and, thus, ADHD is not recognized. Overall, research supports the high rate of ADHD symptoms in individuals with intellectual disabilities, which cannot be accounted for by confounding associations with other psychiatric disorders (Hastings et al., 2005; Pliszka, 2009; Simonoff, Pickles, Wood, Gringras, & Chadwick, 2007).

Validity of Comorbid Diagnosis

Comorbidity often complicates the diagnosis and course of treatment, especially for those diagnosed with ADHD and ID. In an attempt to combat the aforementioned overshadowing effect, the American Psychiatric Association (2000) recommended that the diagnosis of ADHD be given in children with ID when the symptoms of inattention, impulsivity, or hyperactivity occur in excess of the child's mental age. Historically, children with ID have been excluded from studies of children with ADHD (Handen, McAuliffe, Janosky, Feldman, & Breaux, 1998), and earlier versions of the DSM had exclusion criteria for a diagnosis of ADHD in individuals with ID (Antshel, Phillips, Gordon, Barkley, & Faraone, 2006). Given the recognition and prevalence of ADHD in individuals with ID (Dekker & Koot, 2003), the lack of valid instruments (Antshel et al., 2006), along with rater bias (Miller, Fee, & Netterville, 2004), has come under scrutiny.

The proper diagnosis of comorbid ID and ADHD is especially important, as these individuals have worse outcome trajectories than those with ID alone (Lambert, Sassone, & Sandoval, 1987; Xenitidis, Paliokosta, Rose, Maltezos, & Bramham, 2010). For example, research has consistently indicated that children with ADHD and ID are more likely to exhibit aggression and noncompliance, and display poorer social skills than individuals with ID alone (Carmeli, Klein, & Sohn, 2007; Johnson, Lubetsky, & Sacco, 1995). Ahuja, Martin, Langley, and Thapar (2013) compared clinical characteristics (ADHD subtypes, total number of symptoms, and common comorbidities) of children with ADHD/mild ID to their non-ADHD counterparts. The authors found that children with ADHD and ID are clinically similar to typically developing children with ADHD, except for a higher incidence of conduct problems in the ID sample (Ahuja et al., 2013). Xenitidis et al. (2010) found that adults with comorbid ID/ADHD displayed more severe ADHD symptoms when compared to those with ADHD alone.

Treatment Implications

There is strong evidence for the use of stimulants in children and adolescents with ADHD and comorbid learning disabilities. However, medication becomes less effective with an increased severity of learning difficulties (Fonagy et al., 2015). Although few studies have focused on the treatment of ADHD in children with intellectual disabilities, some studies have shown that an IQ above 50 predicts a better response to stimulant medication than those with more severe intellectual impairments (Aman, Buican, & Arnold, 2003).

Although ADHD is one of the most common forms of comorbidities in children and adults with ID, they often remain undiagnosed (Ahuja et al., 2013). In part, this neglect is due to poorly developed communication skills, as well as the social disadvantage experienced by individuals with cognitive impairments (Lindsey, 2002). This may also be due to the lack of normative data on ADHD symptoms in an ID population (Ahuja et al., 2013). As such, there is a paucity of research examining the effectiveness of intervention and treatment services with these individuals (Ahuja et al., 2013).

Treatment of ADHD Without ID

Evidence-based treatments for typically developing individuals with ADHD are well established and under continuous refinement (Barkley, 2015). Documentation of positive outcomes have been obtained with medication as well as behavioral treatments. There has been some support for other treatments such as electroencephalographic (EEG) biofeedback, and cognitive-behavioral or self-control training for children with ADHD (Fabiano et al., 2009). However, these treatments are not supported by randomized controlled trial (RCT) studies, which is contrary to medication and behavioral interventions (Fabiano et al., 2009). Therefore, the following descriptions of treatment focuses on well-established pharmacological and behavioral interventions.

Pharmacological Treatments Several studies have supported the use of stimulant medications for improving impulse control, fine motor coordination, sustained attention, and reaction time (Aron, Dowson, Sahakian, & Robbins, 2003; Konrad, Gunther, Hanisch, & Herpertz-Dahlmann, 2004; Solanto, Arnsten, & Castellanos, 2001). Higher doses tend to be associated with more vigorous responses (Arnsten & Rubia, 2012; Swanson et al., 2013). Although side effects are common, stimulant medications are generally well tolerated (Barkley, 2015). Common side effects include decreased appetite, weight loss, insomnia, headache, stomachache, tics, irritability, and slight increases in heart rate and blood pressure (Barkley, 2015; Martinez-Raga, Knecht, Szerman, & Martinez, 2013; Olfson et al., 2012). In addition, the frequency of side effects increases as medication dosages increase (Meaux, Hester, Smith, & Shoptaw, 2006).

Common stimulant medications used in the treatment of ADHD symptoms include methylphenidate and amphetamine (Barkley, 2015). These medications work by affecting the central nervous system to address ADHD symptoms and are either short acting (effective for up to four hours) or extended release (effective for up to 12 hours; Fonagy et al., 2015). A meta-analysis done by Faraone and Buitelaar (2010) examined 23 double-blinded, placebo-controlled studies of stimulant use for children and adolescents with ADHD. This study compared the effectiveness of methylphenidate and amphetamine on ADHD symptoms and found that amphetamine had slightly higher effect sizes than methylphenidate, suggesting that amphetamine products may be more efficacious than methylphenidate (Faraone & Buitelaar, 2010). Another meta-analysis examined the benefits and safety of medication for individuals with ADHD. The authors found that when accounting for efficacy and safety, the use of methylphenidate was preferred for children and adolescents, while amphetamines were preferred for adults for short-term treatment of ADHD symptoms. Results of this study suggested that individual factors, including age and course of treatment, are important considerations while implementing stimulant medications for ADHD symptom management (Cortese et al., 2015).

Within the past 15 years, nonstimulant medications for ADHD have been developed, tested, and approved by the FDA. Nonstimulant medications include atomoxetine, clonidine extended release (Clon-ER), and guanfacine extended release (GXR; Jain, Segal, Kollins, & Khayrallah, 2011; Kollins et al., 2011; Tanaka, Rohde, Jin, Feldman, & Upadhyaya, 2013). These medications are intended to be administered once or twice per day and generally have a longer duration of action than stimulant medications (Barkley, 2015). In addition, children with ADHD have an initial response rate of between 65 and 75% for any single medication, and trying a second medication for initial non-responders may increase a positive response rate to 80-90% (Barkley, 2015).

Nonpharmacological Treatments There are a number of empirically supported behavioral interventions that effectively aid in the management of ADHD symptoms (Chacko et al., 2009; Evans, Owens, Wymbs, & Ray, 2018; Pfiffner et al., 2016). Behavioral interventions are typically structured in settings where impairment occurs and often includes home, school, and community environments (Barkley, 2015). Therapeutic benefits have also been found within

specialty settings, such as summer treatment programs, that include intensive behavioral interventions (Pelham Jr & Fabiano, 2008).

Classroom management and behavioral parent training (BPT) are also effective and well supported by research for treating children and adolescents with ADHD (Pelham Jr, Wheeler, & Chronis, 1998). Since parents are essential for decision-making and advocacy of their child's assessment and treatment needs, they play a pivotal role in their children's functioning across domains (Barkley, 2015). Parent training generally includes psychoeducation on ADHD, functional assessment, giving effective instructions, increased attention to children's positive behavior, and praise for compliance and desired behavior. Parent training may include establishing an incentive system (e.g., token economy) and using timeout for noncompliant, disruptive, and aggressive behavior (Barkley, 2015; Chacko et al., 2009).

Behavioral interventions, which increase collaboration between parents, teachers, and the child, provide a comprehensive approach and often result in optimal outcomes across settings for children and adolescents with ADHD (Pelham Jr & Fabiano, 2008; Pfiffner et al., 2016). A study conducted by Pfiffner et al. (2016) provided support for use of the Child Life and Attention Skills (CLAS) program, an effective intervention integrating psychosocial treatment across home and school settings. The results revealed that integration of the intervention among parent, teacher, and child is superior to parent training alone, especially for youth with predominantly inattentive symptom presentation (Pfiffner et al., 2016).

Evans et al. (2018) examined evidence-based psychosocial treatments for children and adolescents with ADHD and encouraged the consideration of developmental level when choosing an effective psychosocial intervention. For example, BPT is a well-established form of treatment for ADHD but has proven more effective within a preschool and elementary population (Evans et al., 2018). Study results also concluded that behavioral classroom management intervention, as well as combined behavior management interventions, were optimally effective for preschool and elementary aged children (Evans et al., 2018). In addition, training in organizational skills were the most effective intervention for elementary school children and adolescents, and behavioral peer interventions were maximally effective for elementary school youth with ADHD (Evans et al., 2018).

Combined Treatment Approaches Combined psychosocial and pharmacological treatment is frequently studied, with one of the most prominent series of studies originating from the NIMH collaborative Multimodal Treatment Study of Children with Attention-Deficit/Hyperactivity Disorder (MTA; Arnold et al., 1997; Jensen et al., 1999). One aim of this research was to examine short-term effectiveness of medication and behavioral interventions in the treatment of ADHD. The MTA was designed with random assignment of children (N = 571) to four treatment groups: behavior modification alone, medication alone, the combination of behavior modification and medication, and community comparison. Initial conclusions from the MTA suggested that medication alone and the combination treatment were the most effective for treating ADHD and related problems. However, Schwarz (2013) later questioned this conclusion and reported that the combined treatment was superior to medication alone when considering all outcomes for those with ADHD. The broader literature has continued supporting the use of combined behavioral and medication intervention for optimal improvement of academic performance, oppositional and aggressive behaviors, internalizing symptoms, social skills, and parent-child relations for children and adolescents with ADHD (Barkley, 2015; Majewicz-Hefley & Carlson, 2007; Van der Oord, Prins, Oosterlaan, & Emmelkamp, 2008).

Further, Fabiano et al. (2007) examined the importance of the medication dose-response curve in combination with psychosocial interventions. The results supported the use of low doses due to the dose-response curve of behavioral intervention. Specifically, lower doses of stimulant medication resulted in similar adjustment outcomes as high-level behavioral interventions, with fewer side effects than what is typically experienced with psychopharmacological medications for ADHD (Fabiano et al., 2007).

The current literature generally lacks studies exploring the combined treatment outcomes for ado-

lescents and adults. Similarly, there is a lack of research focused on treating ADHD as a lifelong, chronic disorder. The majority of studies focus on the short-term intervention effects, without studying the long-term effects. In addition, since ADHD is often comorbid with other disorders (such as ID), future research is essential in understanding optimal outcomes for those experiencing more severe developmental deficits. The remainder of this chapter will focus on the limited research addressing treatment options for those experiencing comorbid ID/ADHD.

Treatment of Comorbid ID/ADHD

Medication

Of the few studies evaluating the treatment of comorbid ID/ADHD, the majority of studies have examined the effectiveness of drug therapy, and, almost exclusively, stimulant medication (Handen, Sagady, & McAuliffe-Bellin, 2009; Handen et al., 1992; Simonoff et al., 2013; Handen, McAuliffe, & Caro-Martinez, 1996; Aman et al., 2003; Pearson et al., 2004a; Pearson et al., 2004b; Handen, Feldman, Lurier, & Murray, 1999). In addition, research has demonstrated some support for the use of atomoxetine and risperidone in treating this population (Fernández-Jaén, Fernández-Mayoralas, Pérez, Jareño, & Díaz, 2010; Correia Filho et al., 2005).

The use of medication with individuals with ID/ADHD typically beings with a health evaluation by a medical doctor and, at times, includes an evaluation by a psychologist. Once prescribed, the medication is taken consistently and monitored for effectiveness and side effects by the prescribing physician. The following table summarizes research findings on the effects of medication in individuals with ID/ADHD supporting medication's effectiveness across numerous outcome variables.

Psychopharmacological Research in Individuals with ADHD and ID

Citation	Medication	Sample	Outcome
Handen et al., 2009	Methylphenidate	40 children ages 6–13 with moderate ID to borderline intellectual functioning (23 of which had comorbid ADHD)	Social/play behavior improved
Handen et al., 1992	Methylphenidate	12 children ages 6–9 with IQs of 50–74 and comorbid ADHD	Improved ADHD symptoms, class work, on-task behavior, and attention skills
Simonoff et al., 2013	Methylphenidate	122 children ages 7–15 with comorbid ADHD and IQ of 30-69	Reduced ADHD symptoms
Handen et al., 1996	Methylphenidate	44 children ages 6–13 with IQ range of 44–77 and comorbid ADHD	Improved attention and activity; improved work completion (only for higher doses) and accuracy (limited to the lower doses in the weekday classroom)
Aman et al., 2003	Methylphenidate	90 children ages 4–17 with IQs ranging from untestable to 90 and comorbid ADHD	Improved attention, overactivity, and conduct; improved performance on some cognitive tests
Pearson, Lane, et al., 2004; Pearson, Santos, et al., 2004	Methylphenidate	24 children with mild to moderate ID and comorbid ADHD	Improved behavior and cognitive performance
Handen et al., 1999	Methylphenidate	Children ages 4–5 with IQ ranging from 40 to 78 and comorbid ID	Improved ADHD symptoms and play
Correia Filho et al. (2005)	Methylphenidate versus risperidone	46 children ages 6–16 with ADHD and low IQ (referred to as MR in this article)	Improvements for both methylphenidate and risperidone; risperidone more effective than methylphenidate
Fernández-Jaén et al., 2010	Atomoxetine	48 children ages 5–19 with ADHD and ID	Decreased ADHD symptoms

Methylphenidate

As seen in the abovementioned table, methylphenidate is the most commonly prescribed medication for individuals with ID/ADHD. Research on the treatment of ADHD in individuals with ID with methylphenidate indicate positive improvements in a variety of domains. Specifically, in samples of children with ID/ADHD, methylphenidate has been associated with improvements in ADHD symptoms (Handen et al., 1992; Handen et al., 1996; Pearson, Lane, et al., 2004; Pearson, Santos, et al., 2004; Simonoff et al., 2013), cooperative play, class work, and task engagement (Handen et al., 2009). Moreover, research has demonstrated that methylphenidate provides benefits in children as young as preschool age (Handen et al., 1999).

There is mixed evidence pertaining to the relationship between the dose of methylphenidate and positive outcomes for children and adolescents with ID/ADHD. Pearson and colleagues (Pearson, Lane, et al., 2004; Pearson, Santos, et al., 2004) found that on average cognitive performance across tasks improved more at higher doses of stimulant medication. Notably, the results suggested that improvements resulting from stimulant medication in behavior and cognitive domains were unrelated to one another (Pearson, Lane, et al., 2004; Pearson, Santos, et al., 2004). For example, Handen et al. (1992) reported positive results for class work, ADHD symptoms, attention skills, and being on-task. These improvements were seen only at the higher doses of methylphenidate, and there were no improvements in social behavior or learning (Handen et al., 1992). In contrast, Handen et al. (1996) found improvements in work completion at higher doses but improvements in work accuracy at lower doses.

Simonoff et al. (2013) conducted one of the largest studies on the effectiveness of methylphenidate for children with ID/ADHD. The study was unique in that medication was titrated and the participants were followed for a longer period of time (16 weeks) than is typically done. Consistent with previous research, the results included reductions in ADHD symptoms; however, there were no significant moderators of these effects when examining IQ, ASD, and severity level of ADHD (Simonoff et al., 2013).

Despite the positive outcomes of medication use in individuals with ID/ADHD, the outcomes are seen in only 40–50% of individuals, which is lower than that seen in typically developing children with ADHD (i.e., 70–80%; Tarrant et al., 2018). Although the effectiveness rates are lower relative to individuals with ADHD alone, using stimulant medication has been associated with a number of positive results in research over the past few decades.

Atomoxetine

Atomoxetine has demonstrated positive effects in children with ID/ADHD, with effectiveness rates comparable to those seen in individuals without ID; however, the effectiveness of atomoxetine was lower than that found with methylphenidate in typically developing children with ADHD (Fernández-Jaén et al., 2010). Moreover, Mazzone, Reale, Mannino, Cocuzza, and Vitiello (2011) found the effects of atomoxetine on children with ADHD was inversely related to IQ. That is, the medication was increasingly more effective as children's intellect increased (Mazzone et al., 2011). Their study did not specifically conduct research in the ID population, but rather examined effectiveness of atomoxetine in 55 children ages 5–15 with ADHD and IQs ranging from 43 to 117 (Mazzone et al., 2011). Yet, the findings are meaningful in that individuals with IQs less than 85 had lower response rates (Mazzone et al., 2011). Additionally, in a study by Fernández-Jaén et al. (2010), a subset of individuals with ID/ADHD experienced side effects when taking atomoxetine. In summary, atomoxetine has demonstrated positive effects, but the empirical support specific to an ID/ADHD population is scarce; hence, additional research is needed to better understand the appropriateness of treatment with atomoxetine in individuals with ID/ADHD.

Risperidone

Risperidone is an anti-psychotic medication widely used in treating individuals with schizophrenia, bipolar disorder, and ASD, and is often prescribed to individuals with ID/ADHD or ID with conduct problems (Thomson, Maltezos, Paliokosta, & Xenitidis, 2009). Research on the effectiveness of risperidone for individuals with ID/ADHD is very limited. One study conducted by Correia Filho et al. (2005) found that risperidone was more effective than methylphenidate (Correia Filho et al., 2005). Notably, this study did not have a control group (Correia Filho et al., 2005). Other researchers have indicated that individuals with ID/ADHD may benefit from taking the combination of risperidone and stimulant medication (Aman, Binder, & Turgay, 2004). Conversely, in a meta-analysis, Thomson et al. (2009) concluded that the use of risperidone in individuals with ID/ADHD has limited support due to the quality of available research (i.e., inconsistent participant inclusion criteria and lack of RCT studies). Moreover, Fonagy et al. (2015) warned against the use of risperidone in children with ADHD, suggesting that unlike methylphenidate and atomoxetine, if anti-psychotics are used, it is recommended only for brief durations (Fonagy et al., 2015).

Despite a lack of evidence that risperidone is effective with individuals with ID/ADHD (Thomson et al., 2009), the medication is commonly prescribed for individuals with ID. The current state of literature lacks RCT studies supporting the use of risperidone with individuals with comorbid ADHD and ID (Thomson et al., 2009). Clearly much more research is needed comparing the effectiveness and side effects of risperidone in comparison to stimulant medication.

Medication Considerations

Side Effects One consideration of pharmacological treatment in individuals with ID/ADHD is the increased risk of side effects relative to typically developing children with ADHD. Handen, Feldman, Gosling, Breaux, and McAuliffe (1991) examined the side effects of methylphenidate in children with ID/ADHD, researching 13 potential areas of impairment: tics, drowsiness, sadness, staring, withdrawal, irritability, reduced appetite, anxiety, dizziness, mood, activity, stomach distress, and headaches. The authors found that the use of methylphenidate was associated with motor tics and social withdrawal in children with ID/ADHD (Handen et al., 1991). Other research reported increased sleep problems, reduced appetite, weight loss, staring, and drowsiness (Simonoff et al., 2013; Handen et al., 1992). Individuals with ID/ADHD treated with atomoxetine experienced side effects that included irritability, sleep problems, stomach discomfort, and disordered eating (Fernández-Jaén et al., 2010). Lastly, the side effects of risperidone include uncontrollable body movements and weight gain (Correia Filho et al., 2005)

Limitations of Effectiveness Several variables have been associated with the effectiveness of medication in individuals with ID/ADHD. For example, severity of cognitive impairment appears to moderate the effectiveness of methylphenidate (Aman et al., 2003), although this finding is inconsistent across studies (Simonoff et al., 2013). Mazzone et al. (2011) found that the effectiveness of atomoxetine was negatively associated with intelligence. Further, researchers have suggested that higher doses lead to more positive results (Simonoff et al., 2013). To complicate matters further, higher doses of medication may increase the severity of side effects (Simonoff et al., 2013; Handen et al., 1999).

Future Psychopharmacological Research

Much of the research that supports the use of pharmacological treatment for individuals with ID/ADHD is now dated. In addition, the majority of literature on this population in the last decade focuses on systematic reviews and meta-analyses, all concluding that more research is needed

(Courtenay & Elstner, 2016; Reilly & Holland, 2011; Tarrant et al., 2018; Thomson et al., 2009). Given the medical advancements (i.e., increased availability of various medications accessible now), there is a need for more research conducted to determine the most appropriate medication for this population, taking into consideration effectiveness as well as side effects. The primary measure of improvements within the available research involves parent and teacher behavior ratings, but there is a need for additional measures of objective improvements. Additionally, across medications, the research is largely lacking for adults with ID/ADHD. Researchers may also consider further exploring the potential of combining treatment approaches, whether that includes medication combinations (methylphenidate and atomoxetine) or psychosocial treatments in combination with medication to treat this population.

Nonpharmacological Treatments

There are several important considerations that should be taken into account when selecting a nonpharmacological treatment regimen for those with ADHD and comorbid disorders. These include ADHD symptom severity, comorbid disorder(s), environmental factors that may hinder treatment efficacy, cognitive functioning, age, and other practical factors (e.g., treatment cost and time to implement the treatment; Brown, 2009).

There are also unique factors to consider when planning treatment for adults with ID/ADHD. For example, researchers have found that subjective well-being is linked with treatment efficacy in individuals with ID, as well as typically developing adults. Subjective well-being is defined as one's overall happiness, satisfaction, or feeling of contentment with life (Araten-Bergman, 2015). In one study, the author examined factors related to adults with ID/ADHD's sense of well-being (Araten-Bergman, 2015). This study found that adults with ID/ADHD reported a greater sense of well-being when their basic needs were met, such as medical care, stable living conditions, and source of income (Araten-Bergman, 2015). However, the severity and impairment of one's ADHD symptoms and functional impairment diminished their feelings of subjective well-being. This was mediated by their psychosocial factors, such as social support and vocational skills, suggesting that treatment should focus on creating and cultivating support systems and skills development in adult populations (Araten-Bergman, 2015).

Psychosocial Treatments

Despite the proliferation of research on individuals without cognitive impairment, there are few studies examining the impact of psychosocial treatments for individuals with ID/ADHD. As such, information is sparse on how established, psychosocial treatments can be adapted for individuals with ID/ADHD. One hindrance to this is the large variability in individuals' IQs and adaptive functioning levels in those with ID (Thapar, 2017). Second, researchers have just begun to recognize the comorbidity of ID/ADHD. However, recent studies have found that adults with comorbid ID/ADHD showed greater deficits in attention and response inhibition than those with ADHD alone, even after controlling for intelligence. This suggests that individuals with ID demonstrated more severe and impairing ADHD symptoms, even after controlling for attention symptoms, which cannot be accounted for by their intellectual disability (Araten-Bergman, 2015).

The inclusion of individuals with ID in treatment research has been limited (Thapar, 2017; Rowles & Findling, 2010). Many therapeutic trials excluded those with an IQ of 80 or below (McBrien, Turk, & Letch, 2006). With the fifth edition of the DSM, researchers have called for the inclusion of those with ID to be included in treatment studies, as the research has shown many commonalities with other neurodevelopmental disorders (i.e., ADHD and ASD; Thapar, 2017). Many researchers have called for further research of psychosocial treatments with children with ID/ADHD, especially given the possible lack of response to medication, side effects, and

opportunity to provide them with more than one treatment option (Huang & Ruedrich, 2007; Rowles & Findling, 2010).

Behavioral Parent Training (BPT) Although BPT primarily has been provided to parents with typically developing children, there are an increasing number of studies evaluating the effectiveness of BPT with parents of children with ID/ADHD. Parent training with parents of children with ID/ADHD include many of the techniques taught to parents of children with ADHD alone (e.g., providing clear instructions, contingent praise, using timeout). Additionally, parents of children with ID, with or without ADHD, are often taught skills for shaping behavior, as well as backward and forward chaining.

Stepping Stones Triple P (SSTP) is a BPT program designed specifically for parents of children with disabilities. SSTP is a multilevel family intervention designed to target challenging behavior that hinders learning and increases parental stress. The main goal of SSTP is to provide parents with skills for interacting more positively and warmly with their children, and to increase parenting confidence (Roux, Sofronoff, & Sanders, 2013).

Due the unique challenges that families of children with disabilities face (e.g., new skill development, managing difficult behavior, and parental stress), the authors created a program that addressed a variety of disabilities. The authors believe that parents of children with varying behavior disorders may differ in their behavior problems; however, all challenging behaviors are similarly maintained by environmental stimuli. The intervention focuses on understanding the learned, bidirectional relationship between the parent and child that is currently maintaining the child's dysfunctional behavior. It then replaces the dysfunctional relationship with alternative positive parenting (Sanders, 1999). Overall, the standard SSTP program research has shown that the program reduces problematic behaviors in children with IDs, as well as increase parenting confidence (Roux et al., 2013).

Other parenting programs include the Incredible Years program. This program has also been cited as a successful intervention for children with ID. The Incredible Year program encompasses many of the same components as other parenting program packages (e.g., increasing positive parenting, problem-solving, communication skills, and contingency management), but adds the focus of the collaborative parent-teacher relationship (Jones, Daley, Hutchings, Bywater, & Eames, 2007, 2008). This program utilizes a collaborative approach to teach skills through the use of videotape modeling and role-playing (Chronis, Chacko, Fabiano, Wymbs, & Pelham, 2004).

Family-Based Intervention

There is evidence that participation and engagement in family activities, such as routines, play activities, and family gatherings, provide great benefit for child development (Bronfenbrenner, 1995; Dunst et al., 2001; Axelsson, Granlund, & Wilder, 2013). Axelsson et al. (2013) found that children with profound ID and multiple disabilities engaged in family activities less often than typically developing children. Further, they found that engagement in certain family activities (e.g., watching movies together, playing with pets, story reading, having dinner together, going to the playground or library) were positively correlated with health, cognition, and communication skills in children with profound ID. Of note, cognition and engagement were most strongly correlated in unstructured, child-driven activities (Axelsson et al., 2013). As such, children with ID/ADHD may benefit from adapting family living patterns and routines to child-driven activities in order to increase child engagement and appropriately account for the child's intellectual and cognitive level.

In a follow-up study, Axelsson, Imms, and Wilder (2014) identified several strategies for increasing social engagement in adolescents with profound ID and comorbid disorders. These strategies included availability and acceptability of the activity, adequate knowledge of the child, a posi-

tive attitude towards working with the child, and the child/adolescents' sense of belonging and opportunity to influence the activity (Axelsson et al., 2014). Although the use of participation-facilitating strategies may be helpful in increasing engagement in children and adolescents with ID, further research in this area is necessary to test the efficacy of these strategies in clinical trials.

School-Based Interventions

Researchers suggest that interventions for ADHD with comorbid disorders should be done in the context of applied behavior analysis (ABA), regardless of the student's age or intellectual abilities. ABA is used to structure interventions for treating children with ID/ADHD. Although originally developed for children with ASD, numerous studies have shown favorable results for the treatment of autism, as well as children with ID (Matson et al., 2012). ABA involves the use of operant conditioning methods and focuses on identifying antecedents and consequences of behavior, and focuses on breaking the chain of events to teach new behaviors. Determining antecedents and consequence of a problematic behavior leads directly to formulating treatment for decreasing problematic behavior and increasing positive behavior.

Positive Behavior Support There are several school-based and teacher-led interventions that have been proven effective at managing ADHD symptoms and increasing skill acquisition in individuals with ID. The use of positive behavior support (PBS), an intervention that utilizes educational methods to promote positive behavior and render problem behavior inefficient and ineffective (Carr et al., 2002), has demonstrated efficacy in managing behavior problems in children. There is a plethora of research supporting the use of PBS in the classroom (Bambara & Kern, 2005; Carr et al., 2002; Cooper, Heron, & Heward, 1987; Touchette, MacDonald, & Langer, 1985; Dunlap, Kern-Dunlap, Clarke, & Robbins, 1991). Children with ID benefit greatly from such interventions in managing behavior problems, including those related to ADHD (i.e., inattention, hyperactivity, and impulsivity), and in developing necessary life skills (Dunlap, Iovannone, Wilson, Kincaid, & Strain, 2010).

Procedures related to PBS are informed by ABA principles and often include conducting a functional behavioral assessment (FBA). This process involves understanding the behavioral patterns associated with contextual triggers, antecedents, and consequences, as well as the development of a behavioral support plan based on data collected during the FBA (Dunlap et al., 2010). In addition, the behavioral support plan entails individualizing intervention elements specific to the child.

Dunlap et al. (2010) developed a model of PBS intervention, Prevent-Teach-Reinforce (PTR), targeting challenging behavior problems in various classroom settings. The PTR model is a step-by-step program for developing and successfully employing an intervention implemented by school personnel. PTR can also be an effective intervention for children with ID and a comorbid disorder. This intervention is intended for use in general or special education classrooms. This evidence-based approach has been implemented with fidelity with young children (Strain, Wilson, Wilson, & Dunlap, 2011; Sears, Blair, Iovannone, & Crosland, 2013), and has been effective in decreasing challenging behavior in children with ID/ADHD.

Technology in the Classroom The use of mobile and computer technologies for facilitating skill acquisition in children with ID is emerging (Ayres, Mechling, & Sansosti, 2013; Goldsmith & LeBlanc, 2004). For a review about some of these emerging technologies, see Ayres et al. (2013). Technology devices allow children to learn from a tactile, hands-on approach, and students with ID often prefer this learning modality (Shane & Albert, 2008). It is likely that children with ID/ADHD would benefit from integrating mobile and computer-based learning in the classroom. As such, parents and teachers may consider the implementation of such technologies to aid in classroom instruction and to be used at home. Technologies (i.e., instructional and assis-

tive) can be implemented in the school environment through a students' individualized education plan, as covered under the Individuals with Disabilities Education Improvement Act (2004).

Research demonstrates that these approaches have been beneficial in teaching life skills (e.g., cooking, vocational skills, and pedestrian travel) and making improvements in self-efficacy (i.e., promotion of self-management and self-instructions). Several studies have assessed the implementation of video-based instruction with children with ID. Modalities of these interventions range from use of DVD, personal digital assistants (PDAs), iPod, and mobile phones. For example, Mechling et al. (2008) used video-based instruction through a portable DVD player to teach cooking skills to a group of young adults with moderate ID. Video modeling has also been used to instruct vocational tasks (Van Laarhoven, Johnson, Van Laarhoven-Myers, Grider, & Grider, 2009) and assist with pedestrian travel (Mechling & Seid, 2011) among individuals with ID. Additionally, Cihak, Kessler, and Alberto (2007, 2008) found a hand-held prompting system, which included the use of pictures and audio instructions, was successful at improving independent transition between tasks in students with ID. It is likely that individuals with ID/ADHD would similarly benefit from the use of such technologies to address relevant symptoms.

Skills Training

Organizational skills training can be an essential intervention for adolescents and adults with ADHD. This typically includes skills training in time management, prioritizing and setting goals, meeting deadlines, developing routines, and managing tasks of daily living. An important aspect of this intervention is teaching individuals to utilize checklists and planners to manage assignments, deadlines, and other tasks. Concrete organization skills are taught, and may include how to effectively manage one's time through creating a to-do list, planning out their day based on tasks that need to be completed, and checking items off once completed. For those with ID/ADHD, this could be utilized by helping them create a checklist of a task, such has how to complete some daily living activity. A meta-analysis study showed that organizational skills training effectively improved organizational skills, academics, and inattention deficits (Bikic, Reichow, McCauley, Ibrahim, & Sukhodolsky, 2017). However, this intervention has not yet been examined in populations with ID/ADHD.

Cognitive Training Executive functioning (EF) deficits, such as those related to attention, task-switching, inhibition, and working memory, are characteristic of individuals with ADHD (Pennington & Ozonoff, 1996). These deficits are associated with lower academic achievement and language development, as well as reduced behavioral stability (Barkley, 1997; Biederman et al., 2004), and are likely exacerbated in children with learning problems and ID (Kirk, Gray, Riby, & Cornish, 2015). In recent years, there has been a surge of research suggesting that typically developing children and those with ADHD demonstrate improvements in working memory (for review, see: Cortese et al., 2015; Kirk et al., 2015). This research is based on evidence of brain plasticity and the assumption that neural networks can be strengthened through controlled training trials (Vinogradov, Fisher, & de Villers-Sidani, 2012). Although more research is required to understand the long-term benefits of cognitive training in children with ADHD, it appears that training efforts are effective at improving targeted neuropsychological processes (Cortese et al., 2015).

There has been some evidence that cognitive training supports improved neuropsychological functioning in children with comorbid learning problems and ADHD. Promising results from Gray et al. (2012) demonstrated improvements in working memory in learning disabled adolescents diagnosed with ADHD using a computerized working memory training program. Research assessing the efficiency of cognitive training in individuals with ID/ADHD is limited, but results are promising. Kirk et al. (2015) published a review summarizing the potential application of EF interventions for children with ID.

Söderqvist, Bergman Nutley, Ottersen, Grill, and Klingberg (2012) conducted one of the few studies on cognitive training programs for children with ID. The authors found that the participants demonstrated improvements in verbal working memory and language functioning 5 weeks after the training program; however, improvements were not sustained after 1 year, suggesting that perhaps intensity and frequency of the intervention may require adaptation to see long-term benefits in children with ID (Söderqvist et al., 2012). Additionally, intervention gains varied significantly between participants. Factors associated with the highest levels of improvement included female gender, single-diagnosis, and higher baseline verbal working memory capacity (Söderqvist et al., 2012).

Kirk et al. (2015) recommended that current cognitive training interventions be adapted for children with ID by using short, frequent training intervals to ensure that children can sufficiently attend to and engage with the training material. They also highlight the importance of providing frequent feedback and reinforcement during the intervention. Furthermore, Cortese et al. (2015) suggested that training approaches targeting multiple neuropsychological processes may show the best outcomes in terms of transferring skills to everyday activities. Although further research is required to successfully adapt cognitive training programs to the needs of children and adolescents with comorbid ID and ADHD, this is a promising area of intervention.

Other Treatment Options

Neurofeedback There is some evidence to support the use of neurofeedback as a treatment option for those with ID/ADHD. Neurofeedback is a behavioral intervention that enhances self-regulation through brain activity (Arns, Heinrichc, & Strehl, 2014). Electrodes are used to measure brain activity and provide the individual with feedback (Breteler, Pesch, Nadorp, Best, & Tomasoa, 2012). The ultimate goal is to increase or decrease the specific brain activity based on the individual's symptoms. It is thought that by targeting specific brain patterns associated with the individual's symptoms, there will ultimately be a change in brain activity and cognitive functioning in order to produce symptom reduction. Typically, neurofeedback interventions include neurofeedback training where the individual engages in activities, such as completing puzzles or listening to music, where the person's brain activity is initially measured and modulated throughout sessions.

Since the literature has shown that neurofeedback has positive outcomes in those with ADHD (i.e., increase attention and improve impulsivity; Arns, Heinrichc, & Strehl, 2014), researchers have begun to evaluate the use of neurofeedback with individuals with mild ID (i.e., IQ between 70 and 50) and ADHD. Breteler et al. (2012), evaluated the efficacy of neurofeedback in children with ID/ADHD living in residential setting. The researchers found that attention improved as did task concentration; however, there appeared to be minimal, if any, impact on impulse control (Breteler et al., 2012). This is the first intervention with children with ID/ADHD, and thus results should be interpreted with caution.

Multimodal There is some evidence that multimodal treatment approaches (e.g., pharmacological and behavioral interventions simultaneously) are more effective than unimodal interventions for individuals with ADHD (Hinshaw, Arnold, & MTA Cooperative Group, 2015. The differential benefit of multiple versus single interventions has only been minimally examined in individuals with ID/ADHD. A meta-analysis of intervention effects on treating challenging behavior (e.g., tantrum behavior, hyperactivity) in individuals with ID indicated they were equally effective (Heyvaert, Maes, & Onghena, 2010).

Summary

Research on the treatment of ID/ADHD is scarce. The most well-established treatment is medication, with a primary focus on methylphenidate within pharmacological treatment of individuals

with ID/ADHD. Methylphenidate has been associated with improvements across multiple domains. Additionally, research has demonstrated reductions in ADHD symptoms with the use of atomoxetine in children with ID/ADHD. In a similar way, behavior and parenting programs have resulted in improvements for individuals with ID/ADHD. Overall, the primary treatment approaches include medication and behavioral treatment; however, more research is needed to further support and expand treatment of individuals with ID/ADHD.

Limitations and Considerations

Although there are several existing treatment options, there are numerous limitations. First, there is limited research on the effectiveness and feasibility of the treatment options done in this comorbid population. Further, there is limited research of the spectrum of ID and many studies are done with individuals with an IQ 80 or above and few with 70 or above. Second, much of the research that has been done has focused on children and families and there has been limited research on adult populations. Lastly, there is much more research that needs to be done on this comorbid population in order to better understand their symptom presentation and proper treatment options.

In addition to the limited evidence-based treatment approaches available to treat ADHD in persons with ID, there are considerations for the available treatments. First, due to the cognitive functioning of these individuals, there may be problems with informed consent (Courtenay & Elstner, 2016). This is particularly problematic given the limited information available help make informed decisions based on empirical support, whether it would be the individuals or their caretakers making the decisions. Another concern is the side effects associated with the medication recommended for treatment in those with ADHD and ID. Moreover, it is possible that there is a negative relationship with severity of ID and effectiveness of treatment within this population.

References

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., Buican, B., & Arnold, L. E. (2003). Methylphenidate treatment in children with borderline IQ and mental retardation: Analysis of three aggregated studies. *Journal of Child and Adolescent Psychopharmacology, 13*(1), 29–40.

Aman, M. G., Binder, C., & Turgay, A. (2004). Risperidone effects in the presence/absence of psychostimulant medicine in children with ADHD, other disruptive behavior disorders, and subaverage IQ. *Journal of Child and Adolescent Psychopharmacology, 14*(2), 243–254.

American Association on Intellectual and Developmental Disabilities (2018). Definition of intellectual disability. Retrieved from http://aaidd.org/intellectual-disability/definition#.We4Egkdrx-U.

American Psychiatric Association. (2000). *Diagnostic and statistical manual of mental disorders, 4th edition, text revision (DSM-IV-TR)*. Washington, DC: Author.

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

Antshel, K. M., Phillips, M. H., Gordon, M., Barkley, R., & Faraone, S. V. (2006). Is ADHD a valid disorder in children with intellectual delays? *Clinical Psychology Review, 26*(5), 555–572.

Araten-Bergman, T. (2015). The subjective well-being of individuals diagnosed with comorbid intellectual disability and attention deficit hyperactivity disorders. *Quality of life research, 24*(8), 1875–1886.

Arnold, L. E., Abikoff, H. B., Cantwell, D. P., Connors, C. K., Elliott, G., Greenhill, L. L., et al. (1997). National Institute of Mental Health Collaborative Multimodal Treatment Study of Children with ADHD (the MTA): Design challenges and choices. *Archives of General Psychiatry, 54*, 865–870.

Arns, M., Heinrich, H., & Strehl, U. (2014). Evaluation of neurofeedback in ADHD: the long and winding road. *Biological psychology, 95*, 108–115.

Arnsten, A. F., & Rubia, K. (2012). Neurobiological circuits regulating attention, cognitive control, motivation, and emotion: disruptions in neurodevelopmental psychiatric disorders. *Journal of the American Academy of Child & Adolescent Psychiatry, 51*(4), 356–367.

Aron, A. R., Dowson, J. H., Sahakian, B. J., & Robbins, T. W. (2003). Methylphenidate improves response inhibition in adults with attention- deficit/hyperactivity disorder. *Biol Psychiatry, 54*, 1465–1468.

Axelsson, A. K., Granlund, M., & Wilder, J. (2013). Engagement in family activities: a quantitative, comparative study of children with profound intellectual and multiple disabilities and children with typical development. *Child: Care, Health and Development, 39*(4), 523–534.

Axelsson, A. K., Imms, C., & Wilder, J. (2014). Strategies that facilitate participation in family activities of children and adolescents with profound intellectual and multiple disabilities: Parents' and personal assistants' experiences. *Disability and Rehabilitation, 36*(25), 2169–2177.

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 disabilities and/or Autism Spectrum Disorders: Considerations for the future of school psychology. *Psychology in the Schools, 50*(3), 259–271.

Bambara, L., & Kern, L. (Eds.). (2005). *Individualized supports for students with problem behaviors: Designing positive behavior plans*. New York, NY: Guilford.

Barkley, R. A. (1997). Behavioral inhibition, sustained attention, and executive functions: constructing a unifying theory of ADHD. *Psychological Bulletin, 121*(1), 65.

Barkley, R. A. (Ed.). (2015). *Attention-deficit/hyperactivity disorder: A handbook for diagnosis and treatment* (4th ed.). New York, NY: Guilford Press.

Biederman, J., Monuteaux, M. C., Doyle, A. E., Seidman, L. J., Wilens, T. E., Ferrero, F., & Faraone, S. V. (2004). Impact of executive function deficits and attention-deficit/hyperactivity disorder (ADHD) on academic outcomes in children. *Journal of Consulting and Clinical Psychology, 72*(5), 757.

Bikic, A., Reichow, B., McCauley, S. A., Ibrahim, K., & Sukhodolsky, D. G. (2017). Meta-analysis of organizational skills interventions for children and adolescents with Attention-Deficit/Hyperactivity Disorder. *Clinical psychology review, 52*, 108–123.

Breteler, R., Pesch, W., Nadorp, M., Best, N., & Tomasoa, X. (2012). Neurofeedback in residential children and adolescents with mild mental retardation and ADHD behavior. *Journal of Neurotherapy, 16*(3), 172–182.

Bronfenbrenner, U. (1995). Developmental ecology through space and time: a future perspective. In p. moen, g. h. elder, K. Lüscher, & U. Bronfenbrenner (Eds.), *Examining lives in context: perspectives on the ecology of human development* (1st ed., pp. 619–647). Washington, DC, USA: American Psychological Association.

Brown, T. E. (2009). *ADHD comorbidities: Handbook for ADHD complications in children and adults*. American Psychiatric Pub.

Capone, G. T., Goyal, P., Ares, W., & Lannigan, E. (2006). Neurobehavioral disorders in children, adolescents, and young adults with Down syndrome. *Am J Med Genet C Semin Med Genet, 142C*, 158–172.

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.

Carr, E. G., Dunlap, G., Horner, R. H., Koegel, R. L., Turnbull, A. P., Sailor, W., et al. (2002). Positive behavior support: Evolution of an applied science. *Journal of Positive Behavior Interventions, 4*, 4–16.

Chacko, A., Wymbs, B. T., Arnold, F. W., Pelham, W. E., Swanger-Gagne, M., Girio, E. L., et al. (2009). Enhancing traditional behavioral parent training for single-mothers of children with ADHD. *Journal of Clinical Child and Adolescent Psychology, 38*, 206–218.

Chronis, A. M., Chacko, A., Fabiano, G. A., Wymbs, B. T., & Pelham, W. E. (2004). Enhancements to the behavioral parent training paradigm for families of children with ADHD: Review and future directions. *Clinical child and family psychology review, 7*(1), 1–27.

Cihak, D. F., Kessler, K. B., & Alberto, P. A. (2007). Generalized use of a handheld prompting system. *Research in Developmental Disabilities, 28*(4), 397–408.

Cihak, D. F., Kessler, K., & Alberto, P. A. (2008). Use of a handheld prompting system to transition independently through vocational tasks for students with moderate and severe intellectual disabilities. *Education and Training in Developmental Disabilities*, 102–110.

Cooper, J. O., Heron, T. E., & Heward, W. L. (1987). *Applied Behavior Analysis*. Upper Saddle River, NJ: Merrill.

Correia Filho, A. G., Bodanese, R., Silva, T. L., Alvares, J. P., Aman, M., & Rohde, L. A. (2005). Comparison of risperidone and methylphenidate for reducing ADHD symptoms in children and adolescents with moderate mental retardation. *Journal of the American Academy of Child & Adolescent Psychiatry, 44*, 748–755.

Cortese, S., Panei, P., Arcieri, R., Germinario, E. A., Capuano, A., Margari, L., & Curatolo, P. (2015). Safety of methylphenidate and atomoxetine in children with attention-deficit/hyperactivity disorder (ADHD): Data from the Italian National ADHD Registry. *CNS drugs, 29*(10), 865–877.

Courtenay, K., & Elstner, S. (2016). Drug therapy in ADHD in people with intellectual disabilities. *Advances in Mental Health and Intellectual Disabilities, 10*(1), 27–35.

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.

Dunlap, G., Iovannone, R., Wilson, K. J., Kincaid, D. K., & Strain, P. (2010). Prevent-Teach-Reinforce: A standardized model of school-based behavioral intervention. *Journal of Positive Behavior Interventions, 12*(1), 9–22.

Dunlap, G., Kern-Dunlap, L., Clarke, S., & Robbins, F. R. (1991). Functional assessment, curriculum revision, and severe behavior problems. *Journal of Applied Behavior Analysis, 24*, 387–397.

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.

Dykens, E. M. (2007). Psychiatric and behavioral disorders in persons with Down syndrome. *Mental retarda-*

tion and developmental disabilities research reviews, 13(3), 272–278.

Epstein, M. H., Cullinan, D., & Polloway, E. A. (1986). Patterns of maladjustment among mentally retarded children and youth. *American journal of mental deficiency, 91*(2), 127–134.

Evans, S. W., Owens, J. S., Wymbs, B. T., & Ray, A. R. (2018). Evidence-based psychosocial treatments for children and adolescents with attention deficit/hyperactivity disorder. *Journal of Clinical Child and Adolescent Psychology, 47*(2), 157–198. https://doi.org/10.1080/15374416.2017.1390757

Fabiano, G. A., Pelham, W. E., Jr., Coles, E. K., Gnagy, E. M., Chronis-Tuscano, A., & O'Connor, B. C. (2009). A meta-analysis of behavioral treatments for attention-deficit/hyperactivity disorder. *Clinical psychology review, 29*(2), 129–140.

Fabiano, G. A., Pelham, W. E., Jr., Gnagy, E. M., Burrows-MacLean, L., Coles, E. K., Chacko, A., et al. (2007). The single and combined effects of multiple intensities of behavioral modification and methylphenidate for children with attention deficit hyperactivity disorder in a classroom setting. *School Psychology Review, 36*, 195–216.

Faraone, S. V., & Buitelaar, J. (2010). Comparing the efficacy of stimulants for ADHD in children and adolescents using meta-analysis. *European child & adolescent psychiatry, 19*(4), 353–364.

Fernández-Jaén, A., Fernández-Mayoralas, D. M., Pérez, B. C., Jareño, N. M., & Díaz, M. D. R. C. (2010). Atomoxetine for attention deficit hyperactivity disorder in mental retardation. *Pediatric Neurology, 43*(5), 341–347.

Fisher, W., Burd, L., Kuna, D. P., & Berg, D. J. (1985). Attention deficit disorders and the hyperactivities in multiply disabled children. *Rehabilitation literature.*

Fonagy, P., Cottrell, D., Phillips, J., Bevington, D., Glaser, D., & Allison, E. (2015). *What works for whom? a critical review of treatments for children and adolescents* (2nd ed.). New York, NY: The Guilford Press.

Frazier, J. A., Biederman, J., Bellordre, C. A., Garfield, S. B., Geller, D. A., Coffey, B. J., & Faraone, S. V. (2001). Should the diagnosis of attention-deficit/hyperactivity disorder be considered in children with pervasive developmental disorder? *Journal of Attention Disorders, 4*(4), 203–211.

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.

Gray, S. A., Chaban, P., Martinussen, R., Goldberg, R., Gotlieb, H., Kronitz, R., & Tannock, R. (2012). Effects of a computerized working memory training program on working memory, attention, and academics in adolescents with severe LD and comorbid ADHD: A randomized controlled trial. *Journal of Child Psychology and Psychiatry, 53*(12), 1277–1284.

Handen, B. L., Breaux, A. M., Janosky, J., McAuliffe, S., Feldman, H., & Gosling, A. (1992). Effects and non-effects of methylphenidate in children with mental retardation and ADHD. *Journal of the American Academy of Child Adolescent Psychiatry, 31*(3), 455–461.

Handen, B. L., Feldman, H., Gosling, A., Breaux, A. M., & McAuliffe, S. (1991). Adverse side effects of methylphenidate among mentally retarded children with ADHD. *Journal of American Academy of Child Adolescent Psychiatry, 30*(2), 241–245.

Handen, B. L., Feldman, H. M., Lurier, A., & Murray, P. H. (1999). Efficacy of methylphenidate among preschool children with developmental disabilities and ADHD. *Journal of the American Academy of Child & Adolescent Psychiatry, 38*(7), 805–812.

Handen, B., McAuliffe, S., & Caro-Martinez, L. (1996). Learning effects of methylphenidate in children with mental retardation. *Journal of Developmental and Physical Disability, 8*, 335–346.

Handen, B. L., McAuliffe, S., Janosky, J., Feldman, H., & Breaux, A. M. (1998). A playroom observation procedure to assess children with mental retardation and ADHD. *Journal of Abnormal Child Psychology, 26*(4), 269–277.

Handen, B. L., Sagady, A. E., & McAuliffe-Bellin, S. (2009). Methylphenidate and play skills in children with intellectual disability and ADHD. *Journal of Mental Health Research in Intellectual Disabilities, 2*(1), 1–10.

Hastings, R. P., Beck, A., Daley, D., & Hill, C. (2005). Symptoms of ADHD and their correlates in children with intellectual disabilities. *Research in Developmental Disabilities, 26*(5), 456–468.

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.

Hinshaw, S. P., Arnold, L. E., & MTA Cooperative Group. (2015). Attention-deficit hyperactivity disorder, multimodal treatment, and longitudinal outcome: evidence, paradox, and challenge. *Wiley Interdisciplinary Reviews: Cognitive Science, 6*(1), 39–52.

Huang, H., & Ruedrich, S. (2007). Recent advances in the diagnosis and treatment of attention-deficit-hyperactivity disorder in individuals with intellectual disability. *Mental Health Aspects of Developmental Disabilities, 10*(4), 121–128.

Individuals with Disabilities Education Improvement Act (IDEA), H. R. 1350, 108th Congress (2004).

Jain, R., Segal, S., Kollins, S. H., & Khayrallah, M. (2011). Clonidine extended-release tablets for pediatric patients with attention-deficit/hyperactivity disorder. *Journal of the American Academy of Child & Adolescent Psychiatry, 50*(2), 171–179.

Jensen, P. S., Arnold, L. E., Richters, J. E., Severe, J. B., Vereen, D., Vitiello, B., et al. (1999). Moderators and mediators of treatment response for children with attention-deficit/hyperactivity disorder- the multimodal treatment study of children with attention-deficit/hyperactivity disorder. *Archives of General Psychiatry, 56*(12), 1088–1096.

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.

Jones, K., Daley, D., Hutchings, J., Bywater, T., & Eames, C. (2007). Efficacy of the Incredible Years Basic parent training program as an early intervention for children with conduct problems and ADHD. *Child: Care, Health & Development, 33*, 749–756.

Jones, K., Daley, D., Hutchings, J., Bywater, T., & Eames, C. (2008). Efficacy of the Incredible Years Program as an early intervention for children with conduct problems and ADHD: Longterm follow-up. *Child: Care, Health & Development, 34*(3), 380–390. https://doi.org/10.1111/j.1365-2214.2008.00817.x

Jopp, D. A., & Keys, C. B. (2001). Diagnostic overshadowing reviewed and reconsidered. *American Journal on Mental Retardation, 106*(5), 416–433.

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.

Koller, H., Richardson, S. A., Katz, M., & McLaren, J. (1983). Behavior disturbance since childhood among a 5-year birth cohort of all mentally retarded young adults in a city. *American Journal of Mental Deficiency*.

Kollins, S. H., Jain, R., Brams, M., Segal, S., Findling, R. L., Wigal, S. B., & Khayrallah, M. (2011). Clonidine extended-release tablets as add-on therapy to psychostimulants in children and adolescents with ADHD. *Pediatrics, 127*(6), e1406.

Konrad, K., Gunther, T., Hanisch, C., & Herpertz-Dahlmann, B. (2004). Differential effects of methylphenidate on attentional functions in children with attention-deficit/hyperactivity disorder. *J Am Acad Child Adolesc Psych, 43*, 191–198.

Kutcher, S., Aman, M., Brooks, S. J., Buitelaar, J., van Daalen, E., Fegert, J., … Tyano, S. (2004). International consensus statement on attention-deficit/hyperactivity disorder (ADHD) and disruptive behaviour disorders (DBDs): clinical implications and treatment practice suggestions. *Eur Neuropsychopharmacol, 14*, 11–28.

Linda C. Mechling, David L. Gast, Elizabeth A. Fields, (2008) Evaluation of a Portable DVD Player and System of Least Prompts to Self-Prompt Cooking Task Completion by Young Adults With Moderate Intellectual Disabilities. *The Journal of Special Education 42*(3):179–190

La Malfa, G., Lassi, S., Bertelli, M., Pallanti, S., & Albertini, G. (2008). Detecting attention-deficit/hyperactivity disorder (ADHD) in adults with intellectual disability: The use of Conners' Adult ADHD Rating Scales (CAARS). *Research in developmental disabilities, 29*(2), 158–164.

Lambert, N., Sassone, D., & Sandoval, J. (1987). Persistence of hyperactivity symptoms from childhood to adolescence and associated outcomes. *Am J Orthopsychiatry, 57*, 22–32.

Lindsey, M. P. (2002). Comprehensive health care services for people with learning disabilities. *Advances in Psychiatric Treatment, 8*(2), 138–147.

McBrien, H., Turk, J., & Letch, N. (2006). The management of ADHD and associated problems in a young person with cleidocranial dysostosis (CCD) and mild intellectual disability. *Clinical child psychology and psychiatry, 11*(3), 445–456.

Majewicz-Hefley, A., & Carlson, J. S. (2007). A meta-analysis of combined treatments for children diagnosed with ADHD. *Journal of Attention Disorders, 10*(3), 239–250.

Martinez-Raga, J., Knecht, C., Szerman, N., & Martinez, M. I. (2013). Risk of serious cardiovascular problems with medications for attention-deficit hyperactivity disorder. *CNS Drugs, 27*(1), 15–30.

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., Turygin, N. C., Beighley, J., Rieske, R., Tureck, K., & Matson, M. L. (2012). Applied behavior analysis in Autism Spectrum Disorders: Recent developments, strengths, and pitfalls. *Research in Autism Spectrum Disorders, 6*(1), 144–150.

Mazzone, L., Reale, L., Mannino, V., Cocuzza, M., & Vitiello, B. (2011). Lower IQ is associated with decreased clinical response to atomoxetine in children and adolescents with attention-deficit hyperactivity disorder. *CNS Drugs, 25*(6), 503–509.

Meaux, J. B., Hester, C., Smith, B., & Shoptaw, A. (2006). Stimulant medications: A trade-off? The lived experience of adolescents with ADHD. *Journal for Specialists in Pediatric Nursing, 11*(4), 214–226.

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*, 220–237.

Miller, M. L., Fee, V. E., & Netterville, A. K. (2004). Psychometric properties of ADHD rating scales among children with mental retardation I: Reliability. *Research in Developmental Disabilities, 25*(5), 459–476.

Olfson, M., Huang, C., Gerhard, T., Winterstein, A. G., Crystal, S., Allison, P. D., et al. (2012). Stimulants and cardiovascular events in youth with attention-deficit/hyperactivity disorder. *Journal of the American Academy of Child and Adolescent Psychiatry, 51*(2), 147–156.

Pearson, D. A., & Aman, M. G. (1994). Ratings of hyperactivity and developmental indices: Should clinicians correct for developmental level? *Journal of autism and developmental disorders, 24*(4), 395–411.

Pearson, D. A., Lane, D. M., Santos, C. W., Casat, C. D., Jerger, S. W., Loveland, K. A., & Roache, J. D. (2004a). Effects of methylphenidate treatment in children with mental retardation and ADHD: Individual variation in medication response. *Journal of the American Academy of Child & Adolescent Psychiatry, 43*(6), 686–698.

Pearson, D. A., Santos, C. W., Casat, C. D., Lane, D. M., Jerger, S. W., Roache, J. D., & Cleveland, L. A. (2004b). Treatment effects of methylphenidate on cognitive functioning in children with mental retardation and ADHD. *Journal of the American Academy of Child & Adolescent Psychiatry, 43*(6), 677–685.

Pelham, W. E., Jr., & Fabiano, G. A. (2008). Evidence-based psychosocial treatments for attention-deficit/hyperactivity disorder. *Journal of Clinical Child & Adolescent Psychology, 37*(1), 184–214.

Pelham, W. E., Jr., Wheeler, T., & Chronis, A. (1998). Empirically supported psychosocial treatments for attention deficit hyperactivity disorder. *Journal of clinical child psychology, 27*(2), 190–205.

Pennington, B. F., & Ozonoff, S. (1996). Executive functions and developmental psychopathology. *Journal of child psychology and psychiatry, 37*(1), 51–87.

Pfiffner, L. J., Rooney, M., Haack, L., Villodas, M., Delucchi, K., & McBurnett, K. (2016). A randomized controlled trial of a school-implemented school–home intervention for attention-deficit/hyperactivity disorder symptoms and impairment. *Journal of the American Academy of Child & Adolescent Psychiatry, 55*(9), 762–770.

Pliszka, S. R. (2009). *Treating ADHD and comorbid disorders: Psychosocial and psychopharmacological interventions*. Guilford Press.

Reilly, C., & Holland, N. (2011). Symptoms of attention deficit hyperactivity disorder in children and adults with intellectual disability: A review. *Journal of Applied Research in Intellectual Disabilities, 24*(4), 291–309.

Roux, G., Sofronoff, K., & Sanders, M. (2013). A randomized controlled trial of group stepping stones triple P: A Mixed-Disability trial. *Family process, 52*(3), 411–424.

Rowles, B. M., & Findling, R. L. (2010). Review of pharmacotherapy options for the treatment of attention-deficit/hyperactivity disorder (ADHD) and ADHD-like symptoms in children and adolescents with developmental disorders. *Developmental disabilities research reviews, 16*(3), 273–282.

Sanders, M. R. (1999). Triple P-Positive Parenting Program: Towards an empirically validated multilevel parenting and family support strategy for the prevention of behavior and emotional problems in children. *Clinical child and family psychology review, 2*(2), 71–90.

Schwarz, A. (2013). A.D.H.D. experts re-evaluate study's zeal for drugs. The New York Times. Retrieved from http://www.nytimes.com/2013/12/30/health/adhd-experts-re-evaluate-studys-zeal-for-drugs.html?_r=0

Sears, K. M., Blair, K. S. C., Iovannone, R., & Crosland, K. (2013). Using the prevent-teach-reinforce model with families of young children with ASD. *Journal of Autism and Developmental Disorders, 43*(5), 1005–1016.

Shane, H. C., & Albert, P. D. (2008). Electronic screen media for persons with autism spectrum disorders: Results of a survey. *Journal of Autism and Developmental Disorders, 38*, 1499–1508.

Simonoff, E., Pickles, A., Wood, N., Gringras, P., & Chadwick, O. (2007). ADHD symptoms in children with mild intellectual disability. *Journal of the American Academy of Child & Adolescent Psychiatry, 46*(5), 591–600.

Simonoff, E., Taylor, E., Baird, G., Bernard, S., Chadwick, O., Liang, H., & Wood, N. (2013). Randomized controlled double-blind trial of optimal dose methylphenidate in children and adolescents with severe attention deficit hyperactivity disorder and intellectual disability. *Journal of Child Psychology and Psychiatry, 54*(5), 527–535.

Söderqvist, S., Bergman Nutley, S., Ottersen, J., Grill, K. M., & Klingberg, T. (2012). Computerized training of non-verbal reasoning and working memory in children with intellectual disability. *Frontiers in Human Neuroscience, 6*, 271.

Solanto, M. V., Arnsten, A. F. T., & Castellanos, F. X. (2001). *Stimulant drugs and ADHD*. Oxford: Oxford University Press.

Strain, P. S., Wilson, K., Wilson, K., & Dunlap, G. (2011). Prevent-teach-reinforce: Addressing problem behaviors of students with autism in general education classrooms. Behavioral Disorders, 160-171.

Swanson, J. M., Sergeant, J. A., Taylor, E., Sonuga-Barke, E. J. S., Jensen, P. S., & Cantwell, D. P. (1998). Attention-deficit hyperactivity disorder and hyperkinetic disorder. *The Lancet, 351*(9100), 429–433.

Swanson, J. M., Wigal, T., Kollins, S., Newcorn, J., Wang, G.-J., Fowler, J., et al. (2013). The dopamine hypothesis of ADHD and brain response to stimulant medication. In B. R. Kar (Ed.), *Cognition and brain development: Converging evidence from various methodologies* (pp. 127–143). Washington, DC: American Psychological Association.

Tanaka, Y., Rohde, L. A., Jin, L., Feldman, P. D., & Upadhyaya, H. P. (2013). A meta-analysis of the consistency of atomoxetine treatment effects in pediatric patients with attention-deficit/hyperactivity disorder from 15 clinical trials across four geographic regions. *Journal of child and adolescent psychopharmacology, 23*(4), 262–270.

Tarrant, N., Roy, M., Deb, S., Odedra, S., Retzer, A., & Roy, A. (2018). The effectiveness of methylphenidate in the management of attention deficit hyperactivity disorder (ADHD) in people with intellectual disabilities: A systematic review. *Research in Developmental Disabilities, 83*, 217–232.

Thapar, A. (2017). Intellectual disability and attention-deficit/hyperactivity disorder: what does the clinical and genetic overlap mean for practice and research? *Journal of the American Academy of Child & Adolescent Psychiatry, 56*(2), 105–106.

Thomson, A., Maltezos, S., Paliokosta, E., & Xenitidis, K. (2009). Risperidone for attention-deficit hyperactivity disorder in people with intellectual disabilities. *Cochrane Database of Systematic Reviews, 2*.

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.

Van der Oord, S., Prins, P. J., Oosterlaan, J., & Emmelkamp, P. M. (2008). Efficacy of methylphenidate, psychosocial treatments and their combination in school-aged children with ADHD: A meta-analysis. *Clinical psychology review, 28*(5), 783–800.

Van Laarhoven, T., Johnson, J. W., Van Laarhoven-Myers, T., Grider, K. L., & Grider, K. M. (2009). The effectiveness of using a video iPod as a prompting device in employment settings. *Journal of Behavioral Education, 18*(2), 119.

Vinogradov, S., Fisher, M., & de Villers-Sidani, E. (2012). Cognitive training for impaired neural systems in neuropsychiatric illness. *Neuro- psychopharmacology, 37*, 43–76.

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.

White, M., Nichols, C., Cook, R., Spengler, P., Walker, B., & Look, K. (1995). Diagnostic overshadowing and mental retardation: A meta-analysis. *American Journal on Mental Retardation, 100*, 293–298.

Xenitidis, K., Paliokosta, E., Rose, E., Maltezos, S., & Bramham, J. (2010). ADHD symptom presentation and trajectory in adults with borderline and mild intellectual disability. *Journal of Intellectual Disability Research, 54*(7), 668–677.

Treatment of Substance Abuse in Dual Diagnosis

32

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Introduction

While for individuals with an (above) average IQ and adaptive skills who have substance use–related problems, numerous interventions are available for prevention, early intervention, treatment and aftercare, this is not the case for individuals with ID. In the reviews by Kerr, Lawrence, Darbyshire, Middleton, and Fitzsimmons (2013) and Van Duijvenbode et al. (2015), it was found that there is a limited number of substance use (disorder) interventions adapted to the needs and learning style of those with ID, and those that are available generally are predominantly aimed at reducing tobacco and alcohol use. Though the scarcity of interventions is understandable given the under-recognition of substance use and its related problems in individuals with ID, it is also problematic. First, individuals with ID are underserved with respect to their substance use–related issues, be it from questions with regard to "safe" substance use consumption to multidimensional treatment for complex addiction problems. For them, the lack of suitable interventions can lead to substance use initiation without risk awareness, to progression of substance use into substance use disorder, and to poorer outcomes of treatment. Second, the lack of interventions also is problematic for those who work with individuals with ID and substance-related issues. Staff from both intellectual disability services and addiction treatment or other (e.g., forensic psychiatric) facilities lack resources on how to help substance-using individuals with ID, and may become discouraged from helping these persons. Moreover, lack of treatment progression may lead to stigmatization or blaming those with dual diagnosis or triple diagnosis.

Nevertheless, the number of studies assessing the feasibility and outcomes of interventions for substance (ab)use in individuals with ID has increased during the past decade. The aim of this chapter is to present a short overview of interventions that are adapted to individuals with dual diagnosis: ID and substance use abuse. Psychological and pharmacological interventions are summarized and attention will be given to how to adapt interventions to clients with ID. A distinction is made between dual diagnosis and triple diagnosis, the latter being a condition that is seldom addressed in the literature. Finally, developments regarding personalized treatments will be described and attention is paid to screening and assessment and collaboration between organizations.

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Psychological and Pharmacological Interventions

Psychoeducation and Prevention

Substance-related knowledge in individuals with ID is fragmented at best, and often full of misconceptions based on biased information from television and internet as well as from substance-using peers. While in mainstream education substance use prevention programs are often available, this generally is not the case in special education programs. Even though it is widely known that educational programs aimed at the general public are not (very) effective in substance use (disorder) prevention, the question is if some type of preventative programs tailored to the needs of individuals with ID are needed to increase risk awareness, ability to withstand offerings of substance use, and ability to ask for help when needed. Such programs need to be offered at an appropriate time (preferably before substance use initiation, but not prematurely as it may not yet be relevant), and in a way that is suitable for the needs of the individual.

There are very few studies published on prevention of substance use in young people with ID. A controlled study on a prevention program targeted at alcohol and tobacco use in 12–16-year-old children with ID was conducted by Kiewik, VanDerNagel, Engels, and De Jong (2017). Participants were students from three secondary special-needs schools who were assigned to an experimental or control condition. In the experimental condition, the students received a Dutch prevention program called *Prepared on time* that aims to delay initiation of use of alcohol and tobacco. It is an e-learning program including games, videos, and quizzes to increase students' substance knowledge, to provide appropriate samples of refusal skills, and to strengthen students' ability to make own choices. Students in the control condition followed their standard educational curriculum. Results of this study showed that a classroom-based e-learning program is feasible in students with mild to moderate ID, but had limited effect on their knowledge, attitude, and intention to use tobacco and alcohol. This may be related to the fact that a large subgroup of individuals within this study had already initiated smoking and drinking. In addition, prevention programs may be more effective if they do not (only) target children and adolescents, but include their parents and caregivers. As with many issues in educating individuals, showing (modeling) rather than talking about healthy and responsible behavior may be the best way to prevent substance use–related problems.

Cognitive Behavioral Treatment

Most substance use disorder treatment programs are based on cognitive behavioral approaches. Individuals learn – either within a group or within individual treatment – to identify risky situations, behaviors, thoughts, and feelings, and learn self-control techniques to reduce risks and alter their behaviors. Cognitive behavioral treatment (CBT) is widely used in individuals with mild ID or borderline intellectual functioning (IQ 50–85). It has, for example, been shown to be effective in reducing anger and violence in individuals of this target group (see Didden, Nijman, Delforterie, & Keulen-De Vos, 2019). CBT is a composite treatment with several components – including, for example, cognitive restructuring, relaxation, understanding and regulation of emotions, and skill building – designed to reduce substance use–related problems. The assumptions underlying CBT are that behavior, thoughts, and emotions are interconnected, and that cognitive distortions and maladaptive coping strategies increase the risk for psychological and behavioral problems. These problems can be reduced by improving information processing and the learning of adaptive coping skills. Therapists use functional analyses and case formulations to understand the nature and cause of the problems and as a guide to develop treatment programs (Didden et al., 2019).

Examples of programs developed for individuals with ID and substance use disorders are as follows: *Cognitive Behavioural Treatment – Plus* (VanDerNagel & Kiewik, 2016; Kiewik, VanDerNagel, Engels, & De Jong, submitted;

available in Dutch through the Dutch Addiction Association), *Extended Brief Intervention for Alcohol Misuse – LD* (Kouimtsidis et al., 2017; available online in English), *Alcohol and Substance Abuse Programme – Intellectual Disability,* and *Take it Personal!* Kouimtsidis et al. assessed the feasibility of a manualized 8-week brief extended intervention (BEI) in 30 adults with moderate to mild ID who lived in the community and who reported alcohol-related problems. Participants were assigned to BEI or control group (care as usual and/or advice to stop drinking). The intervention consisted of motivational and cognitive behavioral techniques to affect change in motivation, cognitions, and behavior. Results show that after 12 weeks the proportion of participants with harmful drinking had decreased by 67% and 47% for the intervention and control group, respectively. Participants' and caregivers' feedback on their experience with BEI was positive. Kiewik et al. conducted a feasibility study on CBT – Plus, which is adapted to adults with mild ID or borderline intellectual functioning. Adaptation of the original CBT program which has been shown effective in individuals without ID consisted of the following: a workbook for clients was developed with easy language and visual cues, the number of sessions was increased from 9 to 18, and client's confidants were involved during alternate sessions. An important outcome measure was used that was developed for use in clients with mild ID: *Substance use and misuse in intellectual disability – Questionnaire* (SUMID-Q). Next to the SUMID-Q, experiences of clients, confidants, and therapists with the CBT Plus program were explored through interviews. Results showed that the users highly valued the CBT Plus intervention and that most clients (i.e., 70%) completed the treatment. These clients all showed a reduction in the use of alcohol, stimulants, and cannabis after the treatment as assessed with the SUMID-Q.

Many individuals with ID who are admitted to forensic facilities have a history of substance abuse, and in many cases there is a link between offending behavior or recidivism and abuse of alcohol and other substances (Lindsay, 2009). Sakdalan, Kittner, and Judd (2017) explored the effectiveness of a program called *Alcohol and Substance Abuse Programme – Intellectual Disability* (ASAP-ID) developed for forensic clients with ID. ASAP-ID is a 27-week program with weekly sessions that last approximately 1.5 hours. The program is adapted to clients with ID including repetition, visual aids, role-play, and other techniques. Clients were given homework to complete with their support staff. Staff members were sometimes present during sessions to support their clients. The program incorporates procedures of motivational interviewing, psychoeducation, relapse prevention, and skill acquisition. The program was tested in 6 participants with moderate to mild ID who had a Maori background. During their stay in the facility, participants were prohibited from using substances. Data show differences between pre- and post-tests on several outcome measures. The findings show a marked improvement in participants' confidence to stay clean and sober in risky situation as well as an improvement in overall readiness for change.

Pharmacology

Substance use disorder treatment can be supported by pharmacological interventions. These include (a) interventions to reduce risks and symptoms during detoxification and withdrawal, (b) interventions to reduce relapse, (c) substitution therapy, and (d) treatment of co-occurring somatic and psychiatric conditions. All of these interventions require specialist knowledge that may be hard to find for individuals with ID. Regardless, assessment for the need of pharmacological treatment should not be omitted in those with substance use disorder, given the risks associated with both prolonged substance use, and those associated with the detoxification and withdrawal process.

To our knowledge, no studies are available regarding the pharmacology for substance use disorder in individuals with ID. Clinical experience shows that general guidelines for pharmacology in addiction medicine are appropriate,

with some remarks and issues that require special attention. During detoxification, pharmacological interventions may be required to reduce withdrawal symptoms. Contrary to common belief, this is not mainly for the patient's comfort, but also to avoid severe, potential debilitating, or even lethal complications of withdrawal. Alcohol withdrawal is especially associated with severe complications such as delirium and seizures. These can be avoided by prescribed benzodiazepines in a tapering schedule. In addition, individuals with alcohol use disorder should be prescribed intramuscular vitamin B1 (thiamine) to avoid Wernicke's encephalopathy. GHB (gamma hydroxybutyric acid) withdrawal is also known for its severe complications (including delirium, excited delirium, psychosis, aggression) during detoxification that often requires intensive monitoring and intervention in specialized inpatient wards. Prescribed benzodiazepines in a tapering schedule are also often needed in case of benzodiazepine dependency to avoid severe side effects. Withdrawal from other substances – though temporarily debilitating – from a medical point of view is generally less risky or complicated. Nevertheless, often these patients can benefit from medical interventions to reduce withdrawal symptoms, and increase the likelihood of a successful detoxification. Individuals with ID may require more intensive medical supervision during detoxification, especially if they have co-occurring medical issues, such as epilepsy, a lack of support and monitoring from their social environment, or limited abilities to seek medical support when needed. In addition, in some patients with ID, symptom relief is especially important to complete the detoxification process.

Pharmacological interventions to reduce the risk of relapse can generally be divided into two groups: (a) medication to reduce craving and (b) aversive medication. Pharmacotherapy to reduce craving include, for instance, acamprosate or baclofen for alcohol use disorder, naltrexone for alcohol or opiate use disorder, and bupropion for tobacco use disorder. In individuals with ID (and their caregivers), it is important to explain in detail that these medications do not take away all cravings; they provide some support, but are not a cure from addiction. In addition, these medications should be taken regularly as prescribed (i.e., not on a "take-as-needed" basis). Aversive medication is available for alcohol use disorders, with medications such as disulfiram. It intervenes with the normal alcohol metabolism, resulting in the accumulation of toxic by-products if taken together with alcohol. Patients who take this drug and use alcohol experience side effects, such as severe nausea, hot sweats, and palpitations that should discourage them from drinking. Though these effects are generally not severe, prescribing this type of medication for individuals with ID should be done with great precaution.

Maintenance therapy or "replacement therapy"' may be needed in some patients with prolonged substance use disorder. Strategies are mainly available for tobacco use disorder (e.g., use of nicotine plasters or lozenges) and opiate use disorder (e.g., within methadone clinics). While the patients within these programs are still taking psychoactive substances, they do so in a more regulated and safe manner, thus reducing the risks and complication associated with substance use. For some patients, replacement or maintenance therapy is a short-term intervention, but others benefit from long-term support by these programs. Regular monitoring of effects and side effects and general health of patients within these programs remains necessary.

Last but not least, as substance use disorders are associated with a number of co-occurring illnesses (including malnutrition and vitamin/mineral deficiencies, infectious diseases, liver disease, and psychiatric disorders), treatment of these disorders may also require pharmacological interventions. Addiction treatment centers often have screening programs to assess the need for such treatment. In individuals with ID such screening is even more important, given the increased likelihood of untreated physical or mental disorders within this group.

Adapting Treatments to Individuals with ID

Treatment of substance use disorder is not fundamentally different for individuals with ID than for others. Inpatient or outpatient and pharmacological or psychological interventions can all be suitable for patients regardless on their intellectual and adaptive abilities. Thus, based on a multidisciplinary assessment, each individual with substance use disorder should be offered a tailor-made program to meet their individual treatment requirements (Kiewik, 2018). However, treating individuals with ID will require some adaptation (Van Duijvenbode et al., 2015). Several authors have shown that these adaptations are feasible in a wide range of programs, namely, motivational interviewing (e.g., Frielink & Embregts, 2013), cognitive behavioral therapy (e.g., Kouimtsidis et al., 2017; VanDerNagel & Kiewik, 2016), and, the 12-step program (e.g., Jurewicz, 2017). Adaptations of treatment programs include reduction of language complexity, inclusion of real-life exercises, and using a less information dense program with more repetition. Treatment procedures may need to be adapted with tailoring the number and length of appointments to the needs of the individual patient, collaboration with a mentor or confidant from the patient's social environment, length of treatment and follow-up, and – in group therapy – more homogeneous groups (i.e., with patients with similar intellectual and adaptive abilities). Generally, individuals with ID will need more support to be able to apply new information and skills in daily life. Welcoming a confidant within the intake and assessment procedure, and within each or alternating therapy sessions, can provide an important bridge to daily life (VanDerNagel, Kemna, Barendregt, & Wits, submitted; Kiewik, 2018). However, working within the therapist-patient-confidant triade requires specific therapist skills, especially when patient and confidant have different opinions as to the magnitude and scope of the problems, or with regard to the preferred solutions.

Lindsay (2009) stated that psychological treatment requires adaptations for individuals with ID especially with regard to communication. Adaptation requires, for example, adjustments of vocabulary and syntax in addition to continuing self-monitoring. He provides the following basic recommendations:

- Use short sentences that contain a single concept.
- Use words of fewer than three syllables.
- Ask clients to summarize the session in order to assess their understanding and retention.
- Use inductive methods (e.g., Socratic dialogue).
- Use role-play.
- Increase motivation to change.
- Work with significant others and relatives.

Hronis, Roberts, and Kneebone (2017) provided suggestions for adapting CBT to individuals with ID who present with cognitive skill deficits. Below are some examples:

- Attention: use shorter, more frequent sessions, reduce task length (smaller units), prevent distractions.
- Working memory: use memory aids (e.g., visual prompts), present one task at a time, use short, simple, subject-verb-object sentences.
- Executive functions: use structured sessions (e.g., visual schedule), minimize switching between tasks, redirect uninhibited responses.

Group and individual cognitive behavioral approaches are often used in the treatment of substance use–related problems in individuals with ID. However, participants may lack appropriate skills required for participating in such programs. They have difficulties understanding the cognitive components even if the program is adapted to their learning style and needs. It has been suggested that those who lack prerequisite skills may benefit from pre-therapy in which they are taught CBT basic concepts such as identifying, differentiating, and linking emotions, thoughts, and events (see Tsimopoulou, Stenfert Kroese, Unwin, Azmi, & Jones, 2018).

Inpatient treatment can both be helpful and unsettling for individuals with ID. Being away

from their day-to-day environment and intensive monitoring can be helpful to stop substance use, and this may be needed to provide medical treatment as well. However, homesickness may occur, and patients may be overburdened with social and intellectual requirements that come with living with other patients in a ward. This may lead to patients leaving treatment prematurely and to conflicts with other patients or staff (often resulting in patients being sent away as being unmotivated, aggressive, or obstinate). Proper preparation of inpatient treatment with both patient and his caregivers at home may reduce the risks of such unwanted outcome. A structured day program (when needed not (only) in words but with pictures), more one-on-one staff attention, scheduled calls or visits with caregivers or family, increased staff support during group interactions, as well as focus on the individuals abilities (i.e., rewarding and acknowledging them for helping staff setting the table) are needed to ensure a successful clinical stay during, for example, detoxification. While detoxification requires specialist knowledge and skills regarding substance use disorders that is generally found in addiction treatment centers, for long-term clinical treatment after detoxification, individuals with ID may be better off in specialized wards for individuals with ID. However, as substance use disorders generally require long-term follow-up, and are associated with relapses (even during clinical treatment), their treatment will require substantial knowledge of substance use disorders and its treatment. Preferably, during all treatment phases there is an intensive cross-system collaboration between addiction medicine and disability services.

Triple Diagnosis

Substance abuse in individuals with ID is often accompanied by other problems, such as financial problems, housing problems, or unemployment. Also mental health problems are common in individuals with substance abuse. For example, European research shows that approximately 50% of those diagnosed with SUD also have a co-occurring mental disorder, although these percentages differ widely between studies depending on, for example, the sample and specific combination of SUD and mental disorder (European Monitoring Center for Drugs and Drug Addiction, 2015). If SUD and mental disorders co-occur in individuals with ID, this is called triple diagnosis.

Prevalence

There is little information about the prevalence of triple diagnosis. In most research articles on either SUD or mental disorders in individuals with ID, the prevalence rates suggest at least some overlap between SUD and mental disorders. For example, Hassiotis et al. (2011) report high prevalence rates of personality disorders (82%), neuroticism (53%), and psychotic disorders (11%) in a group of 170 detainees with ID. In addition, 61% was diagnosed with an alcohol use disorder and 56% with a drug use disorder. However, it remained unclear how many subjects had a mental disorder and SUD.

The results of the articles in which information about triple diagnosis can be found are difficult to compare due to differences in sample content, setting, methodology, and definitions of SUD, ID, and mental disorders. As a result, the prevalence rates of triple diagnosis vary between 11% (Holden & Neff, 2000) and 54% (Slayter, 2010). Especially among individuals receiving (long-term) residential care and among individuals receiving involuntary care (e.g., within the judicial domain), relatively high prevalence rates of triple diagnosis are found.

Recently, we conducted a study on the prevalence of triple diagnosis in 75 clients with ID who were receiving treatment and care from a forensic, addiction, or ID facility (Van Duijvenbode, VanDerNagel, Janssen van Raay, & Didden, 2019). Results revealed that almost half of the clients used different substances (poly users), and 90% were diagnosed with SUD. The three most often used substances were alcohol (57%), cannabis (47%), or cocaine (41%). Most prevalent mental disorders next to SUD were

personality disorder (32%) and developmental disorders (e.g., autism spectrum disorder, attention deficit hyperactive disorder; 36%). Trauma-related disorders (24%), mood disorders (17%), and anxiety disorders (5%) were less often classified. Aggressive and rule-breaking behavior were seen in 87% of the sample, and 75% of the clients were known to police and justice. The range of problems of clients with triple diagnosis is therefore diverse and involves different combinations of mental disorders and types of substances that are used. This means that individuals with triple diagnosis form a heterogeneous group. The common denominator is that the problems are complex and intertwined.

SUD, ID, and mental disorders are seldom separate disorders "accidentally" co-occurring in a specific individual. Instead, they often reinforce each other negatively. Indeed, research shows that individuals with a dual or triple diagnosis often experience more severe symptoms and have a worse treatment prognosis (e.g., Lambert, LePage, & Schmitt, 2003; Langas, Malt, & Opjordsmoen, 2011; VanDerNagel, Kiewik, & Didden, 2017). In addition, SUD, ID, and mental disorders often jointly lead to additional psychosocial problems, such as financial problems, unemployment, or delinquency. These social problems, in turn, have a negative influence on overall functioning, thereby creating a vicious circle or downwards spiral.

Referral

Because SUD, ID, and mental disorders are often intertwined and do not fit in one box, treatment of triple diagnosis is challenging. One of the questions that often immediately rises is: Where can a patient with triple diagnosis be referred to? Addiction medicine? Mental health care? ID care? A useful guideline in the referral of patients with dual or triple diagnosis is the four-quadrant model of Minkoff (2001). Minkoff described that individuals with dual diagnosis – in this case: the combination of SUD and mental disorder – can be subdivided into four groups, based on the severity of both the SUD and mental disorder. VanDerNagel then transformed the two-dimensional quadrants into a three-dimensional cube to also take into account the possible influence of ID on referral and treatment (VanDerNagel et al., 2017).

The bottom four quadrants correspond to the original model. Although these individuals have ID, they are expected to benefit sufficiently from regular treatment. The first quadrant involves patients with a mild mental disorder and a mild to moderate SUD. These patients will often benefit from outpatient treatment in a primary health care setting. The second quadrant describes patients with a severe and/or persistent mental disorder and a mild to moderate SUD. These patients can best be referred to a mental health care facility for treatment. The third quadrant describes a group of patients with a mild mental disorder and a moderate to severe SUD. Because treatment of SUD will likely improve psychological functioning and well-being, these patients are usually referred to addiction medicine. The last quadrant involves patients with a severe and persistent mental disorder and moderate to severe SUD. In these cases, an integrated and specialized double-diagnosis treatment is necessary, either within mental health care or addiction medicine (Fig. 32.1).

In the upper four quadrants, the characteristics of ID must be taken into account to benefit from treatment. In other words, these individuals need tailored treatments, designed specifically for individuals with ID. The fifth quadrant contains individuals with ID, mild mental disorder, and mild to moderate SUD. Locus of treatment in these cases should be within ID care, complemented with consultation from professionals working in mental health care or addiction medicine if necessary. The sixth quadrant describes individuals with ID, a severe and/or persistent mental disorder, and a mild to moderate SUD. They are preferably treated by teams within mental health care specialized in the treatment of mental disorders in individuals with ID. Similarly, the seventh quadrant contains individuals with ID, a mild mental disorder, and a moderate to severe SUD. They are preferably treated by teams within addiction medicine specialized in the

		Severity of the addiction	
		Low	High
Severity of mental disorder	Low	A (mental health care/general practitioner)	B (addiction medicine)
	High	C (mental health care)	D (integrated treatment) Severe mental disorder and substance abuse

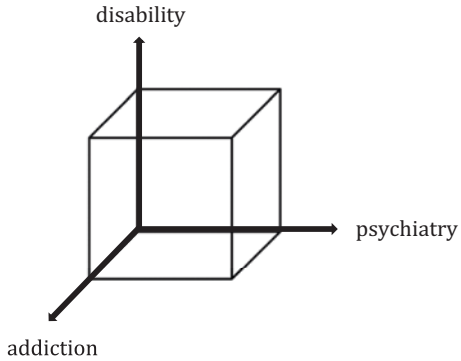

Fig. 32.1 Matrix by Minkoff (2001) and cube by VanderNagel et al. (2017)

treatment of SUD in individuals with ID. Last, the eighth quadrant describes individuals with IDD, a severe and persistent mental disorder, and a moderate to severe SUD. In these cases, an integrated and specialized triple-diagnosis treatment is necessary, where knowledge and expertise on (the treatment of) ID, SUD, and mental disorders are combined within one specialized team of professionals.

Treatment

Treatment protocols for individuals with triple diagnosis are scarce. In fact, we know of only one protocol that is currently being developed for use in individuals with ID. *Seeking Safety Plus* is a Dutch addendum for the original Seeking Safety treatment protocol (Najavits, 2002), designed for individuals with ID, SUD, and trauma-related disorders. It focuses on stabilization and creating a safe context by developing recovery and coping skills. It is currently being pilot tested for applicability, usefulness, and effectiveness.

Despite the lack of treatment protocols for triple diagnosis, a number of suggestions can be made to improve the care and treatment for patients with triple diagnosis. First, it is vital to analyze the patient's history of care. A case file study of 75 patients with triple diagnosis shows that they have often received care from multiple organizations (see Van Duijvenbode et al., 2019). Yet, information about why patients were in contact with health care professionals (either within ID care, mental health care or addiction medicine), what has been done, and how the progression was, was often not available in patient records. Requesting this information is important to be able to learn from what previously worked (and did not work) and thus direct future treatment decisions.

Second, a thorough screening and assessment is essential. Patient records should at the very least contain information about functioning (intellectual, adaptive, social-emotional), mental

health symptoms, substance use (types of substances, frequency, amount, severity of SUD, function), and additional psychosocial problems (such as information about social network, schooling/work, housing, finances) in order to arrive at an integrative theory and a hypothesis about how the elements of mental disorder, SUD, ID, and psychosocial problems are intertwined and related to each other.

Last, organizations within ID care, mental health care, and addiction medicine must collaborate (Van Duijvenbode et al., 2015). Having insufficient knowledge or expertise on a certain subject (e.g., the care for individuals with ID or treating SUD) should not be used as an "excuse" to leave problems untreated or refer patients. Individuals with triple diagnosis require specialized care from multidisciplinary teams in which knowledge and expertise regarding ID, SUD, and mental disorders is present. Collaboration and fertilization between ID care, addiction medicine, and mental health care are key factors in achieving this.

Personalized Treatments

Recently, the focus in mental health care moved from a "one size fits all" approach towards a more personalized approach. With regard to SUD treatment, the National Institute on Drug Abuse (2012) stated that SUD treatment should meet the characteristics of the client and should pay attention to specific problems associated with the SUD. Or in other words, treatment should be personalized. Traditionally, treatment effectiveness is determined based on a group approach, such as a randomized controlled trial, reducing individual scores to overall mean scores, and resulting in treatment-related products also aimed at groups. Not surprisingly, protocolled SUD treatment in individuals with ID is often aimed at a broad heterogeneous group of individuals using one general approach and leaving little room for targeted individualized intervention. First approaches to personalization of SUD treatment focus on identifying subgroups of clients defined by underlying psychological or biological mechanisms (Insel & Cuthbert, 2015). To further improve SUD treatment in individuals with ID differentiation in this heterogeneous group is necessary, as one size does not fit all.

An effective personalized approach is personality-based treatment. This type of treatment is based on the personality dimensions such as anxiety sensitivity, negative thinking, impulsivity, and sensation seeking (Conrod et al., 2013). Ample research in samples of people with SUD has shown that people with SUD can generally be differentiated in these four personality profiles. In addition, these personality profiles have also been identified as an important risk factor for the development of SU(D) in individuals with average intelligence (e.g., Woicik, Stewart, Phil, & Conrod, 2009). More recently, there is also evidence for the role of these personality dimensions in SU(D) individuals with ID (Poelen, Schijven, Otten, & Didden, 2016).

The differentiation in four personality dimensions reflects the theoretical perspective that vulnerability to SU(D) can be explained by a sensitivity to either negative or positive reinforcement processes that maintain substance use (Woicik et al., 2009). The dimensions, namely, anxiety sensitivity and negative thinking are mainly related to substance use maintained by negative reinforcement, that is, substance use to cope with negative emotional states (Comeau, Stewart, & Loba, 2001; Cooper, Frone, Russell, & Mudar, 1995). However, the specific facets of the personality dimensions such as anxiety sensitivity and negative thinking determine the nature of negative reinforcement of substance use. Anxiety sensitivity is defined as the fear of symptoms of physical arousal and is related to self-medication of anxious symptoms through the use of alcohol and/or drugs (Comeau et al., 2001; Conrod, Pihl, & Vassileva, 1998; Woicik et al., 2009). Negative reinforcement related to negative thinking is characterized by substance use to relief negative affect (Hecimovic, Barrett, Darredeau, & Stewart, 2014; Woicik et al., 2009). The personality dimensions, namely, impulsivity and sensation seeking are associated with a vulnerability to positive reinforcement and positively rewarding effects of substances (Woicik

et al., 2009). Sensation seeking is characterized by the desire for intense and novel experiences and is specifically linked to substance use to attain positive affect (Castellanos-Ryan, Rubia, & Conrod, 2011; Woicik et al., 2009). Impulsivity, finally, is defined as the inability to control behavior when faced with immediate (positive) reinforcement (Castellanos-Ryan et al., 2011). From this theoretical perspective, it can be concluded that each personality dimension is related to specific risky or harmful motives for substance use that may lead to subsequent SUD.

Screening and assessment of clients' personality profile enables clinicians to provide personality-targeted treatment. This type of treatment aims at training competencies to clients to deal with specific personality dimensions and associated motives for substance use using motivational interviewing and cognitive behavioral therapy. By training personality-specific skills to improve management of personality risk, substance use linked to the specific personality profile will be reduced. Personality-targeted interventions do not result in changes in personality, but they change the relationship between personality dimensions and substance use. Personality-based treatment is more effective in reducing substance use in people with average intelligence than interventions not differentiating on individual factors (Conrod et al., 2013). As these personality dimensions are a proxy of behavioral and mental problems, personality-based treatment is also a promising strategy for effective treatment in individuals with ID as comorbid behavioral and mental health problems are highly prevalent in this group.

Examples of successful personalized treatments in individuals with ID are the treatment programs *Take it Personal!* and *Take it Personal!*+. These programs are specifically developed and adjusted to the needs and learning style of individuals with ID. Both programs integrate a personality-focused approach with elements of existing effective treatment protocols such as motivational interviewing and cognitive behavioral therapy. The programs consist of the following components that are crucial for successful treatment of SUD in individuals with ID: (a) motivation to behavior change, (b) psychoeducation regarding personality profile, (c) setting goals and make a plan to change, (d) recognition of personality profile and coherent signals of problematic behavior, (e) functional analysis, (f) increasing self-control, (g) behavioral coping training and cognitive coping training, and (h) relapse prevention. The *Take it Personal!*interventions focus on psychoeducation about the participants' personality profile and related problematic coping behavior such as substance use or aggression. Clients become familiar with their personality profile and learn to deal with their personality through exercises. Daily life experiences and physical, cognitive, and behavioral reactions will be analyzed. In the intervention participants will set individual goals, which they will encounter during the training. Clients will identify personality-specific thoughts and cognitions that lead to problematic behavior. For example, the intervention aimed at persons with the personality profile "Impulsive" will focus on "thinking before taking action." Simultaneously, the participants will be trained to use cognitive restructuring techniques to counter such tendencies. Participants make a personalized "changing plan" aimed at changing their problematic behavior related to their personality profile. To meet the needs of people with ID, the interventions consist of two weekly A and B sessions paying attention to the same topic. In one of the sessions, a confidential person (i.e., a person from the social network of the client or a professional caregiver) will be present. This design provides in the needs of self-control and support of people with ID. Also, the content of the treatment will be repeated, which is essential in treatment for people with intellectual disabilities. In addition, the generalizability of the treatment to everyday life will be supported by the confidential person.

The next step in personalized care in treatment of SUD in individuals with ID could take into account the highly dynamic and idiosyncratic nature of treatment, which is often neglected in the traditional and more conventional approaches. Such treatment should target, time, and adapt effective intervention efforts to meet the personal

and dynamically changing needs of clients, especially if clients are immersed in highly volatile developmental processes, such as individuals who are recovering from addiction. The dynamic and idiosyncratic nature of treatment processes and how these processes affect treatment outcomes is currently not well understood. As a consequence, there are no reliable guidelines to help clinicians create adaptive, personalized treatments. Clinicians establish their treatment plans based on intuition and clinical experience, without any standardized assistance.

The personalized network approach is an approach that could offer an attractive client-centered alternative to more conventional ways of looking at treatment of SUD in individuals with ID (Borsboom & Cramer, 2013; Fried et al., 2017). From a Dynamic Systems perspective, mental disorders (among which SUDs) are not understood as entity-like categories caused by one underlying factor but as networks of self-organizing and self-maintaining components of cognition, behavior, emotion, and somatic functioning (Schiepek, Heinzel, Karch, Plöderl, & Strunk, 2016). Treatment can be seen as a perturbation to that network, potentially triggering a reorganization towards more healthy patterns of functioning (Hayes, Yasinski, Barnes, & Bockting, 2015).

The approach starts with the creation of Idiographic System Modelling (ISM) components, which requires a session in which the client and the clinician together try to describe the current situation or problem in terms of symptoms and problems over the last weeks or months. In the second step, the intercorrelations between the components are graphically mapped in a procedure that takes two to three sessions, ensuring that clients will find the ISMs meaningful. In a third step, the ISM components are translated into personalized assessments for daily process monitoring leading to a set of personalized questions, such as: "Did I sleep well today?" The fourth step consists of daily monitoring of these symptoms resulting in contextualized and personalized process data that should be accessible to the therapist and the client at all time.

We argue that conventional Routine Outcome Monitoring (ROM)-data should be expanded or replaced with more contextualized and personalized process data that provide insight in the variations in clients' daily life experiences in their real-life contexts. Enriching standardized ROM data will help to better understand long-term treatment outcomes in the context of short-term idiosyncratic variations. Combining standardized and personalized outcome data is essential for significantly enhancing treatment outcomes by allowing clinicians to tailor their interventions and provide more personalized care, and for clients to gain more insight into their conditions and control over their treatment process.

Conclusion

Although significant progress has been made during the last decade, the evidence base of treatment of substance abuse in individuals with ID is still small. The literature describes a small range of different intervention approaches (e.g., education, motivational interviewing, cognitive behavioral techniques) that have been used in different settings (e.g., secure unit in forensic facility, services for ID care). As can be expected, interventions were adapted to the needs and learning style of individuals who had moderate/mild ID to borderline intellectual functioning. Most studies were directed towards increasing motivation to prevent, reduce substance or stop substance (ab) use. However, educating individuals with ID about the (adverse) consequences of substance (ab)use did not lead to decrease in actual substance use. As far as we know, interventions targeting illicit drugs or prescribed medications in individuals with IDD have not been reported in the literature. Neither have studies on interventions for other types of addictions such as excessive gaming and internet use in individuals with ID. For example, Jenaro et al. (2018) found that many individuals among a sample of 216 youth with ID show excessive patterns of use of internet which was associated with increased levels of psychological distress.

Case Identification

Screening for and assessment of both ID and substance (ab)use are important when designing treatments. These include both identifying individuals with moderate/mild ID and borderline intellectual functioning in addiction medicine or forensic psychiatric hospitals and recognizing substance (ab)use in those with ID. The first is not routinely done yet, even though there are some promising developments in these settings. For example, Braatveit, Torsheim, and Hove (2018) used the WAIS-IV identifying ID in 84 inpatients of treatment facilities for substance use disorder. Results showed that mean full scale IQ was 87 (range 61–118); mean Vineland II score was 96 (range 50–120). Among this sample, 7% was classified with an ID, 25% had borderline intellectual functioning, and 68% had average intellectual functioning. It should be noted that prior to the study, none of the inpatients was diagnosed with an ID. The authors used the Hayes Ability Screening Index and found that the screener had good psychometric properties for screening for ID among inpatients with SUD.

Substance (ab)use may be especially high among individuals with ID who reside in forensic or mental health care settings. For example, in a retrospective file study, Salavert et al. (2018) assessed prevalence rates of different types of substances among 88 clients with ID who were admitted to a psychiatric hospital over a period of 10 years. Almost half of the sample had mild ID, 3% had moderate ID, 3% had severe ID, and in the remaining cases the ID was unspecified. More than 35% of the sample met criteria for a substance use disorder, and most often this was related to cannabis (25%), alcohol (22%), and cocaine (14%). Most clients (ab)used more than one type of substance. Triple diagnosis was common (also see "Triple Diagnosis").

Assessment of substance (ab)use and substance use patterns on an individual basis is probably the most straightforward way to improve early detection and intervention in individuals with ID. There is a scarcity of instruments that have been shown reliable and valid in assessing substance use and abuse of individuals with ID (also see Chap. 18). VanDerNagel, Kiewik, Buitelaar, and De Jong (2011) have developed the *Substance Use and Misuse in Intellectual Disability – Questionnaire*(SumID-Q). This instrument is adapted for use in individuals with mild ID or borderline intellectual functioning (IQ 50–85) and measures substance use, its risk factors, and consequences. In an interview format, substance use and abuse are discussed in an empathic, open, and non-confrontational manner with the client. If outcomes of the SumID-Q reveals that an individual uses one or more substances, further assessment is necessary to reveal whether DSM-5 criteria for a substance use disorder are met, and how substance use is related to biological, social, and psychological risk factors. The diagnostic process includes a clinical interview, retrieval of information from significant others (family or professional care givers), and a comprehensive health check. Special attention should be given to the possibility of polysubstance use, and co-occurring symptoms of a psychiatric disorder. The latter can be both result of SUD and a risk factor for SUD, and generally warrants a comprehensive multicomponent treatment approach.

VanDerNagel, Kiewik, Dijk, et al. (2017) compared the outcomes of the self-report version of the SumID-Q to the SumID-Q proxy version (completed by clients' caregivers) to biomarkers (data collected by hair, urine, sweat patches) of substance use in 112 clients with mild ID to borderline intellectual functioning who lived in several Dutch facilities providing care to clients with ID. The authors found that agreement between the three strategies varied across substances and type of biomarker. It was found that biomarker analysis seemed of limited additional value compared to self-report and proxy report in the assessment of substance use, especially considering the additional costs and lower willingness of clients to participate in biomarker analysis.

Collaboration Between Organizations

In many cases, treatment of substance abuse in individuals with ID requires collaboration

between facilities in ID care, addiction medicine, and mental health care. However, staff in these facilities report a lack of expertise when working with these clients. A survey by VanDerNagel et al. (2011) among 39 ID care organizations in the Netherlands showed that most had inadequate expertise with substance use of clients with ID. Respondents also noted that substance users face a number of psychosocial problems that the service providers were poorly equipped to address. Individuals with ID experience barriers to accessing substance abuse treatment, for example in addiction services. When in treatment, the drop out may be relatively high. For example, McGillivray, Gaskin, Newton, and Richardson (2016) found that the drop out of alcohol and/or drugs programs in prison was much higher in prisoners with ID than in those prisoners without ID. The higher drop out levels were attributed to staff who were inexperienced in providing treatment to individuals with ID. Many individuals with ID may have negative experiences with treatment in mainstream addiction centers (see Taggart, McLaughlin, Quinn, & McFarlane, 2007).

Van Duijvenbode et al. (2015) and other researchers have identified a need for more cross-system collaboration and the use of integrated treatment approaches for the benefit of individuals with dual and triple diagnosis. Cross-system collaboration also implies involvement of ID services in prevention, care, and treatment for those with comorbid SUD and ID. This includes establishing policies regarding the prevention of substance use by clients, and staff members to protect other clients and staff members from the harmful effects and undesirable role models of clients' (and staff's) substance use, while avoiding repressive policies that may discourage clients to seek help. Organizations of ID care need to acknowledge SUD as a complex and potentially serious health problem that warrants clinical attention, intensified staff support, and possibly referral to an addiction center, rather than seeing SUD as a behavioral problem that can be remediated by relatively simple measures (Van Duijvenbode et al., 2015). Addiction centers, on the other hand, not only need to adapt their treatment protocols and patient communication to the need and learning style of those with ID, but also need to learn how to work together with and learn from staff in ID care organizations to provide optimal care for this patient group.

References

Borsboom, D., & Cramer, A. O. (2013). Network analysis: An integrative approach to the structure of psychopathology. *Annual Review of Clinical Psychology, 9*, 91–121.

Braatveit, K., Torsheim, T., & Hove, O. (2018). Screening for intellectual disabilities: A validation of the Hayes ability screening index for in-patients with substance use disorder. *Nordic Journal of Psychiatry, 72*, 387–392.

Castellanos-Ryan, N., Rubia, K., & Conrod, P. J. (2011). Response inhibition and reward response bias mediate the predictive relationships between impulsivity and sensation seeking and common and unique variance in conduct disorder and substance misuse. *Alcoholism Clinical Experimental Research, 35*, 140–155.

Comeau, N., Stewart, S. H., & Loba, P. (2001). The relations of trait anxiety, anxiety sensitivity, and sensation seeking to adolescents' motivations for alcohol, cigarette, and marijuana use. *Addictive Behaviors, 26*, 803–825.

Conrod, P. J., O'Leary-Barrett, M., Newton, N., Topper, L., Castellanos-Ryan, N., Mackie, C., et al. (2013). Effectiveness of a selective, personality-targeted prevention program for adolescent alcohol use and misuse: A cluster randomized controlled trial adolescent alcohol misuse prevention. *JAMA Psychiatry, 70*, 334–342.

Conrod, P. J., Pihl, R. O., & Vassileva, J. (1998). Differential sensitivity to alcohol reinforcement in groups of men at risk for distinct alcoholism subtypes. *Alcoholism Clinical and Experimental Research, 22*, 585–597.

Cooper, L. M., Frone, M. R., Russell, M., & Mudar, P. (1995). Drinking to regulate positive and negative emotions: A motivational model of alcohol use. *Journal of Personality and Social Psychology, 69*, 990–1005.

Didden, R., Nijman, H., Delforterie, M., & Keulen-De Vos, M. (2019). Treatment of anger and violence in individuals with intellectual disability. In W. Lindsay, L. Craig, & D. Griffiths (Eds.), *The Wiley handbook of what works for offenders with intellectual and developmental disabilities: An evidence-based approach to theory, assessment and treatment (pp. 297-309)*. London, UK: Wiley.

European Monitoring Centre for Drugs and Drug Addiction. (2015). *Comorbidity of substance use and mental disorders in Europe*. Lisbon, Portugal:

European Monitoring Centre for Drugs and Drug Addiction.

Fried, E. I., van Borkulo, C. D., Cramer, A. O., Boschloo, L., Schoevers, R. A., & Borsboom, D. (2017). Mental disorders as networks of problems: A review of recent insights. *Social Psychiatry and Psychiatric Epidemiology, 52*, 1–10.

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.

Hassiotis, A., Gazizova, D., Akinlonu, L., Bebbington, P., Meltzer, H., & Strydom, A. (2011). Psychiatric morbidity in prisoners with intellectual disabilities: Analysis of prison survey data for England and Wales. *British Journal of Psychiatry, 199*, 156–157.

Hayes, A. M., Yasinski, C., Barnes, J. B., & Bockting, C. L. (2015). Network destabilization and transition in depression: New methods for studying the dynamics of therapeutic change. *Clinical Psychology Review, 41*, 27–39.

Hecimovic, K., Barrett, S. P., Darredeau, C., & Stewart, S. H. (2014). Cannabis use motives and personality risk factors. *Addictive Behaviors, 39*, 729–732.

Holden, P., & Neff, J. A. (2000). Intensive outpatient treatment of persons with mental retardation and psychiatric disorder: A preliminary study. *Mental Retardation, 38*, 27–32.

Hronis, A., Roberts, A., & Kneebone, I. (2017). A review of cognitive impairments with children with intellectual disabilities: Implications for cognitive behaviour therapy. *British Journal of Clinical Psychology, 56*, 189–207.

Insel, T. R., & Cuthbert, B. N. (2015). Medicine. Brain disorders? Precisely. *Science, 348*, 499–500.

Jenaro, C., Flores, N., Cruz, M., Perez, M., Vega, V., & Torres, V. (2018). Internet and cell phone usage patterns among young adults with intellectual disabilities. *Journal of Applied Research in Intellectual Disabilities, 31*, 259–272.

Jurewicz, H. (2017). *The challenged addict, addiction recovery with concurring developmental disorders*. Madison, CT: Bick Publishing House.

Kerr, S., Lawrence, M., Darbyshire, C., Middleton, A., & 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. (2018). *Prevention and intervention of substance use and misuse in persons with intellectual disabilities*. Doctoral thesis, Radboud University, Nijmegen, the Netherlands.

Kiewik, M., VanDerNagel, J., Engels, R., & de Jong, C. (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.

Kiewik, M., VanDerNagel, J., Engels, R., & De Jong, C. (submitted). *Cognitive behavior therapy for adults with mild to borderline intellectual disabilities and substance use disorders: a feasibility study*.

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*, 216.

Lambert, M., LePage, J., & Schmitt, A. (2003). Five-year outcomes following psychiatric consultation to a tertiary care emergency room. *American Journal of Psychiatry, 160*, 1350–1353.

Langas, A.-M., Malt, U., & Opjordsmoen, S. (2011). Comorbid mental disorders in substance users from a single catchment area. A clinical study. *BMC Psychiatry, 11*, 25.

Lindsay, W. (2009). Adaptations and developments in treatment programmes for offenders with developmental disabilities. *Psychiatry, Psychology and Law, 16*, S18–S35.

McGillivray, J., Gaskin, C., Newton, D., & Richardson, B. (2016). Substance use, offending and participation in alcohol and drug treatment programmes: A comparison of prisoners with and without intellectual disabilities. *Journal of Applied Research in Intellectual Disabilities, 29*, 289–294.

Minkoff, K. (2001). Level of care determination for individuals with co-occurring psychiatric and substance disorders. *Psychiatric Rehabilitation Skills, 5*, 163–196.

Najavits, L. (2002). *Seeking safety: A treatment manual for PTSD and substance abuse*. New York, NY: Guilford Press.

National Institute on Drug Abuse (2012). *Principles of drug addiction treatment: A research-based guide* (3rd ed.). Bethesda, MA: NIDA.

Poelen, E. A. P., Schijven, E. P., Otten, R., & Didden, R. (2016). Personality dimensions and substance use in individuals with mild to borderline intellectual disabilities. *Research in Developmental Disabilities, 63*, 142–150.

Sakdalan, J., Kittner, D., & Judd, D. (2017). ASAP-ID: Substance abuse programme for a forensic ID population. *Journal of Intellectual Disabilities and Offending Behaviour, 8*, 157–164.

Salavert, J., Clarabuch, A., Fernandez-Gomez, F., Barrau, V., Giraldez, M., & Borras, J. (2018). Substance use disorders in patients with intellectual disability admitted to psychiatric hospitalisation. *Journal of Intellectual Disability Research, 62*, 923–930.

Schiepek, G., Heinzel, S., Karch, S., Plöderl, M., & Strunk, G. (2016). Synergetics in psychology: Patterns and pattern transitions in human change processes. In G. Wunner & A. Pelster (Eds.), *Selforganization in complex systems: The past, present, and future of Synergetics* (pp. 181–208). Cham, Switzerland: Springer.

Slayter, E. (2010). Disparities in access to substance abuse treatment among people with intellectual disabilities and serious mental illness. *Health and Social Work, 35*, 49–59.

Taggart, L., McLaughlin, D., Quinn, B., & McFarlane, C. (2007). Listening to people with intellectual disabilities who misuse alcohol and drugs. *Health and Social Care in the Community, 15*, 360–368.

Tsimopoulou, I., Stenfert Kroese, B., Unwin, G., Azmi, S., & Jones, C. (2018). A case series to examine whether people with learning disabilities can learn prerequisite skills for cognitive behaviour therapy. *The Cognitive Behaviour Therapist, 11*, e1.

Van Duijvenbode, N., VanDerNagel, J., Janssen van Raay, M., & Didden, R. (2019). Triple trouble: tobben of teamwork? [Triple trouble: tricky or teamwork?]. *De Psycholoog, 54*, 10-19.

Van Duijvenbode, N., VanDerNagel, J. E. L., Didden, R., Engels, R., Buitelaar, J., Kiewik, M., et al. (2015). Substance use disorders in individuals with mild to borderline intellectual disabilities: Current status and future directions. *Research in Developmental Disabilities, 38*, 319–328.

VanDerNagel, J., Kemna, L., Barendregt, C., & Wits, E. (submitted). *Recognized and treated accordingly: improving access to addiction care for individuals with mild intellectual disabilities*.

VanDerNagel, J., & 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, The Netherlands: Perspectief.

VanDerNagel, J., Kiewik, M., Buitelaar, J., & De Jong, C. (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., Kiewik, M., & Didden, R. (2017). *Handboek LVB en verslaving [handbook mild ID and addiction]*. Amsterdam, The Netherlands: Boom.

VanDerNagel, J., Kiewik, M., Dijk, V., Didden, R., Korzilius, H., … De Jong, C. (2017). Substance use in individuals with mild to borderline intellectual disability: A comparison between self-report, collateral-report, and biomarker analysis. *Research in Developmental Disabilities, 63*, 151–159.

Woicik, P. A., Stewart, S. H., Phil, R. O., & Conrod, P. J. (2009). The substance use risk profile scale: A scale measuring traits linked to reinforcement-specific substance use profiles. *Addictive Behaviors, 34*, 1042–1055.

33

Treatment of Aggression and Property Destruction in Persons with Dual Diagnosis

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Individuals with autism spectrum disorders (ASD) and intellectual/developmental disabilities (I/DD) sometimes display severe aggression and property destruction. As the individual advances in age, such behavior can be exceptionally challenging to families, school personnel, and other care providers. At times it becomes difficult for a family to find an appropriate residence, school placement, or adult program as a result of such extreme behavior (Emerson et al., 2001). Aggression can take many forms, including but not limited to biting, scratching, kicking, slapping, punching, and hair pulling (Thompson, Fisher, Piazza, & Kuhn, 1998). Property destruction can range from simple and relatively unharmful forms such as rending a magazine to destroying televisions and computers or breaking windows (Fisher, Greer, Fuhrman, Saini, & Simmons, 2018).

In this chapter, we will describe a progression of interventions to consider when aggression and property destruction are displayed by individuals with ASD or I/DD. A first step is to identify the function of the behavior (i.e., why is it occurring?) and strengthen alternative behavior accordingly. Most commonly, aggression and property destruction appear to be inadvertently socially reinforced (Beavers, Iwata, & Lerman, 2013). That is, the behavior produces positive reinforcement in the form of some sort of attention or reaction (Thompson et al., 1998), positive reinforcement in the form of some sort of tangible item such as food or a preferred toy or item (Mace, Pratt, Prager, & Pritchard, 2011), or negative reinforcement in the form of escape or avoidance from instructional activity (Zangrillo, Fisher, Greer, Owen, & DeSouza, 2016), self-care or medical activity (Allen, Loiben, Allen, & Stanley, 1992), or even social contact (Hagopian, Wilson, & Wilder, 2001). Thus, the first step is to teach the individual new ways to obtain the reinforcer(s) previously maintaining aggression or property destruction and to then teach periods of tolerance and delays to reinforcement.

At times, the behavior presents a significant threat of danger and must be stopped immediately. In such cases, substantial modifications are sometimes made in the home, work, or school environment in order to reduce or eliminate the motivation to engage in such behavior. For example, in the case of escape-maintained behavior, there may be a need to temporarily cease all instructional activity to ensure that the behavior does not occur (Piazza, Moes, & Fisher, 1996). Then, the challenge becomes fading the instructional activity back into the schedule. Similarly, in the case of attention-maintained behavior, it may be necessary to provide an individual with

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free and continuous noncontingent attention (Hagopian, Fisher, & Legacy, 1994). Then, the challenge becomes fading away the intensive levels of attention. Examples of this approach will be described further.

The most intrusive interventions for aggression and property destruction involve punishment, seclusion, and restraint. These approaches come with a host of risks, problems associated with social acceptability, and even legal considerations. We will present some of the considerations when using intrusive interventions as a component of treatment for aggression and property destruction.

Last, we will present some ideas for future research on the treatment of aggression and property destruction. For example, it is possible that some features of aggression are phylogenic in nature (as evidenced by animal models of aggression) and may be therefore relatively insensitive to operant-based treatment approaches. Evidence for such phenomena will be described.

Interventions

Below, we will describe commonly used or commonly recommended interventions based on a functional analysis of the behavior. In each case, we will comment on limitations, and we conclude that differential reinforcement of alternative behavior (DRA), which includes functional communication training (FCT) as a variant, is the most supported approach. Other interventions could serve as logical components to an overall treatment package.

Applied behavior-analytic interventions for aggression and property destruction maintained by positive or negative reinforcement often include differential reinforcement (Kunnavatana, Bloom, Samaha, Slocum, & Clay, 2018; Vollmer & Iwata, 1992; Vollmer, Iwata, Zarcone, Smith, & Mazaleski, 1993; Vollmer, Roane, Ringdahl, & Marcus, 1999), noncontingent reinforcement (NCR; Lalli, Casey, & Kates, 1997; Lambert, Bloom, Samaha, Dayton, & Kunnavatana, 2016; Slocum, Grauerholz-Fisher, Peters, & Vollmer, 2017), extinction (Fisher et al., 1993; Hanley, Piazza, Fisher, & Maglieri, 2005), punishment (Hagopian, Fisher, Thibault Sullivan, Acquisto, & LeBlanc, 1998; Hanley et al.), or some combination. If a functional analysis (Iwata, Dorsey, Slifer, Bauman, & Richman, 1982/1994) suggests that aggression or property destruction is socially mediated by positive or negative reinforcement, it is important to consider a range of factors before selecting an intervention. Some of these factors include the age and size of the individual, how often aggression or property destruction occurs, the severity of aggression or property destruction, the context in which aggression or property destruction occurs (e.g., school, home, medical office), and the conditions that might influence whether stakeholders (e.g., caregivers, teachers) are able to implement the treatment with a high level of integrity.

Differential Reinforcement

DRA is a well-established intervention to reduce problem behavior, such as aggression or property destruction (Briggs, Fisher, Greer, & Kimball, 2018; Hagopian et al., 1998). The implementation of DRA involves providing a higher rate, quality, and magnitude of reinforcement contingent on appropriate behavior rather than problem behavior. One example of DRA is functional communication training (FCT), wherein the alternative response is a communicative response that produces the same reinforcer previously maintaining problem behavior (see Fig. 33.1). However, not all DRA is FCT. For example, sometimes it is best to reinforce behavior such as compliance with instructional activity with positive reinforcers, even though the problem behavior is maintained by escape (see Fig. 33.2, e.g., Slocum & Vollmer, 2015). DRA may or may not include extinction; in an ideal case, it would, but sometimes it is nearly impossible to entirely place dangerous behavior on extinction (Athens & Vollmer, 2010). According to the matching law, responding should shift from one behavior (i.e., problem behavior) to another behavior (i.e., appropriate behavior) given that appropriate behavior is more likely to result in reinforcement

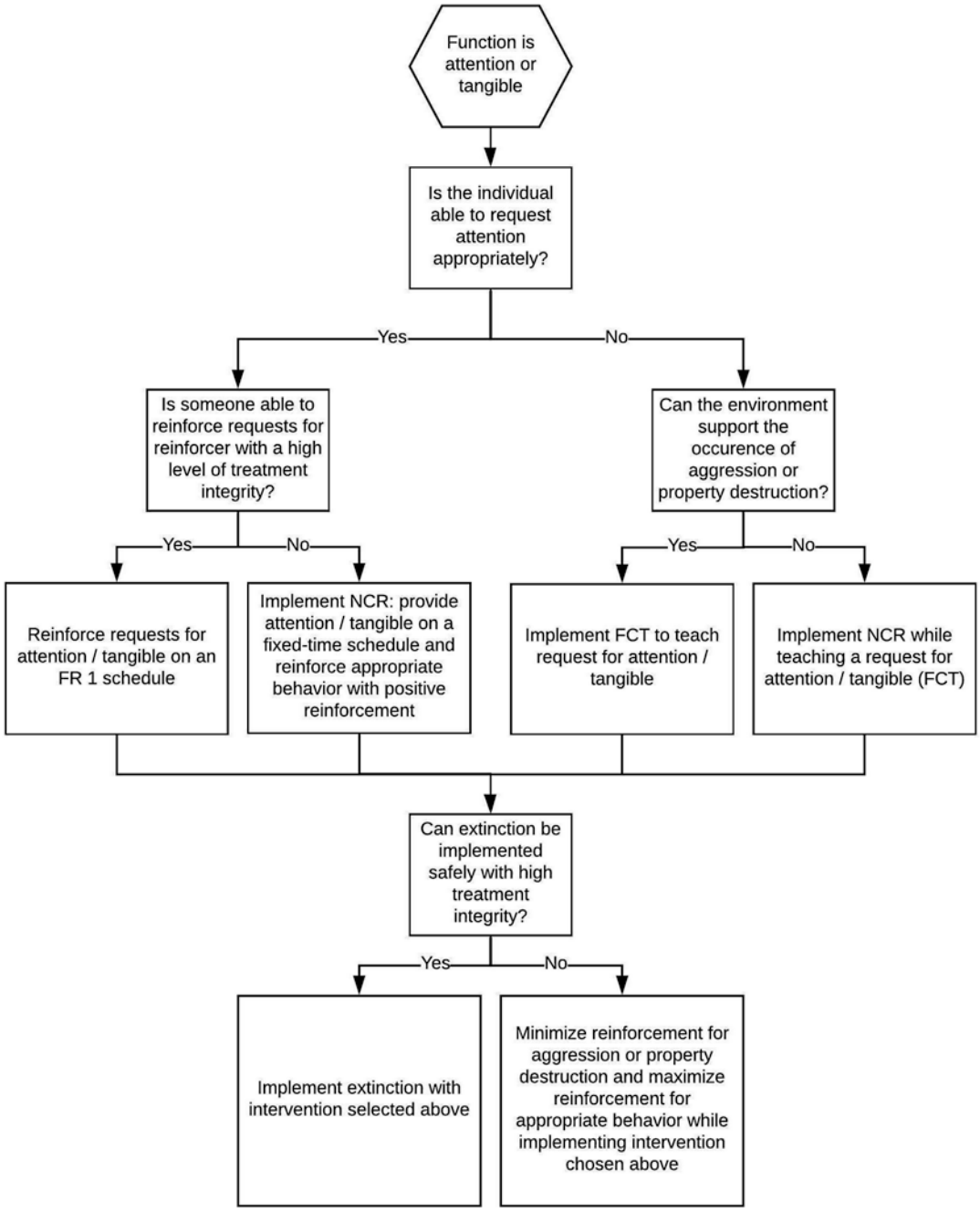

Fig. 33.1 A diagram depicting possible steps to safely minimize aggression and property destruction sensitive to positive reinforcement in the form of access to attention or tangibles

(Borrero & Vollmer, 2002; Herrnstein, 1961; Jacobs, Borrero, & Vollmer, 2013).

The use of DRA as treatment is often chosen for many reasons. First, it is less likely than extinction alone to produce negative side effects, such as emotional responding, extinction bursts, and a decreased likelihood of the occurrence of extinction-induced variability, because the reinforcer is still available contingent on an appropriate response (Lerman & Iwata, 1996). Vollmer, Borrero, Lalli, and Daniel (1999) evaluated DRA with two individuals whose severe

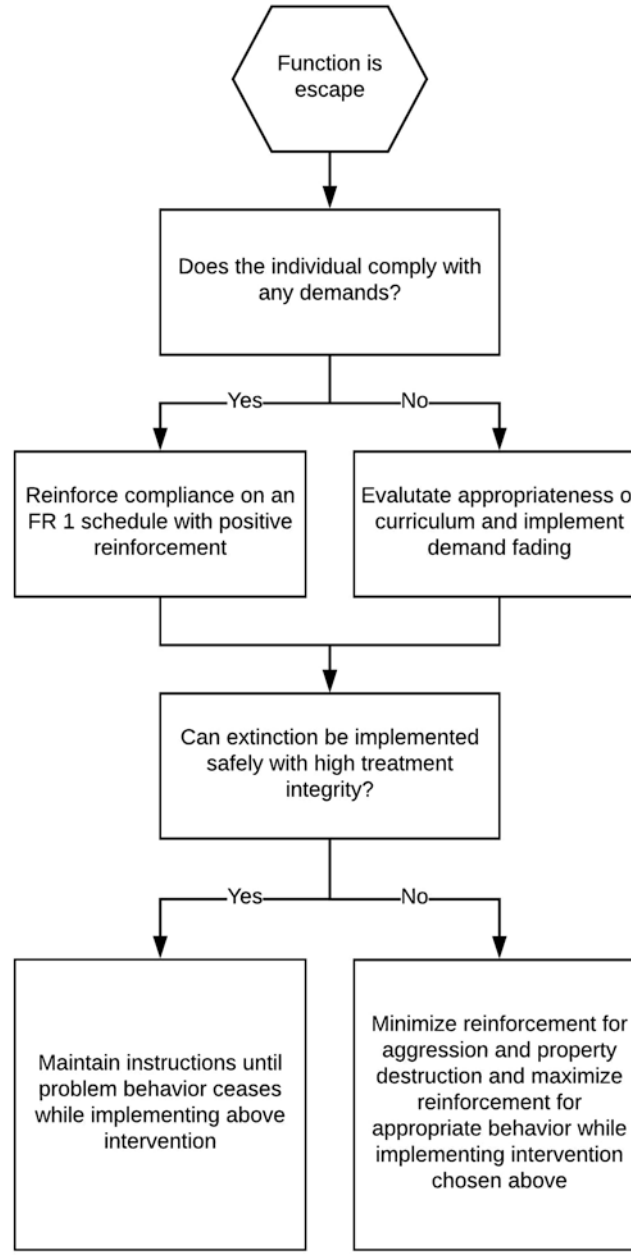

Fig. 33.2 A diagram depicting possible steps to safely minimize aggression and property destruction sensitive to negative reinforcement in the form of escape from aversive stimuli

aggression was shown to be reinforced by preferred items (i.e., food and television). After teaching both individuals to appropriately mand (i.e., request), an increase in appropriate behavior and a reduction in aggression was obtained. Both individuals, however, required a signal to indicate when the reinforcer was and was not available (e.g., "please wait") to maintain low rates of aggression as the delay to reinforcement increased.

Although the variant of DRA that is FCT is highly effective in reducing problem behavior maintained by attention or tangibles, it may not be ideal for escape-maintained problem behavior. Its use would likely lead to a reduction in problem behavior allowing the individual to escape an aversive situation by providing a break contingent upon a request; however, it may also significantly reduce learning opportunities and fail to address the factors that are resulting in an aversive situ-

ation in the first place. Recently, studies have shown that providing positive reinforcement for alternative behavior, such as compliance, rather than a request, may actually be more effective than negative reinforcement and may still be effective without extinction in most cases (see Fig. 33.2). Slocum and Vollmer (2015) compared the effects of positive and negative reinforcement in five participants, ranging in age between 4 and 8 years, three of whom had been diagnosed with ASD and one had been diagnosed with an I/DD. Results indicated that negative reinforcement, or removal of demands, provided contingent upon compliance was not effective in reducing problem behavior for three of the five participants. In contrast, positive reinforcement delivered contingent upon compliance was effective at reducing problem behavior for all five participants. Apart from the advantage of being able to avoid the use of extinction, the procedure resulted in an increase in compliance, which likely also leads to an increased skill acquisition.

A variation of DRA is differential reinforcement of other behavior (DRO). Unlike DRA, DRO does not identify an alternative appropriate behavior that will be reinforced in place of the problem behavior (Jessel & Borrero, 2014; Jessel & Ingvarsson, 2016). Rather, a reinforcer is delivered after a specific interval of time elapses without the occurrence of problem behavior. The intervention poses several problems which include:

(a) An appropriate alternative behavior is not targeted for increase in place of the problem behavior.
(b) The overall density of reinforcement an individual receives may be reduced.
(c) Other maladaptive or inappropriate behavior, not targeted for reduction, may be inadvertently reinforced.
(d) Reductions in problem behavior may be lower in comparison to interventions that include DRA.
(e) High levels of procedural integrity are required to avoid treatment failures (Athens & Vollmer, 2010; Vollmer, Sloman, & St. Peter Pipkin, 2008).

Therefore, DRO is not often the ideal choice for behavior reduction, at least as an isolated procedure, especially when the behavior is as severe as aggression or property destruction.

Extinction

Another behavior-reduction procedure, extinction, involves terminating the response-reinforcer contingency such that emitting a response that was previously reinforced no longer produces the reinforcer (Iwata, Pace, Cowdery, & Miltenberger, 1994; Lerman, Iwata, & Wallace, 1999). Extinction is often implemented in conjunction with DRA in that the problem behavior is no longer reinforced (Fisher et al., 1993; Hagopian et al., 1998), and instead the appropriate behavior is reinforced. It is important to note that extinction is difficult to implement with high fidelity in environments such as schools and homes due to safety risks (St. Peter Pipkin, Vollmer, & Sloman, 2010). For example, a teacher cannot withhold attention from a student that is aggressing toward another student or breaking equipment. For this reason, it can often be very difficult, or unsafe, to implement extinction in isolation, despite its potential effectiveness. If extinction cannot be implemented with high fidelity, the result will be that the behavior is reinforced on an intermittent schedule, such as a variable ratio (VR) or a variable interval (VI) schedule, which will increase its resistance to extinction, making it more difficult to reduce in the future (Worsdell, Iwata, Hanley, Thompson, & Kahng, 2000). Therefore, whenever possible, extinction should not be used in environments that cannot support the occurrence of problem behavior and should almost never be used without some form of reinforcement for alternative behavior in place, especially with behavior as severe as aggression and property destruction (see Figs. 33.1 and 33.2).

Noncontingent Reinforcement

NCR is the delivery of a reinforcer based on time, independent of responding (Vollmer et al., 1993). If an individual is provided access to the

reinforcer maintaining problem behavior noncontingently, the motivation to obtain that reinforcer will decrease, reducing the likelihood the individual will engage in problem behavior (e.g., aggression, property destruction) to obtain that reinforcer (Vollmer et al). Also, NCR eliminates the contingency between problem behavior and reinforcement, so it functions similarly to a form of extinction (Hagopian et al., 1994). Slocum et al. (2017) evaluated NCR for three participants who engaged in aggression sensitive to social positive reinforcement. Results suggest NCR was effective in decreasing aggression for all three participants. A variant of this procedure is momentary DRO (mDRO), wherein the reinforcer is delivered if and only if the problem behavior is not occurring at the scheduled time of delivery, in order to avoid adventitious reinforcement (e.g., Vollmer, Roane, et al., 1999). Further, NCR is most commonly considered a component of an overall treatment package, not the treatment itself, because no alternative behavior is explicitly strengthened (Vollmer & Sloman, 2005). Often, these additional components might include DRA or extinction (see Fig. 33.1; Lalli et al., 1997; Piazza, Contrucci, Hanley, & Fisher, 1997).

Punishment

Punishment-based procedures involve the delivery (positive punishment) or removal (negative punishment) of a stimulus contingent on the occurrence of a behavior to decrease the future probability of that behavior (Lerman & Vorndran, 2002). It is important to note that a punishment procedure is defined by its function on a behavior and not the topography of the procedure. For example, although removal of a student from class due to destructive behavior sensitive to social negative reinforcement might be called "punishment" by the teacher, disruptive behavior has been reinforced by removing the aversive stimulus (i.e., the classroom environment). Therefore, punishment procedures cannot be implemented for all individuals with the expectation that it will reduce the target behavior. It is also important to consider which procedure is appropriate to use based on (a) the laws and regulations, (b) caregiver preference, (c) the topography of the behavior, and (d) the size and strength of the client or student. Punishment procedures often described in the literature include time-out, hands-down, facial screens, and quiet hands (Donaldson & Vollmer, 2011; Fisher, Piazza, Bowman, Hagopian, & Langdon, 1994; Hanley et al., 2005). It is important to note that many of these procedures might be considered dated and inappropriate to consider in our current culture (e.g., water mist).

Furthermore, there are many questions one should consider prior to implementing a punishment-based procedure. First, have less intrusive reinforcement-based procedures been exhausted, and was the behavior insensitive to other forms of treatment? Second, is the behavior severe enough to warrant punishment? That is, would it be unethical to allow the behavior to occur, and is there a risk of injury to those interacting with the individual? Third, is it possible to combine a reinforcement-based procedure with the punishment-based procedure? Last, and most important, will the individual's caregiver(s) consent to the punishment-based procedure, and is it possible to have the procedure peer-reviewed by other professionals? If the answer is no to any of these questions, it is probably best to refrain from implementing punishment-based procedures. If, however, the answer to all of the questions is yes, punishment might be necessary to reduce the behavior. Research suggests that when DRA or DRA combined with extinction is ineffective in reducing problem behavior, the addition of punishment to DRA and extinction is often effective (Fisher et al., 1993; Hagopian et al., 1998). For example, Greer et al. (2013) implemented time-out, a negative punishment procedure, to reduce aggression and property destruction sensitive to social positive reinforcement. Results suggested that DRA and punishment were necessary to reduce problem behavior and increase appropriate behavior for all four participants.

Instructional Revision

When developing interventions for problem behavior maintained by escape, it is important to explore the reasons why demands or tasks are aversive. Some of the factors that may increase the aversiveness of tasks include long work periods, selecting tasks that are too difficult or easy given the skill set of the individual or are not functional, and insufficient levels of reinforcement for compliance (Geiger, Carr, & Leblanc, 2010). It has been shown that presenting shorter tasks (Moore, Anderson, & Kumar, 2005), providing a choice of activities, and modifying the difficulty of the task may all be effective strategies for reducing problem behavior maintained by escape (Dunlap, Kern-Dunlap, Clarke, & Robbins, 1991). For individuals in school environments or in intervention programs where the level of demands is likely to be high, it may be beneficial to conduct assessments one to two times a year to ensure that presented tasks are appropriate for the student's skill level and take data on compliance during longer work periods. Curricular modifications may be beneficial in preventing problem behaviors; however, appropriate selection of tasks and task length may require a higher level of expertise.

Transition to Natural Environments

In order to translate an effective treatment to settings such as home, school, or employment, several steps should be considered. First, it must be ensured that safe crisis management procedures are in place, independent of the behavior intervention plan. Second, the intervention plan must be arranged to be as practical and efficient as possible. Third, care providers must be effectively trained to conduct the procedures. Fourth, variables influencing behavior in the natural setting must be taken into account and effectively addressed.

Schedule Thinning Procedures

As reinforcement schedules are thinned to be more conducive to the natural environment, the reinforcer is delivered less often and may resemble extinction. One method commonly implemented is to provide competing activities when the reinforcer is unavailable (Austin & Tiger, 2015; Fisher, Kuhn, & Thompson, 1998; Fuhrman, Greer, Zangrillo, & Fisher, 2018; Hagopian, Contrucci Kuhn, Long, & Rush, 2005). Most recently, Fuhrman et al. compared schedule thinning with and without competing activities under a multiple schedule arrangement which signals the availability of reinforcement and extinction with discriminative stimuli. Results suggested that providing competing activities during schedule thinning produced a lower rate of problem behavior relative to schedule thinning without competing activities. However, no formal assessment was conducted to identify which reinforcers would be most effective in mitigating problem behavior during schedule thinning. It would be beneficial to compare different features of competing items and activities (e.g., matched vs. unmatched stimuli, demands for behavior sensitive to social positive reinforcement) to identify the most effective procedures to utilize within certain contexts. These procedures are not limited to DRA and should be used in conjunction with other procedures such as NCR (Slocum et al., 2017).

Care Provider Training

Training care providers to implement behavior interventions is important given that they spend a considerable amount of time with the individual. Behavioral skills training is a common method for caregiver training and has been effectively implemented to teach parents to carry out interventions for problem behavior (Van Camp et al., 2008), feeding disorders (Mueller, Piazza, Moore, & Kelley, 2003), and social skills deficits

(Dogan et al., 2017) and resulted in positive changes in child behavior (Marcus, Swanson, & Vollmer, 2001). The three main components of behavioral skills training are instructions (verbal or written), modeling, and role-play with feedback (Marcus et al.). The instructions provide an overview of the intervention and are often provided, while the trainer models the procedure either with the client, another trainer, or the individual being trained. Following a model, the trainee is asked to practice implementing the intervention either with the trainer or with their child. During the role-play, both corrective and positive feedback are provided either immediately after the care provider engages in the target response or at the end of the session. Training continues until the trainee has demonstrated that they are able to implement the intervention at a specified level of accuracy. Although it is common for all three components to be included in the training, it has been shown that opportunities to practice and receive feedback are the components that are necessary to ensure that care providers are able to correctly implement the procedures (Drifke, Tiger, & Wierzba, 2017; Severtson & Carr, 2012).

Variables Influencing Behavior

Within an individual's natural environment (e.g., school, home), a reinforcer may be available, but an error of omission (i.e., failure to deliver a reinforcer contingent on an appropriate response; St. Peter Pipkin et al., 2010) results in an increase in the problem behavior, called resurgence. Typically, the resurgence paradigm includes reinforcement of problem behavior, extinction of problem behavior while reinforcing appropriate behavior, and then extinction of problem and appropriate behavior, which may resemble omission errors an individual might encounter. Resurgence is typically described by an increase in problem behavior when problem and appropriate behavior are no longer reinforced (Briggs et al., 2018; Volkert, Lerman, Call, & Trosclair-Lasserre, 2009; see Fig. 33.3 for a diagram summarizing resurgence). Briggs et al. found

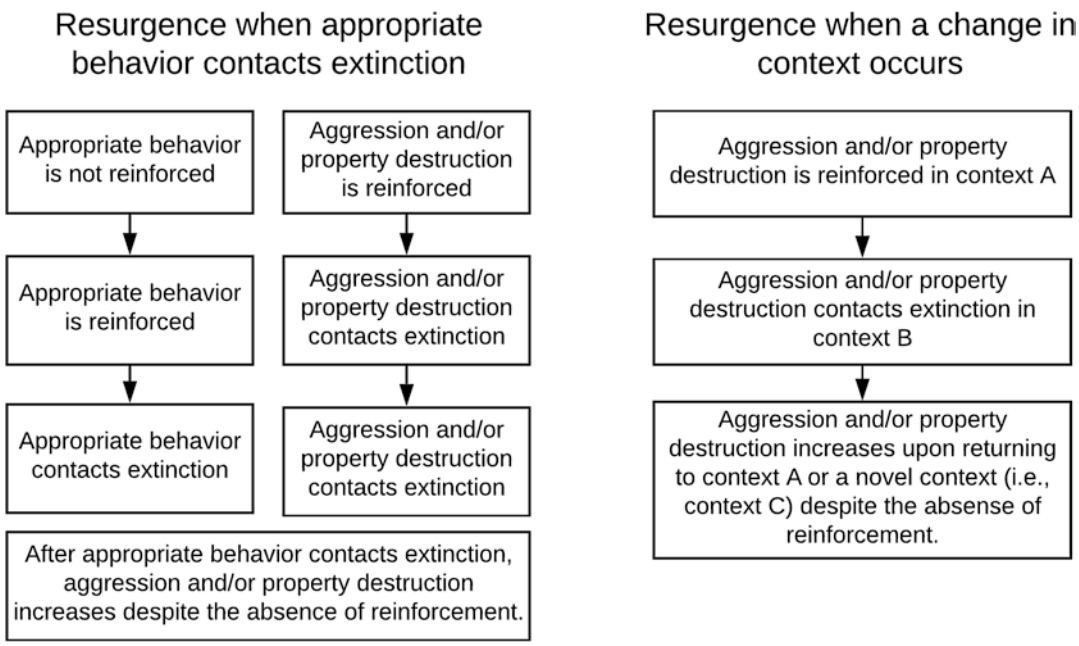

Fig. 33.3 A diagram depicting resurgence of aggression and/or property destruction when appropriate behavior contacts extinction

resurgence to occur in 10 out of 14 applications when transitioning from dense to lean reinforcement schedules for problem behavior sensitive to social positive reinforcement. These results suggest resurgence is prevalent and something one must account for when programming behavior-reduction procedures.

One method to mitigate resurgence is to incorporate multiple schedules. Multiple schedules signal the availability and unavailability of reinforcement using differently colored stimuli (Fuhrman, Fisher, & Greer, 2016; Landa & Hanley, 2016; Saini, Miller, & Fisher, 2016; Shamlian et al., 2016). Fuhrman et al. evaluated the effects of multiple schedules on resurgence of problem behavior and found lower rates of resurgence when implementing a multiple schedule relative to its absence. These results suggest the use of multiple schedules might mitigate problem behavior under conditions replicating errors of omission.

A second method to mitigate resurgence may include programming multiple topographies of alternative behavior (Bloom & Lambert, 2015; Lambert, Bloom, Samaha, Dayton, & Rodewald, 2015). Lambert et al. compared two DRA interventions, traditional and serial. Traditional DRA involved teaching a single alternative response, while serial DRA involved teaching multiple alternative responses. During the resurgence test, an increase in the alternative behavior was more likely to occur relative to the target (i.e., problem) behavior. Although these were arbitrary responses (switch flipping), the results suggest there is value to teaching multiple alternative (i.e., appropriate) responses to mitigate resurgence when an individual contacts extinction, whether it is programmed or occurs due to an omission error.

Instructional Fading

Some environments, such as a classroom, cannot support even one occurrence of aggression or property destruction, as it would pose a serious risk of harm to others. One option to prevent the occurrence of problem behavior is to implement instructional fading or the removal and systematic reintroduction of all demands that have been shown to evoke problem behavior in the past (Piazza et al., 1996). Prior to reintroducing demands, it may be beneficial to conduct a demand assessment in order to determine which tasks are most likely to evoke problem behavior. The assessment is conducted by tracking both rates of problem behavior and compliance for each specific instruction. Demands for which there are high compliance and low problem behavior are classified as high-probability requests (high-p), and those for which compliance is low and problem behavior is high are classified as low-probability requests (low-p). When reintroducing demands, it has been recommended that it is more effective to begin by presenting three to four high-p requests, for which every occurrence of compliance is reinforced, prior to presenting a single low-p request (Zuluaga & Normand, 2008). Instructional fading is effective at eliminating problem behavior through the removal of the aversive stimulus and any need for the individual to engage in problem behavior. The main advantage of this type of intervention is that problem behavior will likely drop to near-zero rates (Dunlap et al., 1991; Geiger et al., 2010). The disadvantage of removing demands is that there will be little to no instructional time which may reduce rates of skill acquisition, though in the long term those demands will be gradually reintroduced.

Crisis Management

When dealing with aggression and property destruction, it is probable that there will be moments when the behavior poses a risk of harm to the individual themselves, to others, or to the environment. These moments are often referred to a crisis and may require the use of physical containment to reduce the risk of harm. Prior to working with individuals with severe problem behavior, it is essential that one familiarizes themselves with the medical, ethical, and legal obligations that arise with these types of

behaviors. The moment it is known that the problem behavior poses a risk of harm, either because there is a record of it occurring in the past or because an incident occurred recently, a behavior intervention plan must be developed and approved by the individuals' parents or legal guardian. In addition, those implementing crisis management procedures must be properly trained not only on the procedures themselves but also in de-escalation techniques.

The two common components of a crisis management procedure are restraint and seclusion. *Restraint* is defined as restricting an individual's ability to move freely either by physically holding them or using a mechanical device for an extended period of time (Knox & Holloman, 2012; Vollmer et al., 2011). *Seclusion* is defined as the confinement of an individual from others in a separate area or room from which they are prevented from leaving (Knox & Holloman, 2012; Vollmer et al.). Both are emergency procedures used to prevent the occurrence of behavior that poses an immediate safety risk to the patient themselves or others and are not to be used as treatment or intervention for problem behavior.

Best practice in the use of restraint and seclusion, first and foremost, requires that neither of the procedures are used without an intervention being in place for problem behavior. If no risk of harm is present, the use of the procedure is not warranted. It is essential that guidelines are established that clearly outline what constitutes a crisis that would warrant the use of the procedures, such as environment and definition of the behavior, as well as termination criteria (Vollmer et al., 2011). When restraint or seclusion is part of any treatment plan, it is essential that it includes (a) other less intrusive interventions based on positive reinforcement, (b) a functional assessment, (c) ongoing evaluation based on objective data, (d) oversight and supervision by someone with experience in treating severe problem behavior, and (e) evidence-based, best practices from the current literature (APBA, 2010, Vollmer et al.).

Another consideration with the inclusion of restraint or seclusion as an emergency procedure is the welfare of the patient, their right to choose, and their right to be provided with the least restrictive treatment (Johnston & Sherman, 1993; Vollmer et al., 2011). Definitions of least restrictive treatments vary across disciplines. Within the field of behavior analysis, least restrictive treatment focuses more on outcomes and is commonly defined as the intervention that results in the greatest benefit and the least amount of risk while considering factors such as anticipated success and duration, as well as potential distress caused by both the behavior and the treatment procedure (ABAI, 2010; Vollmer et al.). In one case study, it was argued that the use of ambulatory restraints was the least restrictive treatment for a violent adolescent, as their use allowed her to be integrated with her peers and access other forms of treatment, something that was not possible without them (Troutman, Myers, Borchardt, Kowalski, & Bubrick, 1998). Definitions, however, that are based on the form of the treatment itself rather than the outcome of that treatment would most likely conclude that restraint would always be the most restrictive option (Johnston & Sherman, 1993).

The use of both restraint and seclusion remains controversial due to the potential risks posed to both those implementing the procedure (e.g., legal action) and the individual being restrained (e.g., injury or death). There is little evidence to support any therapeutic value from the use of restraint (Mohr, Petti, & Mohr, 2003); however, this may not always be the case for seclusion. Surveys conducted with patients following seclusion have indicated that a small percentage report positive effects, such as that it helped them calm down (Meehan, Bergen, & Fjeldsoe, 2004). The controversy arises from the fact that the use of restraint and seclusion has led to serious injury and death in several cases, although one could argue that risks are more likely to occur in cases of misuse or abuse of restraint. For example, in 2006, a 7-year-old girl suffocated after being restrained for 40 min at a mental health clinic that had at least two prior complaints of inappropriate use of restraint in the same year (Reynolds, 2006). In this case, the injury and subsequent death of the child were likely because the restraint was implemented incorrectly. In other cases, the use of restraint may have prevented an injury or

death. For example, a 40-year-old man with schizophrenia plunged head first into a window leading to a fractured neck and eventually death. The family was awarded $7 million because the nurse failed to restrain him and was determined to be negligent (Grant, 2005).

Position Statements

In an attempt to better define how and when restraint or seclusion is warranted, several organizations have released position statements on the topic. Both the Association for Behavior Analysis International (ABAI) and the Association of Professional Behavior Analysts continue to support the use of both as necessary emergency interventions in situations where the behavior could cause serious harm. Both require that it be included as part of a behavior intervention plan (BIP) with other less restrictive interventions that have been derived from a functional assessment (ABAI, 2010; APBA, 2010). Other medically based organizations such as the American Psychiatric Nurses Association (APNA) and the National Association of Psychiatric Healthy Systems (NAPHS) also support the use of both procedures in combination with less restrictive procedures; however, the APNA has included a goal to eliminate their use entirely, and the NAPHS requires debriefing immediately following each occurrence (APNA, Updated, 2014). All the organizations require oversight by a trained professional as well as specific training in crisis intervention and de-escalation techniques.

In contrast, one group, the Autism National Committee (AUTCOM), has taken the position of condemning the use of restraint in any circumstance as they believe it to be a direct violation of human rights of individuals with disabilities (AUTCOM, 1999). The World Health Organization (WHO) also published a document in which it is stated that "seclusion and restraint are never justified, even in extreme circumstances (such as when individuals behavior violently, are at potential risk of harm etc.)" (Funk & Drew, 2017, p. 30). Both insist that if an individual engages in a behavior that poses an imminent threat of harm, it is always a sign of treatment failure and that there was likely something that could have been done to prevent the incident from happening in the first place. Another group, the Council of Parent Attorneys and Advocates (COPAA), has not taken the position on a total ban of restraint; however, they have taken the position that prone restraints should be banned under all circumstances (COPAA, 2008, Updated 2011).

Restraint and seclusion, despite being controversial, may be necessary components in the treatment of aggression and property destruction in order to ensure the safety of both the individual engaging in the problem behavior and those around them. In cases where death or injury has occurred as a result of either procedure, it has been likely due to misuse or abuse rather than correct implementation. When faced with a situation in which someone is aggressing toward another individual and causing them bodily harm, it is unlikely that most people would argue that the aggressor's right to freedom overrides the victim's right to safety.

Alternative Mechanisms to Consider

Regardless of how precisely an intervention is designed and implemented, there are still situations in which problem behavior persists. When the topography of the behavior is aggression or property destruction, there is a possibility that behavior was thought to be maintained by escape and may actually be phylogenic or respondent behavior. It has been shown in several basic research studies that when presented with noxious or aversive stimuli humans and animals may engage in aggression by attacking or biting (Hutchinson, 1977). There have been several documented cases in animals, such as squirrel monkeys and rats, in which the presentation of an aversive stimulus, such as an electric shock, elicits an attack response either toward an inanimate object or another animal, and it does not decrease even when extinction is implemented (Azrin, Hake, & Hutchinson,

1965; Azrin, Hutchinson, & McLaughlin, 1965; Ulrich, 1966; Ulrich & Azrin, 1962). In a study conducted with humans, university students were asked to submerge their hands in cold water and were then given the opportunity to deliver noise blasts to a confederate (Berkowitz, Cochran, & Embree, 1981). When told that noise blast would be harmful to the confederates' performance, as opposed to being told it would be helpful, the subjects with their hands submerged in painfully cold water were more likely to deliver the blast to the confederate, in contrast to those subjects who had their hands submerged in temperate water. These findings suggest that in the presence of aversive stimuli, both humans and animals may engage in aggressive or retaliatory behavior that is not maintained by escape or negative reinforcement. Therefore, when aggressive behavior, such as biting, occurs in demand situations and is resistant to extinction, it may be important to consider that the behavior may be respondent rather than operant and not socially mediated. Much more research on this topic is needed.

Best Practice (Conclusions)

To summarize, best practice in the treatment of aggression and property destruction involves identification of the function of behavior (via assessment) and differential reinforcement of alternative behavior. Although it is not always possible to place severe behavior on extinction, reinforcement for problem behavior should be minimized in comparison to reinforcement for appropriate alternative behavior. Other techniques can be used to suppress behavior that is extremely dangerous. For example, continuous NCR can be provided to reduce the motivation to engage in dangerous behavior. However, the schedule of reinforcement would need to be signaled and systematically thinned. When restraining or seclusion is used, laws and ethical codes must always be followed.

References

Allen, K. D., Loiben, T., Allen, S. J., & Stanley, R. T. (1992). Dentist-implemented contingent escape for management of disruptive child behavior. *Journal of Applied Behavior Analysis, 25*, 629–636. https://doi.org/10.1901/jaba.1992.25-629

American Psychiatric Nurses Association. (2000; Updated 2014). *APNA position statement on the use of seclusion and restraint.*

Association for Behavior Analysis International. (2010). *Statement on restraint and seclusion*

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.*

Athens, E. S., & Vollmer, T. R. (2010). An investigation of differential reinforcement of alternative behavior without extinction. *Journal of Applied Behavior Analysis, 43*(4), 569–589. https://doi.org/10.1901/jaba.2010.43-569

Austin, J. E., & Tiger, J. H. (2015). Providing alternative reinforcers to facilitate tolerance to delayed reinforcement following functional communication training. *Journal of Applied Behavior Analysis, 48*, 663–668.

Autism National Committee (AUTCOM) (1999). *Position on restraints.*

Azrin, N. H., Hake, D. F., & Hutchinson, R. R. (1965). Elicitation of aggression by a physical blow. *Journal of the Experimental Analysis of Behavior, 8*(1), 55–57. https://doi.org/10.1901/jeab.1965.8-55

Azrin, N. H., Hutchinson, R. R., & McLaughlin, R. (1965). The opportunity for aggression as an operant reinforcer during aversive stimulation. *Journal of the Experimental Analysis of Behavior, 8*(3), 171–180. https://doi.org/10.1901/jeab.1965.8-171

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

Berkowitz, L., Cochran, S. T., & Embree, M. C. (1981). Physical pain and the goal of aversively stimulated aggression. *Journal of Personality and Social Psychology, 40*(4), 687–680. https://doi.org/10.1037/0022-3514.40.4.687

Bloom, S. E., & Lambert, J. M. (2015). Implications for practice: Resurgence and differential reinforcement of alternative responding. *Journal of Applied Behavior Analysis, 48*, 781–784.

Borrero, J. C., & Vollmer, T. R. (2002). An application of the matching law to severe problem behavior. *Journal of Applied Behavior Analysis, 35*(1), 13–27. https://doi.org/10.1901/jaba.2002.35-13

Briggs, A. M., Fisher, W. W., Greer, B. D., & Kimball, R. T. (2018). Prevalence of resurgence of destructive

behavior when thinning reinforcement schedules during functional communication training. *Journal of Applied Behavior Analysis, 51*, 620–633.

COPAA. (2008, Updated 2011). *Declaration of principles opposing the use of restraints, seclusion, and other aversive interventions upon children with disabilities*.

Dogan, R. K., King, M. L., Fischetti, A. T., Lake, C. M., Matthews, T. L., & Warzak, W. J. (2017). Parent-implemented behavioral skills training of social skills. *Journal of Applied Behavior Analysis, 50*, 805–818.

Donaldson, J. M., & Vollmer, T. R. (2011). An evaluation and comparison of time-out procedures with and without release contingencies. *Journal of Applied Behavior Analysis, 44*, 693–705. https://doi.org/10.1901/jaba.2011.44-693

Drifke, M. A., Tiger, J. H., & Wierzba, B. C. (2017). Using behavior skills training to teach parents to implement three-step prompting: A component analysis and generalization assessment. *Learning and Motivation, 57*, 1–14.

Dunlap, G., Kern-Dunlap, L., Clarke, S., & Robbins, F. R. (1991). Functional assessment, curricular revision, and severe behavior problems. *Journal of Applied Behavior Analysis, 24*(2), 387–397. https://doi.org/10.1901/jaba.1991.24-387

Emerson, E., Kieman, 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

Fisher, W. W., Greer, B. D., Fuhrman, A. M., Saini, V., & Simmons, C. A. (2018). Minimizing resurgence of destructive behavior using behavioral momentum theory. *Journal of Applied Behavior Analysis, 51*, 831–853. https://doi.org/10.1002/jaba.499

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*, 543–560.

Fisher, W. W., Piazza, C., Cataldo, M., Harrell, R., Jefferson, G., & Conner, R. (1993). Functional communication training with and without extinction and punishment. *Journal of Applied Behavior Analysis, 26*, 23–36.

Fisher, W. W., Piazza, C. C., Bowman, L. G., Hagopian, L. P., & Langdon, N. A. (1994). Empirically derived consequences: A data-based method for prescribing treatments for destructive behavior. *Research in Developmental Disabilities, 15*, 133–149. https://doi.org/10.1016/0891-4222(94)90018-3

Fuhrman, A. M., Fisher, W. W., & Greer, B. D. (2016). A preliminary investigation on improving functional communication training by mitigating resurgence of destructive behavior. *Journal of Applied Behavior Analysis, 49*, 884–899.

Fuhrman, A. M., Greer, B. D., Zangrillo, A. N., & Fisher, W. W. (2018). Evaluating competing activities to enhance functional communication training during reinforcement schedule thinning. *Journal of Applied Behavior Analysis, 51*, 931–942.

Funk, M., & Drew, N. (2017). *Mental health policy and service development department of mental health and substance abuse*. World Health Organization. Retrieved from http://apps.who.int/bookorders

Geiger, K. B., Carr, J. E., & Leblanc, L. A. (2010). Function-based treatments for escape maintained problem behavior: A treatment selection model for practicing behavior analysts. *Behavior Analysis in Practice, 3*(1), 22–32.

Grant, J. E. (2005). Restraint and monitoring of psychotic or suicidal patients. *Current Psychiatry, 4*(11), 84–86.

Greer, B. D., Neidert, P. L., Dozier, C. L., Payne, S. W., Zonneveld, K. L. M., & Harper, A. M. (2013). Functional analysis and treatment of problem behavior in early education classrooms. *Journal of Applied Behavior Analysis, 46*, 289–295.

Hagopian, L. P., Contrucci Kuhn, S. A., Long, E. S., & Rush, K. S. (2005). Schedule thinning following communication training: Using competing stimuli to enhance tolerance to decrements in reinforcer density. *Journal of Applied Behavior Analysis, 38*, 177–193.

Hagopian, L. P., Fisher, W. W., & Legacy, S. M. (1994). Schedule effects of non-contingent reinforcement on attention-maintained destructive behavior in identical quadruplets. *Journal of Applied Behavior Analysis, 27*, 317–325.

Hagopian, L. P., Fisher, W. W., Thibault Sullivan, M., Acquisto, J., & LeBlanc, L. A. (1998). Effectiveness of functional communication training with and without extinction and punishment: A summary of 21 inpatient cases. *Journal of Applied Behavior Analysis, 31*, 211–235.

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*, 229–232. https://doi.org/10.1901/jaba.2001.34-229

Hanley, G. P., Piazza, C. C., Fisher, W. W., & Maglieri, K. A. (2005). On the effectiveness of and preference for punishment and extinction components of function-based interventions. *Journal of Applied Behavior Analysis, 38*, 51–65.

Herrnstein, R. J. (1961). Relative and absolute strength of response as a function of frequency of reinforcement. *Journal of the Experimental Analysis of Behavior, 4*, 563–573.

Hutchinson, R. R. (1977). By-products of aversive control. In W. K. Honig & J. E. R. Staddon (Eds.), *Handbook of operant behavior. Century psychology series* (pp. 415–430). Englewood Cliffs, NJ: Prentice Hall.

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*, 197–209.

Iwata, B. A., Pace, G. M., Cowdery, G. E., & Miltenberger, R. G. (1994). What makes extinction work: An analy-

sis of procedural form and function. *Journal of Applied Behavior Analysis, 27*, 131–144.

Jacobs, E. A., Borrero, J. C., & Vollmer, T. R. (2013). *Translational applications of quantitative choice models*. Washington, DC: American Psychological Association.

Jessel, J., & Borrero, J. C. (2014). A laboratory comparison of two variations of differential-reinforcement-of-low-rate procedures. *Journal of Applied Behavior Analysis, 47*, 314–324.

Jessel, J., & Ingvarsson, E. T. (2016). Recent advances in applied research on DRO procedures. *Journal of Applied Behavior Analysis, 49*, 991–995.

Johnston, J. M., & Sherman, R. A. (1993). Applying the least restrictive alternative principle to treatment decisions: A legal and behavioral analysis. *The Behavior Analyst, 16*(1), 103–115.

Knox, D., & Holloman, G. (2012). Use and avoidance of seclusion and restraint: Consensus statement of the american association for emergency psychiatry Project BETA seclusion and restraint workgroup. *Western Journal of Emergency Medicine, 13*(1), 35–40. https://doi.org/10.5811/westjem.2011.9.6867

Kunnavatana, S. S., Bloom, S. E., Samaha, A. L., Slocum, T. A., & Clay, C. C. (2018). Manipulating parameters of reinforcement to reduce problem behavior without extinction. *Journal of Applied Behavior Analysis, 51*, 283–302.

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*, 127–137.

Lambert, J. M., Bloom, S. E., Samaha, A. L., Dayton, E., & Kunnavatana, S. S. (2016). Effects of noncontingent reinforcement on the persistence and resurgence of mild aggression. *The Psychological Record, 66*, 283–289.

Lambert, J. M., Bloom, S. E., Samaha, A. L., Dayton, E., & Rodewald, A. M. (2015). Serial alternative response training as intervention for target response resurgence. *Journal of Applied Behavior Analysis, 48*, 765–780.

Landa, R., & Hanley, G. P. (2016). An evaluation of multiple-schedule variations to reduce high-rate requests in the picture exchange communication system. *Journal of Applied Behavior Analysis, 49*, 388–393.

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*, 345–385. 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 the treatment of self-injurious behavior. *Journal of Applied Behavior Analysis, 32*(1), 1–8. https://doi.org/10.1901/jaba.1999.32-1

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.

Mace, C. F., Pratt, J. L., Prager, K. L., & Pritchard, D. (2011). An evaluation of three methods of saying "no" to avoid an escalating response class hierarchy. *Journal of Applied Behavior Analysis, 44*, 83–94. https://doi.org/10.1901/jaba.2011.44-83

Marcus, B. A., Swanson, V., & Vollmer, T. R. (2001). Effects of parent training on parent and child behavior using procedures based on functional analyses. *Behavioral Interventions, 16*, 87–104.

Meehan, T., Bergen, H., & Fjeldsoe, K. (2004). Staff and patient perceptions of seclusion: Has anything changed? *Journal of Advanced Nursing, 47*(1), 33–38. https://doi.org/10.1111/j.1365-2648.2004.03062.x

Mohr, W. K., Petti, T. A., & Mohr, B. D. (2003). Adverse effects associated with physical restraint. *Canadian Journal of Psychiatry, 48*(5), 330–337. https://doi.org/10.1177/070674370304800509

Moore, D. W., Anderson, A., & Kumar, K. (2005). Instructional adaptation in the management of escape-maintained behavior in a classroom. *Journal of Positive Behavior Interventions, 7*(4), 216–223. https://doi.org/10.1177/10983007050070040301

Mueller, M. M., Piazza, C. C., Moore, J. W., & Kelley, M. E. (2003). Training parents to implement pediatric feeding protocols. *Journal of Applied Behavior Analysis, 36*, 545–562. https://doi.org/10.1901/jaba.2003.36-545

Piazza, C. C., Contrucci, S. A., Hanley, G. P., & Fisher, W. W. (1997). Nondirective prompting and noncontingent reinforcement in the treatment of destructive behavior during hygiene routines. *Journal of Applied Behavior Analysis, 30*, 705–708. https://doi.org/10.1901/jaba.1997.30-705

Piazza, C. C., Moes, D. R., & Fisher, W. W. (1996). Differential reinforcement of alternative behavior and demand fading in the treatment of escape-maintained destructive behavior. *Journal of Applied Behavior Analysis, 29*(4), 569–572. https://doi.org/10.1901/jaba.1996.29-569

Reynolds, D. (2006). Medical examiner: Restraint killed girl, 7. *Inclusion Daily Express.*

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*, 1–24.

Severtson, J. M., & Carr, J. E. (2012). Training novice instructors to implement errorless discrete-trial teaching: A sequential analysis. *Behavior Analysis in Practice, 5*, 13–23.

Shamlian, K., 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

Slocum, S. K., Grauerholz-Fisher, E., Peters, K. P., & Vollmer, T. R. (2017). A multicomponent approach to thinning reinforcer delivery during noncontingent reinforcement schedules. *Journal of Applied Behavior Analysis, 51*, 61–69.

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*(3), 563–574. https://doi.org/10.1002/jaba.216

St. Peter Pipkin, C., Vollmer, T. R., & Sloman, K. N. (2010). Effects of treatment integrity failures during differential reinforcement of alternative behavior: A translational model. *Journal of Applied Behavior Analysis, 43*, 47–70.

Thompson, R. H., Fisher, W. W., Piazza, C. C., & Kuhn, D. E. (1998). The evaluation and treatment of aggression maintained by attention and automatic reinforcement. *Journal of Applied Behavior Analysis, 31*, 102–116.

Troutman, B., Myers, K., Borchardt, C., Kowalski, R., & Bubrick, J. (1998). Case study: When restraints are the least restrictive alternative for managing aggression. *Journal of American Academy of Child & Adolescent Psychiatry, 37*(5), 554–558.

Ulrich, R. (1966). Pain as a cause of aggression. *Integrative and Comparative Biology, 6*(4), 643–662. https://doi.org/10.1093/icb/6.4.643

Ulrich, R. E., & Azrin, N. H. (1962). Reflexive fighting in response to aversive stimulation. *Journal of the Experimental Analysis of Behavior, 5*(4), 511–520.

Van Camp, C. M., Vollmer, T. R., Goh, H., Whitehouse, C. M., Reyes, J., Montgomery, J. L., & Borrero, J. C. (2008). Behavior parent training in child welfare: Evaluations of skills acquisition. *Research on Social Work Practice, 18*, 377–391.

Volkert, V. M., Lerman, D. C., Call, N. A., & Trosclair-Lasserre, N. (2009). An evaluation of resurgence during treatment with functional communication training. *Journal of Applied Behavior Analysis, 42*, 145–160.

Vollmer, T. R., Borrero, J. C., Lalli, J. S., & Daniel, D. (1999). Evaluating self-control and impulsivity in children with severe behavior disorders. *Journal of Applied Behavior Analysis, 32*, 451–466.

Vollmer, T. R., Hagopian, L. P., Bailey, J. S., Dorsey, M. F., Hanley, G., Lennox, D., … Spreat, S. (2011). The association for behavior analysis international position statement on restraint and seclusion. *Behavior Analyst, 34*(1), 103–110. https://doi.org/10.1007/BF03392238

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.

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*, 9–21.

Vollmer, T. R., Roane, H. S., Ringdahl, J. E., & Marcus, B. A. (1999). Evaluating treatment challenges with differential reinforcement of alternative behavior. *Journal of Applied Behavior Analysis, 32*, 9–23. https://doi.org/10.1901/jaba.1999.32-9

Vollmer, T. R., & Sloman, K. N. (2005). The historical context of noncontingent reinforcement as a behavioral treatment. *European Journal of Behavior Analysis, 6*, 9–19. https://doi.org/10.1080/15021149.2005.11434242

Vollmer, T. R., Sloman, K. N., & St. Peter Pipkin, C. (2008). Practical implications of data reliability and treatment integrity monitoring. *Behavior Analysis in Practice, 1*, 4–11. https://doi.org/10.1007/BF03391722

Worsdell, A. S., Iwata, B. A., Hanley, G. P., Thompson, R. H., & Kahng, S. W. (2000). Effect of continuous and intermittent reinforcement for problem behavior during functional communication training. *Journal of Applied Behavior Analysis, 33*, 167–179. https://doi.org/10.1901/jaba.2000.33-167

Zangrillo, A. N., Fisher, W. W., Greer, B. D., Owen, T. M., & DeSouza, 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, 63*, 147–156. https://doi.org/10.1080/20473869.2016.1176308

Zuluaga, C. A., & Normand, M. P. (2008). An evaluation of the high-probability instruction sequence with and without programmed reinforcement for compliance with high-probability instructions. *Journal of Applied Behavior Analysis, 41*(3), 453–457. https://doi.org/10.1901/jaba.2008.41-453

34 Self-Injurious Behavior, Rituals, and Stereotypies in Dual Diagnosis

Jessica Akers, Tonya Davis, and Stephanie Gerow

Self-Injurious Behavior, Rituals, and Stereotypies in Dual Diagnosis

This chapter will cover a range of treatment options which target the reduction of self-injury, rituals, and stereotypy, including interventions based on the principles of applied behavior analysis, protective equipment, and medication. These treatment options may be implemented in isolation but are more commonly recommended to serve as components of a treatment package (e.g., protective equipment in combination with a consequence-based intervention). Although discussed within the same chapter, distinct treatment considerations for self-injury and stereotypy will be highlighted throughout. The most significant difference between self-injurious behavior and stereotypy is the acceptable levels of behavior reduction. Due to safety concerns, practitioners should target an 80–100% reduction in self-injurious behavior. In contrast, some level of stereotypy can be accepted as long as it is not overly disruptive. Individuals without intellectual disabilities typically engage in stereotypic behaviors (e.g., leg shaking, finger tapping); thus, complete elimination of stereotypy for individuals with intellectual disabilities seems unwarranted and possibly unethical.

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Applied Behavior Analysis

A substantial research base supports the use of behavioral interventions to reduce challenging behavior, such as self-injurious behavior, rituals, and stereotypy. These interventions, based on the principles of applied behavior analysis, consist of systematically altering the environment to reduce problematic behavior and increase appropriate behavior (Cooper, Heron, & Heward, 2007).

Reinforcement-Based Interventions

Reinforcement occurs when a consequence is delivered contingent upon a behavior and results in the frequency of the behavior increasing in the future. That is, reinforcement is defined by the future persistence of the behavior (Skinner, 1953). Reinforcement can be further defined by the consequence or type of stimulus change. Specifically, positive reinforcement involves the presentation of a stimulus contingent upon the occurrence of the target behavior which results in persistence of behavior or an increase in the future frequency of that behavior. For example, positive reinforcement would have occurred if after a child made his bed his dad let him play

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with his favorite toys and the child continued to make his bed in the future. On the other hand, negative reinforcement involves the removal or termination of an already-present stimulus contingent upon the target behavior which results in persistence of behavior or an increase in the future frequency of that behavior. Following the previous example of bed making, negative reinforcement would have occurred if the dad was nagging the child to make his bed and, contingent upon the child making the bed, the nagging stopped and the child continued to make his bed in the future.

Although reinforcement is a procedure that results in persistence or increased future frequency of a target behavior, many treatment procedures aimed to reduce the future frequency of a target behavior actually involve reinforcement procedures. In such cases, typically treatment procedures involve the reinforcement of a desirable behavior that, if increased, would reduce the future frequency of the undesirable behavior. For example, if aiming to reduce hand flapping, a reinforcement procedure could be implemented to increase the frequency of the individual keeping his hands in his pocket because doing so would also reduce the frequency of hand flapping. Prior to introducing a reinforcement-based intervention, the function of the behavior must be determined. The function of behavior refers to the environmental consequences that have been found to maintain the behavior. Identifying the function of the behavior can lead to more effective and individualized intervention procedures (Hanley, Iwata, & McCord, 2003; Loman & Horner, 2014). The function of behavior is determined using a functional behavior assessment, which often includes interviews, observations, and an experimental functional analysis (Arndorfer & Miltenberger, 1993; see Chap. 24 for detailed description of assessment procedures). Functions of behavior are typically classified into two broad categories: socially maintained behavior and automatically maintained behavior. Typical social functions of challenging behavior include access to attention, access to preferred items or activities, and escape from non-preferred tasks (e.g., academic demands; Hanley et al., 2003). Automatically maintained behavior is not influenced by consequences in the environment; rather, these behaviors are hypothesized to produce an internal form of reinforcement (Cooper et al., 2007; Hanley et al., 2003; Iwata, Dorsey, Slifer, Bauman, & Richman, 1994). Stereotypy is generally maintained by automatic reinforcement (Rapp & Vollmer, 2005), whereas self-injurious behavior has been shown to be maintained by automatic reinforcement in only approximately 20% of cases (Beavers, Iwata, & Lerman, 2013).

Differential Reinforcement of Alternative Behavior The most commonly implemented reinforcement-based procedure for reducing socially maintained challenging behavior is differential reinforcement of alternative behavior (DRA; Tiger, Hanley, & Bruzek, 2008; Wong et al., 2014). DRA consists of providing reinforcement contingent upon an appropriate, alternative behavior and withholding reinforcement contingent upon target challenging behavior (Cooper et al., 2007; Tiger et al., 2008). The alternative behavior can be communicative in nature or another form of alternative, appropriate behavior, such as task completion or appropriate toy play.

Following the selection of the response, the practitioner teaches the individual to engage in the appropriate, alternative response using prompting and repeated, structured practice (Tiger et al., 2008). The practitioner should contrive the evocative situation (i.e., the situation which is associated with challenging behavior) to provide multiple practice opportunities for the individual to engage in the alternative response. The practitioner should use effective prompting procedures, which will vary depending on the individual's skill level, to ensure the individual is successful in engaging in the response. The prompting procedures should be faded (i.e., systematically withdrawn) to promote independence.

Self-injurious behavior poses special concerns for implementing DRA procedures which must be considered. The most effective form of DRA includes extinction (i.e., withholding

reinforcement for the challenging behavior); however, due to safety concerns, implementing extinction with integrity may be ill-advised. Thus, prior to implementing DRA, the practitioner must determine whether the severity of the self-injury suggests the need for blocking procedures (e.g., an adult blocks the individual's hand from contacting her head), protective equipment, or a modification to the procedure such that extinction is not in place. A further discussion of both response blocking and protective equipment will be provided later in the chapter. DRA without extinction, although not ideal, in some cases may be necessary for safety purposes. Researchers have demonstrated that challenging behavior will decrease when different response and reinforcement dimensions are manipulated (see Petscher, Rey, & Bailey, 2009 for a review of DRA). For example, after adjusting the rate of reinforcement for self-injurious behavior, researchers observed a decrease in self-injurious behavior and an increase in the communication response (Worsdell, Iwata, Hanley, Thompson, & Kahng, 2000). This adjustment consisted of gradually increasing the response requirement for the self-injury while keeping the response requirement for the alternative response constant. Initially, one instance of the alternative response resulted in reinforcement, and one instance of self-injury resulted in reinforcement; however, eventually three or four instances of self-injury resulted in reinforcement. The participants began to allocate responding to the alternative response, and their engagement in self-injury substantially decreased.

Functional Communication Training Functional communication training (FCT) is a specific form of DRA in which the alternative behavior consists of a communicative response (Carr & Durand, 1985; Tiger et al., 2008). FCT has been shown to be the most frequently implemented treatment for self-injurious behavior (Kurtz et al., 2003). Teaching a communicative response to replace challenging behavior can be beneficial because the individual can access the reinforcement across settings and situations using the communicative response (Carr & Durand, 1985; Wacker et al., 1990). For example, the individual could learn to say, "break please" to request a break from work. The individual could use this same communicative response across settings and practitioners, and it is likely to result in reinforcement. This can lead to generalized improvements in self-injurious behavior (Durand & Carr, 1991; Falcomata & Wacker, 2013).

Selecting the most appropriate communicative response which will be taught to replace the self-injurious behavior is an important step in implementing FCT. The practitioner must select the mode of communication and the specific response that will be taught. The mode of communication will differ depending on the individual's current communicative repertoire and may consist of vocal responses, speech-generating device, card exchanges, or sign language. It is important to select a mode of communication that is easy for the individual to use or learn, requires a low amount of effort to emit, and is likely to be understood and reinforced by novel communication partners (Horner & Day, 1991; Tiger et al., 2008). Practitioners should consider initially introducing a nonvocal communicative response (e.g., card exchange) because the individual's exposure to evocative situation (e.g., removal of tangible items) can be controlled. For example, immediately following the removal of the tangible item, the practitioner can physically guide the card exchange to ensure the client is only without the tangible for 1–2 s. The exposure cannot be controlled in the same manner with a vocal response because the practitioner cannot physically guide vocal responses. This is of extreme importance as researchers have demonstrated that an exposure as short as 5 s to the evocative situation can lead to levels of self-injury which are higher than baseline levels (Fisher et al., 2018). Once socially significant reductions in self-injury are observed, practitioners can transition to accepting a vocal communicative response.

Schedule-thinning procedures can increase the feasibility of the intervention and promote maintenance and generalization of treatment

gains (Hagopian, Boelter, & Jarmolowicz, 2011). Initially, every instance of engagement in the communication response should result in the function-based reinforcer (i.e., continuous reinforcement schedule). The practitioner should avoid thinning the schedule of reinforcement before the individual makes adequate treatment gains. The length of time required to achieve treatment gains will vary by individual. For example, older individuals who have had a longer history of engaging in self-injurious behavior may require more exposure to the intervention before achieving significant treatment gains. Common FCT schedule-thinning procedures include chained schedules, multiple schedules, delay to reinforcement, and response restriction (Greer, Fisher, Saini, Owen, & Jones, 2016). Schedule-thinning procedures gradually increase the amount of time in which the functional reinforcer is unavailable based on low levels of self-injurious behavior and independent communicative responses. Chained schedules involve systematically increasing the number of compliance responses required prior to communicative responses resulting in access to the functional reinforcer (e.g., Berg, Wacker, Harding, Ganzer, & Barretto, 2007; Lalli, Casey, & Kates, 1995; Mildon, Moore, & Dixon, 2004). The response requirement promotes compliance which makes this procedure especially well suited for treating escape-maintained behavior. Multiple schedules are time-based schedules which do not include a response requirement. These schedules are well suited for behavior maintained by positive reinforcement (e.g., attention; Greer et al., 2016).

DRA for Automatically Maintained Behavior Although not as thoroughly researched as DRA for socially maintained behavior, there is research suggesting the effectiveness of DRA to address automatically maintained behavior. Researchers have demonstrated that introducing a structured work system was effective in reducing stereotypy for three children (Bennett, Reichow, & Wolery, 2011). The primary difference between implementing DRA for behavior which is socially maintained and automatically maintained is the identification of sources of reinforcement. The practitioner should use known reinforcing consequences and/or conduct a preference assessment to identify preferred stimuli (e.g., DeLeon & Iwata, 1996; Fisher et al., 1992) to increase the likelihood of effectiveness. Following the selection of the response and reinforcer, the practitioner must select the schedule of reinforcement for the alternative response. The schedule of reinforcement refers to the specific rules detailing when the alternative behavior will be reinforced. The schedule of reinforcement may be based on a specific number of responses that must be emitted in order for the participant to access reinforcement. For example, a child who engages in head hitting may be required to complete five math problems in order to access reinforcement. Alternatively, the schedule may be based on a duration of time. For instance, the same child who engages in head hitting may instead be required to work on a math worksheet for 3 minutes in order to access reinforcement. The schedule of reinforcement should be gradually faded contingent upon low levels of self-injury or stereotypy and independent engagement in the alternative behavior.

Differential Reinforcement of Incompatible Behavior Differential reinforcement of incompatible behavior (DRI) is procedure similar to DRA; however, the alternative behavior must be incompatible with the target behavior (Brawley, Harris, Allen, Flemming, & Peterson, 1969; Cooper et al., 2007; Jones & Baker, 1990; Lovaas, Freitag, Gold, & Kassorlia, 1965). Although DRI can be implemented to reduce any challenging behavior, it is often used to reduce self-injurious behavior and stereotypy (Jones & Baker, 1990).

In order to implement DRI, the practitioner must first select the incompatible behavior. Several factors should be considered in this selection process. First, the incompatible behavior should be one that is physically incompatible with the target behavior. For example, an individual who hits himself may be required to manipulate toys in order to access potential

reinforcement. Second, the incompatible behavior should be one already in the individual's repertoire. The individual must be able to independently emit the selected incompatible behavior. Third, practitioners should also select incompatible behaviors that are socially acceptable as well as compatible with adaptive behaviors. For instance, placing hands in pockets is a socially acceptable and incompatible behavior for skin picking, but it also limits the individual's ability to complete a number of academic, vocational, or leisure tasks; therefore, it may not be ideal to promote and reinforce hands in pockets for long periods of time.

In addition to selecting the incompatible behavior, the practitioner must also identify the specific stimuli to provide as a potential reinforcement as well as reinforcement schedule. When determining potential reinforcers, the practitioner should first consider reinforcers associated with the function of the target self-injurious behavior or stereotypy. In other words, if the target challenging behavior is maintained by access to tangibles, it is wise to provide access to those tangibles contingent upon the incompatible behavior. As previously mentioned, self-injurious behavior and stereotypy may be maintained by automatic reinforcement. In those cases, practitioners should use a preference assessment to identify preferred stimuli to present contingent upon the incompatible behavior (e.g., DeLeon & Iwata, 1996; Fisher et al., 1992). Finally, in order to implement DRI, the practitioner must select the schedule of reinforcement for the incompatible behavior. The schedule can be response-based or time-based depending on the selected incompatible behavior.

The current literature provides several examples of the use of DRI to successfully reduce self-injurious behavior, rituals, and stereotypy. Luiselli, Colozzi, Helfen, and Pollow (1980) implemented DRI with a participant with an intellectual disability who engaged in vocal stereotypy. The participant was given small snacks if he remained quiet (incompatible behavior) for 1 minute. The 1-minute interval was gradually increased contingent upon the participant's success. The DRI successfully resulted in near-zero levels of vocal stereotypy. Jones and Baker (1988) also implemented DRI with a participant who engaged in self-injury. Prior to the implementation of DRI, the boy wore arm splints and a helmet at all times due to the frequency and intensity of the self-injurious behavior. DRI involved providing reinforcement contingent upon task completion, which was incompatible with self-injury. The success of the DRI intervention allowed the participant to have periods of time in which the restraint and safety devices could be safely removed.

Differential Reinforcement of Other Behavior Differential reinforcement of other behavior (DRO) is a procedure in which potential reinforcement is provided for the absence of the target challenging behavior (Cooper et al., 2007; Reynolds, 1961; Wong et al., 2014). In other words, DRO involves the delivery of reinforcement for the lack of responding. As a result, DRO is sometimes referred to as *differential reinforcement of zero responding*.

To implement DRO, the practitioner determines the interval of time in which the individual must refrain from engaging in the self-injury or stereotypy in order to access the potential reinforcer. This interval of time should be short enough that it is likely the individual will be able to meet the contingency and contact reinforcement in the first attempts; therefore, it is prudent to use baseline data to determine how long the individual currently refrains from engaging in the target behavior to determine an interval that would likely result in success. This initial interval may not result in socially significant decreases in the target behavior due to the short duration, but the interval can be systematically increased over time.

The current literature provides many examples of the implementation of DRO to reduce self-injurious behavior, rituals, and stereotypies. For example, Lustig et al. (2014) implemented DRO with a participant who engaged in near continuous, socially inappropriate stereotypy involving leg movements. Initially, DRO consisted of providing the participant with access to a

preferred toy contingent upon the absence of the leg movements for 10 seconds. As the participant demonstrated success in refraining from engaging in the leg movements for that specified amount of time, the interval duration was slowly increased. The stereotypy was reduced from near continuous levels to zero levels using the DRO procedure.

Procedural Variations When implementing DRO, practitioners have many choices regarding variations in the DRO procedure (Weston, Hodges, & Davis, 2018). These include the consequence provided as potential reinforcement, schedule arrangements, and the use of rules and signals. These procedural variations are described below. It is important to keep in mind that procedures should be customized to the needs of the individual.

Reinforcement Selection The first variation to DRO procedures involves the selection of reinforcement. Ideally, the selected consequence is one that will result in favorable behavior change. In order to increase the likelihood of the effectiveness of the consequence, practitioners should consider using the same variables maintaining the target self-injurious behavior or stereotypy when possible. That is, if a functional analysis concluded the target behavior was maintained by socially mediated variables such as accessing attention or escaping demands, then these variables should be used as reinforcement for DRO. For example, Hammond, Iwata, Fritz, and Dempsey (2011) implemented a momentary DRO to reduce head hitting for a participant with an intellectual disability. A functional analysis concluded that head hitting was maintained by access to tangibles. Those tangibles were then delivered as a consequence if the participant was not engaging in self-injurious behavior, which successfully resulted in a decrease in head hitting.

It is important to note that in many cases, self-injurious behavior and stereotypy may be maintained by automatic reinforcement, thus making it impossible for the practitioner to control access to the maintaining variable(s) to use as reinforcement within the DRO. When using DRO to reduce automatically maintained self-injurious behavior or stereotypy, the practitioner should use known reinforcing consequences and/or conduct a preference assessment to identify preferred stimuli (e.g., DeLeon & Iwata, 1996; Fisher et al., 1992) to increase the likelihood of effectiveness.

Schedule Arrangement The second variation to DRO procedures involves the schedule arrangement. The more frequently implemented schedule is the interval DRO which involves the practitioner delivering reinforcement contingent upon the absence of the target behavior for an entire period of time. The previous example of the DRO procedure to reduce leg movements is an example of an interval DRO. If using an interval DRO procedure, the practitioner may choose to reset the interval if the target behavior occurs prior to the end of the interval. To do so, when the target behavior occurs within the interval, that interval is immediately ended and a new interval is initiated. On the other hand, the practitioner may choose not to reset the interval; in which case, if the target behavior occurs during the interval, the interval continues regardless of the fact that reinforcement can no longer be obtained at the end of that interval. Both approaches are supported in the current literature (Weston et al., 2018).

A *momentary* DRO involves the delivery of reinforcement if the target behavior is absent for a specific moment in time. One benefit of the momentary DRO methodology is that, unlike the interval DRO procedure, the practitioner does not need to observe the individual at all times but instead only at the moment when reinforcement is available. While interval DRO procedures are more prevalent in the research (Weston et al., 2018), there is some evidence to support the effectiveness of momentary DRO as well. For example, Toussaint and Tiger (2012) used a momentary DRO with a participant with multiple disabilities to reduce skin picking. During

10-minute sessions, the participant was alone in a room. A therapist entered the room at randomly scheduled times. If the participant was abstaining from skin picking at that moment, he earned a token to be exchanged for access to watching a video. However, he did not receive a token if he was picking his skin the moment the therapist entered the room. The momentary DRO successfully reduced skin picking to near-zero levels.

Rules and Signals The final variation to DRO procedures involves incorporating a rule or signal to communicate the DRO contingencies. A rule may involve simply explaining to the individual how he or she can earn access to the preferred consequences prior to implementing DRO. A signal, on the other hand, may occur just before the opportunity to earn reinforcement. For example, Hammond and colleagues held the reinforcer in front of the participant just seconds before the potential reinforcement delivery within a momentary DRO to signal the upcoming availability of reinforcement.

Noncontingent reinforcement Unlike the differential reinforcement procedures described, noncontingent reinforcement refers to the reinforcer remaining continuously available regardless of engagement in self-injurious behavior or stereotypy. The stimuli introduced to serve as the reinforcer include preferred stimuli (Higbee, Chang, & Endicott, 2005), matched stimuli (i.e., stimuli which match the sensory stimulation hypothesized to maintain the behavior, e.g., Rapp, Cook, McHugh, & Mann, 2017), or competing stimuli (e.g., Groskreutz, Groskreutz, & Higbee, 2011). Research suggests that stimuli identified using a competing stimulus assessment are more effective in decreasing self-injury and stereotypy as compared to stimuli identified via a preference assessment (Higbee et al., 2005; Rooker, Bonner, Dillon, & Zarcone, 2018). In a review of the literature, researchers determined that noncontingent access to a competing stimulus is the most commonly implemented and most effective intervention implemented in isolation for treating automatically maintained self-injurious behavior (Rooker et al., 2018). For example, researchers treated automatically maintained ear digging for one participant using noncontingent access to matched stimulation in the form of soft acrylic balls which the participant independently placed in his ears (Davis, Dacus, Strickland, Machalicek, & Coviello, 2013). Following the introduction of the matched stimulation, ear digging decreased to zero levels for the majority of sessions. Although effective as an independent intervention, noncontingent reinforcement often serves as component of a treatment package with other interventions such as response cost (Watkins, Paananen, Rudrud, & Rapp, 2011; Watkins & Rapp, 2014), contingent exercise (Kahng, Abt, & Wilder, 2001), and response interruption and redirection (RIRD; Love, Miguel, Fernand, & LaBrie, 2012). Competing stimuli are identified via a competing stimulus assessment, which consists of recording both the percentage of the session in which the individual engages in stereotypy or self-injury and the percentage of the session in which the individual interacts with the target stimulus. Stimuli associated with high levels of engagement and low levels of stereotypy or self-injury are determined to serve as a competing stimulus. Practitioners selecting stimuli to include in the competing stimulus assessment should consider stimuli that are available across settings, stimuli that are less disruptive than the stereotypy, and stimuli that will likely continue to compete with the behavior for an extended period of time.

Punishment-Based Interventions

Punishment occurs when a consequence delivered contingent upon a behavior results in frequency of the behavior decreasing in the future (Azrin & Holz, 1966). Like reinforcement, punishment can be divided into two categories, based on the type of stimulus change. Positive punishment is the presentation of a stimulus change following a behavior that decreases the future frequency of that behavior. On the other hand, negative punishment is the removal of a stimulus following a behavior that decreases the future

frequency of that behavior (Mayer, Sulzer-Azaroff, & Wallace, 2012). For example, a reprimand delivered contingent upon hand flapping that decreases the future frequency of hand flapping is positive punishment. In contrast, removing the individual's toys contingent upon hand flapping that decreases the future frequency of hand flapping is negative punishment. Both of these examples represent punishment because the future frequency of hand flapping decreased.

Punishment procedures may be viewed as controversial. While most can agree that all practitioners working with others should do no harm, provide effective treatment, and use least intrusive approaches, the decision to use punishment procedures is rarely simple (Cooper et al., 2007). Undesirable side effects of punishment are well documented in the literature. These include emotional and aggressive reactions as well as escape or avoidance behavior (Azrin & Holz, 1966). Escape and avoidance may be particularly detrimental if the individual avoids people, settings, or activities that could have positive impacts on the individual's life (e.g., parent, teacher, school, vocational tasks). However, practitioners may be able to minimize or prevent these side effects by implementing interventions that also incorporate reinforcement procedures.

Despite the aforementioned side effects associated with punishment, treatment procedures should be selected based on a variety of factors including the risks associated with the target behavior, the variables controlling the behavior, potential treatment effectiveness, and potential treatment risks (DiGennaro Reed & Lovett, 2008; Foxx, 2005; Van Houten et al., 1988). It is possible that when considering these factors, punishment is a viable treatment procedure. For example, punishment may be a viable treatment or treatment component in cases in which the behavior poses a high risk of injury to the individual and/or the variables maintaining the behavior are unknown. Practitioners have many factors to consider when selecting a treatment, especially if this treatment involves punishment. Moreover, practitioners should be mindful of professional ethical guidelines in the treatment selection process. Thoughtful consideration of each of these factors will allow practitioners to select effective treatments that minimize risk to the individual.

The current literature provides some examples of the successful implementation of punishment procedures. In these examples, oftentimes the punishment procedure is a component of a treatment package that also includes reinforcement procedures. Such punishment procedures include response blocking, hands down, response blocking and redirection (RIRD), and verbal reprimand.

Response Blocking Response blocking involves the practitioner physically blocking the target self-injurious behavior, ritual, or stereotypic response. There are several examples of successful response blocking procedures in the literature. Smith, Russo, and Le (1999) implemented response blocking with a participant with developmental disabilities who engaged in automatically maintained eye poking. Prior to the study, the chronic eye poking had resulted in blindness in both eyes. To implement response blocking, the practitioner placed his hand between the participant's finger and her eye any time she attempted to eye poke. Response blocking resulted in marked decreases in eye poking.

One variation of the response blocking procedures is a hands-down procedure. The hands-down procedure involves physically blocking, but unlike response blocking which is typically terminated when the target behavior is prevented or discontinued, the hands-down procedure involves then redirecting the individual to place his or her hands down on a table or by his or her side for a specific amount of time, often 1–10 seconds. The practitioner should use only enough force to prevent the target challenging behavior rather than completely immobilizing the individual's hands.

Practitioners selecting to implement response blocking should consider the following factors. First, the practitioner must be physically capable of blocking the response without posing risk of injury to himself or herself or the individual. In other words, it may not be physically possible for

a practitioner to block hand biting of an individual who is taller than the practitioner. Similarly, blocking hand-to-head self-injury could pose risk of harm to the practitioner who could be hit during an attempted response block. Second, response blocking requires the practitioner to be in close proximity to the individual for long periods of time, which may not be feasible in some cases. Third, some research indicates that response blocking may increase target challenging behavior during nonintervention periods. Rapp (2006) implemented response blocking that effectively reduced stereotypy when implemented. However, during the 5 minutes after the response blocking sessions in which response blocking was not implemented, the target stereotypy increased above levels observed prior to intervention. Additional research is needed to fully explore these potential drawbacks of response blocking.

Response Interruption and Redirection RIRD consists of interrupting an individual engaging in a challenging behavior and then redirecting him or her to emit several adaptive behaviors (Ahearn, Clark, & MacDonald, 2007). The procedure is a modification of the traditional response blocking procedure but was specifically designed to reduce vocal stereotypy, a topography of behavior that cannot be physically blocked. Although originally designed to treat vocal stereotypy, the procedure has been expanded to reduce motor stereotypy as well (e.g., Ahrens, Lerman, Kodak, Worsdell, & Keegan, 2011).

Ahearn et al. (2007) implemented RIRD in a school setting with four participants with autism spectrum disorder who engaged in vocal stereotypy. When a participant engaged in vocal stereotypy, the teacher interrupted the stereotypic responses by calling the participant's name and initiating eye contact. The teacher then prompted the participant to either respond to questions (e.g., "What is your name?") or imitate vocal responses (e.g., "say ball") until the participant complied with three consecutive directions without engaging in vocal stereotypy. At this time, the teacher praised the participant for use of appropriate language. Vocal stereotypy was reduced among all four participants, and increase in appropriate communication was observed for three of the four participants.

To implement RIRD, the practitioner would need to determine how to interrupt the target behavior. This interruption would need to be effective as well as discrete to reduce the possibility of negative stigmatization. Second, the practitioner would need to determine the adaptive behavior(s) to which the individual would be redirected to emit. These should be socially acceptable behaviors currently within the individual's repertoire. Typically, the redirected behaviors match the topography of the target behavior. That is, if the individual engages in vocal stereotypy, the practitioner directs the individual to emit acceptable vocal responses such as answering questions. In contrast, if the individual engages in motor stereotypy, the practitioner directs the individual to emit an acceptable motor response such as fine motor imitation. However, recent research indicates that the similarity between topography of behavior and the redirected responses may not be necessary (e.g., Ahrens et al., 2011; Cassella, Sidener, Sidener, & Progar, 2011). Finally, the practitioner would need to determine the number of socially acceptable responses the individual will be expected to complete. Typically, immediately after interrupting the target behavior, the practitioner prompts the individual to emit three consecutive correct responses; however, recent research indicates that RIRD may be successful with fewer repetitions (e.g., Saini, Gregory, Uran, & Fantetti, 2015).

Verbal Reprimand In some cases, a simple verbal reprimand contingent upon self-injurious behavior or stereotypy may be effective. Reprimands should involve a short statement that interrupts the target behavior and communicates that the behavior should be terminated. Examples of verbal reprimands include, "keep your hands down" and "stop making sounds." Ideally, a verbal reprimand is as simple and discrete as possible while maintaining effectiveness.

Although the simplicity of verbal reprimands lends to its appeal, research supporting its effectiveness is limited. Richman, Lindauer, Crossland, McKerchar, and Morse (2001) implemented verbal reprimands to reduce breath holding with a participant with intellectual disability and cerebral palsy. While verbal reprimand was successful in reducing breath holding, several variables were evaluated, specifically the volume of the reprimand and the proximity of the individual delivering the reprimand. Loud reprimands within a close proximity were found to be effective, but reprimands delivered with a soft volume or at a distance from the individual were not as effective. Moreover, results indicate that maximum effectiveness was reached only when a DRO was added to verbal reprimands delivered loudly and in close proximity. Rapp, Patel, Ghezzi, O'Flaherty, and Titterington (2009) also found mixed results regarding the effectiveness of verbal reprimands. While verbal reprimands successfully reduced vocal stereotypy for two of three participants, additional procedures were required to reduce vocal stereotypy for the other participant. Due to limited evidence of their effectiveness implemented in isolation, it may be best for verbal reprimands to be used as a component of a treatment package (e.g., Rapp et al., 2017; Richman et al., 2001). As such, if a practitioner implements verbal reprimands as an isolated treatment, it would be necessary to monitor effectiveness closely.

Protective Equipment

Protective equipment can be used to decrease the frequency of self-injurious behavior as well as limit the potential for serious bodily harm (Dorsey, Iwata, Reid, & Davis, 1982; Moore, Fisher, & Pennington, 2004). Examples of protective equipment include helmets, shoulder pads, padded arm sleeves, padded leg sleeves, padded slippers, and boxing gloves.

Protective equipment can be used to physically prevent engagement in the specific self-injurious behavior. Using protective equipment in this manner is described as a mechanical restraint. For example, arm splints make it physically impossible for an individual to poke her eye. This procedure should only be used when the intensity of self-injury is at level in which permanent damage may occur (e.g., blindness). If this type of protective equipment is necessary, practitioners should employ procedures to ensure the individual is exposed to the least restrictive procedures. A rapid restraint analysis can be used to determine the level of restraint that is required to prevent self-injury (Wallace, Iwata, Zhou, & Goff, 1999). Researchers have continued to modify the rapid restraint assessment to expedite the fading procedure and better evaluate the extent to which other adaptive behaviors are inhibited (DeRosa, Roane, Wilson, Novak, & Silkowski, 2015; Deshais, Fisher, Hausman, & Kahng, 2015).

Protective equipment can also be used to block the sensory stimulation which is hypothesized to maintain the self-injury. For example, while wearing a padded arm sleeve, an individual can still bite his arms but will not obtain the same stimulation as when he bites in the absence of the sleeve. This procedure is called sensory extinction. Moore and colleagues demonstrated that self-injurious behavior decreased when padded equipment was in place. The participant engaged in multiple topographies of self-injury which the researchers separated into three categories: self-injury with shoulder, self-injury with hand, and self-injury with leg. The researchers systematically added and removed shoulder pads, padded arm sleeves, and padded leg sleeves. They observed that when the protective equipment was removed from one body part, self-injury related to that body part increased, while self-injury related to the other body parts did not increase. For example, if the padded arm and leg sleeves were in place but shoulder pads were removed, only self-injury with the shoulder increased.

Finally, protective equipment can be used as a means to safely implement extinction procedures for socially maintained self-injurious behavior. If self-injurious behavior is maintained by access to attention from adults, the procedures for DRA suggest to ignore the self-injurious behavior and provide attention contingent upon the alternative

behavior. However, depending on the intensity of the self-injury, this may be unsafe. For example, if an individual bites his arm in order to gain attention, it may be unsafe to ignore this self-biting. The introduction of padded arm sleeves could reduce the risk of harm to the individual and facilitate teaching the request "play with me." It is important to note that even with protective equipment in place, practitioners should remain vigilant in observing their clients as the individual may identify new topographies of self-injury which are not inhibited by the protective equipment.

Using protective equipment alone is not an effective treatment strategy because as soon as the protective equipment is removed, the individual will presumably resume engagement in self-injury. In addition, when the protective equipment is in place, the individual may be unable to engage in other prosocial behavior (e.g., self-feeding). Therefore, protective equipment should always serve as a component of a treatment package with the goal of fading the equipment if possible.

Medication

Due to the dangerous nature of self-injury and disruptive nature of stereotypy, interventions which lead to rapid behavior reductions are ideal. Thus, intensive interventions which include medication as a component may be recommended. In a recent international survey, researchers found that, within their population, 90% of the individuals with intellectual disabilities were currently using psychotropic medication (Perry et al., 2018). Lower estimates have been recorded at 38% (Bowring, Totsika, Hastings, Toogood, & McMahon, 2017) and 49% (Sheehan et al., 2015). Prescription of antipsychotic medication is not restricted to adults with intellectual disabilities. Researchers found a higher percentage of children with intellectual disabilities (2.8%) are prescribed antipsychotics as compared to children without intellectual disabilities (0.15%; Brophy et al., 2018). The majority of individuals with intellectual disabilities who are prescribed psychotropic medications do not have a documented psychiatric disorder (Lunsky et al., 2018; Perry et al., 2018; Sheehan et al., 2015). Therefore, many of these prescriptions are considered off-label because they are being used in a manner which is different from that which they were intended. Although off-label prescribing is a common practice, off-label prescriptions do not receive the same level of scientific evaluation and thus do not provide the same level of confidence in the safety and effectiveness of the drug (Radley, Finkelstein, & Stafford, 2006). According to a nationwide survey of physician prescribing practices (not restricted to individuals with disabilities), antipsychotics and antidepressants are frequently prescribed for off-label use with little to no support from the research (Radley et al., 2006).

Overall, there is a consensus within the literature that the volume of evidence regarding the effectiveness of psychotropic medications for individuals with intellectual disabilities is limited (Ji & Findling, 2016; Matson & Neal, 2009; Tyrer et al., 2008). A review of studies evaluating the evidence of psychotropic drug use with individuals with intellectual disabilities reported that few studies employed double-blind, placebo-controlled procedures. The results of the studies that did employ such procedures were mixed with some reporting significant differences between the control and treatment groups and others reporting no significant differences. For example, one study which included 86 participants, and used a placebo-controlled group and randomization, found no differences in behavior change for the participants in the treatment groups (i.e., risperidone group, haloperidol group) and the control group (Tyrer et al., 2008). In contrast, other studies report that there is evidence that risperidone may be effective in decreasing challenging behavior in individuals with intellectual disabilities (e.g., Hellings et al., 2006). Drug classes which have received some experimental evaluation for their effects on self-injury include typical antipsychotics (e.g., chlorpromazine), atypical antipsychotics (e.g., risperidone), antidepressants (e.g., fluoxetine), antianxiety medications (e.g., buspirone), and

mood stabilizers (e.g., lithium; see Matson & Neal, 2009 for a full review).

Important Considerations In addition to the limited research evidence, there are other factors which should be considered prior to the use of psychotropic medication with individuals with disabilities. First, psychotropic medications may result in decreases in self-injury or stereotypy without treating the underlying cause of the behavior. For example, a side effect of many psychotropic medications is fatigue, in which case self-injury or stereotypy may decrease due to a general lack of energy. Although this is an ideal outcome for the target behavior, this lethargy will persist across the individual's life and may impact other prosocial behaviors. Second, individuals with intellectual disabilities may have difficulties verbally reporting internal effects of the medication. In the absence of external evidence (e.g., vomiting), possible adverse effects may go unnoticed. Finally, some of the common adverse side effects of antipsychotics, antidepressants, and antianxiety medications include dizziness, fatigue, weight gain, sleep problems, and nausea. One severe side effect of some antipsychotic medications is tardive dyskinesia which causes stiff, jerky, involuntary extremity and facial movements and may persist even after discontinuing use of the medication (Tarsy & Baldessarini, 1984).

Recommendations for Practitioners An international guide to psychotropic prescriptions for individuals with intellectual disabilities, released by the world psychiatric association, recommended that if the individual does not have an accompanying diagnosis of a psychiatric disorder, non-medication-based treatments should be evaluated prior to prescribing psychotropic medication (Deb et al., 2009). The report does not prohibit the prescription of psychotropic drugs but does encourage careful consideration of other treatment options. If the treatment team determines medications should be incorporated into the treatment plan, medications should be prescribed at the lowest dose, for the shortest duration necessary (Deb et al., 2009). In addition, accurate and detailed data on the client's engagement in self-injury and stereotypy should be documented. Modifications to prescriptions (e.g., dosage, time of delivery) should be clearly denoted for visual analysis to determine corresponding behavior changes.

A final recommendation for practitioners is to avoid simultaneously introducing multiple treatment changes. For example, if the client's dosage of medication is altered, maintain all the other treatment components. This increases the likelihood that behavior change will be appropriately attributed to the environmental variables which lead to the change. Practitioners should follow this recommendation for all treatment packages (even those which do not include medication) because it is imperative to ensure only the treatment components which impact the behavior are included in the final treatment plan.

Conclusion

In conclusion, there are several treatment strategies for addressing self-injurious behavior and stereotypy. Many of the decisions related to treatment selection should be based on the topography of the target behavior (i.e., self-injury or stereotypy), results from functional behavior assessment, and the age and skill level of the individual. Although effective individually, the research suggests that a treatment package with multiple components may be most effective for addressing self-injury and stereotypy.

References

Ahearn, W. H., Clark, K. M., & MacDonald, R. P. F. (2007). Assessing and treating vocal stereotypy in children with autism. *Journal of Applied Behavior Analysis, 40*, 263–275. https://doi.org/10.1901/jaba.2007.30-60

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

Arndorfer, R. E., & Miltenberger, R. G. (1993). Functional assessment and treatment of challenging behavior: A review with implications for early childhood. *Topics in Early Childhood Special Education, 13*, 82–105. https://doi.org/10.1177/027112149301300109

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, NY: Appleton-Century Crofts.

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

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

Berg, W. K., Wacker, D. P., Harding, J. W., Ganzer, J., & Barretto, A. (2007). An evaluation of multiple dependent variables across distinct classes of antecedent stimuli pre and post functional communication training. *Journal of Early and Intensive Behavior Intervention, 4*, 305–333. https://doi.org/10.1037/h0100346

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*, 604–617. https://doi.org/10.1111/jir.12359

Brawley, E. R., Harris, F. R., Allen, K. E., Flemming, R. S., & Peterson, R. F. (1969). Behavior modification of an autistic child. *Behavioral Science, 14*, 87–97.

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*, 180–191. https://doi.org/10.1089/cap.2017.0003

Carr, E. G., & Durand, V. M. (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

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

Cooper, J. O., Heron, T. E., & Heward, W. L. (2007). *Applied behavior analysis* (2nd ed.). Upper Saddle River, NJ: Pearson Education.

Davis, T. N., Dacus, S., Strickland, E., Machalicek, W., & Coviello, L. (2013). Reduction of automatically maintained self-injurious behavior utilizing noncontingent matched stimuli. *Developmental Neurorehabilitation, 16*, 166–171. https://doi.org/10.3109/17518423.2013.766819

Deb, S., Kwok, H., Bertelli, M., Salvador-Carulla, L., 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*, 181–186.

DeLeon, I. G., & Iwata, B. A. (1996). Evaluation of a multiple-stimulus presentation format for assessing reinforcer preferences. *Journal of Applied Behavior Analysis, 29*, 519–533.

DeRosa, N. M., Roane, H. S., Wilson, J. L., Novak, M. D., & Silkowski, E. L. (2015). Effects of arm-splint rigidity on self-injury and adaptive behavior. *Journal of Applied Behavior Analysis, 48*, 860–864. https://doi.org/10.1002/jaba.250

Deshais, M. A., Fisher, A. B., Hausman, N. L., & Kahng, S. (2015). Further investigation of a rapid restraint analysis. *Journal of Applied Behavior Analysis, 48*, 845–859. https://doi.org/10.1002/jaba.251

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.

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*, 217–230.

Durand, V. M., & Carr, E. G. (1991). Functional communication training to reduce challenging behavior: Maintenance and application in new settings. *Journal of Applied Behavior Analysis, 24*, 251–264. https://doi.org/10.1901/jaba.1991.24-251

Falcomata, T. S., & Wacker, D. P. (2013). On the use of strategies for programming generalization during functional communication training: A review of the literature. *Journal of Developmental and Physical Disabilities, 25*, 5–15. https://doi.org/10.1007/s10882-012-9311-3

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.

Fisher, W. W., Greer, B. D., Mitteer, D. R., Fuhrman, A. M., Romani, P. W., & Zangrillo, A. N. (2018). Further evaluation of differential exposure to establishing operations during functional communication training. *Journal of Applied Behavior Analysis, 51*, 360–373. https://doi.org/10.1002/jaba.451

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.

Greer, B. D., Fisher, W. W., Saini, V., Owen, T. M., & Jones, J. K. (2016). Functional communication training during reinforcement schedule thinning: An analysis of 25 applications. *Journal of Applied Behavior Analysis, 49*, 105–121. https://doi.org/10.1002/jaba.265

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

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

Hammond, J. L., Iwata, B. A., Fritz, J. N., & Dempsey, C. M. (2011). Evaluation of fixed momentary DRO schedules under signaled and unsignaled arrangements. *Journal of Applied Behavior Analysis, 44*(1), 69–81. https://doi.org/10.1901/jaba.2011.44-69

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

Hellings, J. A., Zarcone, J. R., Reese, R. M., Valdovinos, M. G., Marquis, J. G., Fleming, K. K., & Schroeder, S. R. (2006). A crossover study of risperidone in children, adolescents and adults with mental retardation. *Journal of Autism and Developmental Disorders, 36*, 401–411.

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

Horner, R. H., & Day, H. M. (1991). The effects of response efficiency on functionally equivalent competing behaviors. *Journal of Applied Behavior Analysis, 24*, 719–732. https://doi.org/10.1901/jaba.1991.24-719

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). https://doi.org/10.1901/jaba.1994.27-197

Ji, N. Y., & Findling, R. L. (2016). Pharmacotherapy for mental health problems in people with intellectual disability. *Current Opinion in Psychiatry, 29*, 103–125. https://doi.org/10.1097/YCO.0000000000000233

Jones, R. S., & Baker, L. J. (1990). Differential reinforcement and challenging behaviour: A critical review of the DRI schedule. *Behavioural Psychotherapy, 18*(1), 35–47.

Jones, R. S. P., & Baker, L. J. V. (1988). The differential reinforcement of incompatible responses in the reduction of self-injurious behavior: A pilot study. *Behavioural Psycholtherapy, 16*, 323–328.

Kahng, S., Abt, K. A., & Wilder, D. (2001). Treatment of self-injury correlated with mechanical restraints. *Behavioral Interventions, 16*, 105–110. https://doi.org/10.1002/bin.86

Kurtz, P. F., Chin, M. D., Huete, J. M., Tarbox, R. S. F., 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*, 205–219.

Lalli, J. S., Casey, S., & Kates, K. (1995). Reducing escape behavior and increasing task completion with functional communication training, extinction and response chaining. *Journal of Applied Behavior Analysis, 28*, 261–268. https://doi.org/10.1901/jaba.1995.28-261

Loman, S. L., & Horner, R. H. (2014). Examining the efficacy of a basic functional behavioral assessment training package for school personnel. *Journal of Positive Behavior Interventions, 16*, 18–30. https://doi.org/10.1177/1098300712470724

Lovaas, O. I., Freitag, G., Gold, V. J., & Kassorlia, I. C. (1965). Experimental studies in childhood schizophrenia: Analysis of self-destructive behavior. *Journal of Experimental Child Psychology, 2*, 67–84.

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

Luiselli, J. K., Colozzi, G. A., Helfen, C. S., & Pollow, R. S. (1980). Differential reinforcement of incompatible behavior (DRI) in treating classroom management problems of developmentally disabled children. *The Psychological Record, 30*, 261–270.

Lunsky, Y., Khuu, W., Tadrous, M., Vigod, S., Cobigo, V., & Gomes, T. (2018). Antipsychotic use with and without comorbid psychiatric diagnosis among adults with intellectual and developmental disabilities. *Canadian Journal of Psychiatry. Revue Canadienne De Psychiatrie, 63*, 361–369. https://doi.org/10.1177/0706743717727240

Lustig, N., Ringdahl, J., Breznican, G., Romani, P., Scheib, M., & Vinquist, K. (2014). Evaluation and treatment of socially inappropriate stereotypy. *Journal of Developmental & Physical Disabilities, 26*, 225–235.

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

Mayer, G. R., Sulzer-Azaroff, B., & Wallace, M. (2012). *Behavior analysis for lasting change* (2nd ed.). Corwnall-on-Hudson, NY: Sloan Publishing.

Mildon, R. L., Moore, D. W., & Dixon, R. S. (2004). Combining noncontingent escape and functional communication training as a treatment for negatively reinforced disruptive behavior. *Journal of Positive Behavior Interventions, 6*, 92–102. https://doi.org/10.1177/10983007040060020401

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*, 73–77.

Perry, B. I., Kwok, H. F., Mendis, J., Purandare, K., Wijeratne, A., Manjubhashini, S., … Cooray, S. E. (2018). Problem behaviours and psychotropic

medication use in intellectual disability: A multinational cross-sectional survey. *Journal of Intellectual Disability Research, 62*, 140–149. https://doi.org/10.1111/jir.12471

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

Radley, D. C., Finkelstein, S. N., & Stafford, R. S. (2006). Off-label prescribing among office-based physicians. *Archives of Internal Medicine, 166*, 1021–1026.

Rapp, J. T. (2006). Toward an empirical method for identifying matched stimulation for automatically reinforced behavior: A preliminary review. *Journal of Applied Behavior Analysis, 39*, 137–140.

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.

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.

Reynolds, G. S. (1961). Behavioral contrast. *Journal of the Experimental Analysis of Behavior, 4*(1), 57–71. https://doi.org/10.1901/jeab.1961.4-57

Richman, D. M., Lindauer, S. E., Crossland, K. A., McKerchar, T. L., & Morse, P. S. (2001). Functional analysis and treatment of breath holding maintained by nonsocial reinforcement. *Journal of Applied Behavior Analysis, 34*, 531–534.

Rooker, G. W., Bonner, A. C., Dillon, C. M., & Zarcone, J. R. (2018). Behavioral treatment of automatically reinforced sib: 1982–2015. *Journal of Applied Behavior Analysis, 51*, 974–997. https://doi.org/10.1002/jaba.492

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

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*. https://doi.org/10.1136/bmj.h4326

Skinner, B. F. (1953). *Science and human behavior*. New York, NY: Macmillan.

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*, 367–370.

Tarsy, D., & Baldessarini, R. J. (1984). Tardive dyskinesia. *Annual Review of Medicine, 35*, 605–623. Retrieved from http://ezproxy.baylor.edu/login?url=http://search.ebscohost.com/login.aspx?direct=true&db=cmem&AN=6144288&site=ehost-live&scope=site

Tiger, J. F., 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

Toussaint, K. A., & Tiger, J. H. (2012). Reducing covert self-injurious behavior maintained by automatic reinforcement through a variable momentary DRO procedure. *Journal of Applied Behavior Analysis, 45*, 179–184. https://doi.org/10.1901/jaba.2012.45-179

Tyrer, P., Oliver-Africano, P. C., Ahmed, Z., Bouras, N., Cooray, S., Deb, S., … Crawford, M. (2008). Risperidone, haloperidol, and placebo in the treatment of aggressive challenging behaviour in patients with intellectual disability: A randomised controlled trial. *The Lancet, 371*, 57–63. https://doi.org/10.1016/S0140-6736(08)60072-0

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.

Wacker, D. P., Steege, M. W., Northup, J., Sasso, G., Berg, W., Reimers, T., … Donn, L. (1990). A component analysis of functional communication training across three topographies of severe behavior problems. *Journal of Applied Behavior Analysis, 23*, 417–429. https://doi.org/10.1901/jaba.1990.23-417

Wallace, M. D., Iwata, B. A., Zhou, L., & Goff, G. A. (1999). Rapid assessment of the effects of restraint on self-injury and adaptive behavior. *Journal of Applied Behavior Analysis, 32*, 525–528. https://doi.org/10.1901/jaba.1999.32-525

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

Weston, R., Hodges, A., & Davis, T. N. (2018). Differential reinforcement of other behaviors to treat challenging behaviors among children with autism: A systematic and quality review. *Behavior Modification, 42*, 584–609.

Wong, C., Odom, S. L., Hume, K., Cox, A. W., Fettig, A., Kucharczyk, S., … Schultz, T. R. (2014). *Evidence-based practices for children and young adults with autism spectrum disorder*. Chapel Hill, NC: The University of North Carolina, Frank Porter Graham Child Development Institute, Autism Evidence-Based Practice Review Group.

Worsdell, A. S., Iwata, B. A., Hanley, G. P., Thompson, R. H., & Kahng, S. W. (2000). Effects of continuous and intermittent reinforcement for problem behavior during functional communication training. *Journal of Applied Behavior Analysis, 33*, 167–179. https://doi.org/10.1901/jaba.2000.33-167

Treatment of Feeding Problems in Dual Diagnosis

35

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Introduction

A previous chapter in this text focused on the assessment of feeding disorders. As such, the present chapter addresses the treatment of feeding problems. Specifically, this chapter focuses on feeding intervention from an applied behavior analytic (ABA) perspective. There are a number of factors to consider *prior* to initiating a feeding intervention, however. Included among these factors are an interdisciplinary assessment/consultation and the competence and experience of the therapist with treating feeding problems. For example, it is important to understand the extent to which medical and/or developmental factors may be contributing to the presence of a feeding problem prior to developing an intervention (e.g., the ability to chew and swallow food). In the absence of such information, treating a feeding problem could be dangerous and potentially worsen an existing concern. This is a particularly important consideration with feeding problems, as several medical factors may contribute to the development and maintenance of feeding problems, and the multidisciplinary nature of food consumption and digestion requires disciplinary expertise outside of behavior analytic psychology. Similarly, it is imperative for behavior analysts to consider their own expertise and competence before treating a feeding problem. Without specific expertise in the area, a therapist should refer the case to someone with such expertise or only move forward with the appropriate supervision and consultation. The decision to develop a behavioral intervention for a feeding problem should not be taken lightly.

After deciding to treat a feeding problem, a behavior analytic intervention must select a socially significant, measurable target behavior that can be analyzed objectively over time (see Baer, Wolf, & Risley, 1968). Within the feeding literature, common targets include acceptance and consumption of bites of food and inappropriate mealtime behavior (IMB; more on this below). Beyond these common target behaviors, a range of other behaviors have also been the focus of intervention, including chewing (e.g., Volkert, Peterson, Zeleny, & Piazza, 2014), packing (e.g., Silbaugh, Swinnea, & Penrod, 2018), and more. In addition, while frequency counts have been the most common dimension of behavior measured, other dimensions have been considered as well (e.g., inter-response time in the treatment of rapid eating; Page, Griffith, & Penrod, 2017). As with all interventions, behavior

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analysts should carefully consider both the short- and long-term goals of an intervention to select a socially significant target behavior and measurement system (see Wolf, 1978).

The following sections take a closer look at two important variables to consider in the context of a feeding intervention: food preference and IMB.

Food Preference

Although a number of factors may be measured in the context of a feeding intervention, it seems that potential shifts in food preferences are an especially important factor to consider. Indeed, systematically examining whether preferences for various foods change following intervention may provide valuable information that is not captured by changes in the number of bites consumed. In other words, it is one thing to know that an individual is consuming more bites presented by a therapist but another to know that their preferences for previously nonpreferred foods have also shifted (i.e., that previously nonpreferred foods are now preferred). Although only about a third of published feeding studies conducted with children with autism spectrum disorder (ASD) reported the use of stimulus preference assessments (SPAs) during intervention (Silbaugh et al., 2016), clinicians are encouraged to incorporate these procedures into treatment plans to directly assess whether preferences for new foods are established.

Preference Assessments

Stimulus preference assessments often involve the presentation of a series of choices between alternatives and direct observation of a client's approach, selection, or consumption of one option over others. The number of times each option was selected is then divided by the total number of times the item was available to calculate a percentage of selection. These percentages are then rank ordered from highly preferred to moderately and low preferred (DeLeon & Iwata, 1996; Fisher et al., 1992; Pace, Ivancic, Edwards, Iwata, & Page, 1985). Several SPA variations exist (e.g., multiple stimulus without replacement [DeLeon & Iwata, 1996], single stimulus [Pace et al., 1985]), but the paired stimulus preference assessment (Fisher et al., 1992) is most commonly used to determine relative preferences in feeding interventions (Silbaugh et al., 2016). Clinicians can use SPA procedures prior to intervention to confirm if foods reported to be regularly consumed or refused actually evoke such responses. These procedures can also be useful in identifying highly preferred foods and tangible items (e.g., toys, videos) that may function as effective reinforcers during a behavioral intervention.

In addition to employing a SPA to identify target foods and potential reinforcers at the onset of intervention, preference for both target foods and nontarget foods can be assessed before *and* after intervention to determine if there has been a change in relative preferences. If a client demonstrates increases in preference for foods targeted during intervention, this may be indicative of whether treatment outcomes are likely to maintain once treatment has concluded (Penrod & Van Dalen, 2010), since clients are more likely to consume foods that are preferred. Furthermore, if increases in preference for nontarget foods occur, evidence of treatment generalization may also be discovered (Fernand, Penrod, Fu, Whelen, & Medved, 2016; Penrod, Gardella, & Fernand, 2012), and intervention components may be systematically eliminated. Conducting pre- and post-intervention SPAs, therefore, provides an objective gauge of whether a meaningful expansion of the food repertoire has occurred.

Despite the need to better understand these long-term and repertoire-altering effects of feeding interventions, relatively few studies have assessed whether preferences developed as an outcome of treatment. An initial example of how clinicians might incorporate pre- and post-intervention SPAs into practice was outlined in a series of studies conducted by Penrod and Van Dalen (2010; Van Dalen & Penrod, 2010). In the first study (Van Dalen & Penrod, 2010), researchers compared the effects of simultaneous

presentation of highly preferred and nonpreferred foods to a sequential presentation method where preferred foods were presented contingent on consumption of nonpreferred foods. When combined with escape extinction in the form of nonremoval of the spoon (NRS), both presentation methods resulted in increases in consumption, indicating the NRS treatment component was likely responsible for behavior change.

In an extension of this study, Penrod and Van Dalen (2010) examined pre- and post-intervention preference assessment data to explore whether intervention resulted in increased preferences for target and nontarget foods. All three participants (two who participated in the first investigation) developed preferences for previously nonpreferred foods after exposure to those foods during treatment and generalization sessions. These results may explain why treatment outcomes maintain after intervention concludes and may provide insights about the long-term efficacy of feeding treatments.

Fernand et al. (2016) presented another demonstration of changes in preference using pre- and post-intervention SPAs in their study exploring the effects of choice as an antecedent-based intervention for food selectivity. Researchers compared consumption and IMB across choice and no-choice conditions in a multielement single-case experimental design with two participants. In the choice condition, participants were presented with a choice between four nonpreferred foods, while in the no-choice condition, participants were not presented with the opportunity to choose between foods. When choice alone was ineffective in increasing consumption for either participant, NRS was introduced. Once NRS was implemented, choice was effective in increasing consumption for one participant and in decreasing IMB for the other. Moreover, previously nonpreferred foods were selected and consumed in the post-intervention SPA as often or more frequently than previously preferred foods and preferences for several new foods developed for both participants. Collectively, these studies indicate behavioral feeding interventions can facilitate the development of robust food repertoires that are likely to persist in natural mealtime environments. Even if such promising results had not been achieved, incorporating SPAs into treatment plans can still provide valuable data to guide ongoing service delivery.

Whelan and Penrod (2018) provide an example of how SPA data might be used to inform treatment plans. This study examined a sequential meal presentation method with two typically developing, picky eaters. Initially, preferred and nonpreferred foods, identified through SPAs, were presented simultaneously on a plate with no requirement to consume either. When only miniscule amounts of target foods were consumed, an appetizer condition was implemented. This condition was intended to capitalize on the increased motivation to eat at the start of meals as the nonpreferred food was presented for 10 min prior to the presentation of the preferred food. Again, no requirement to consume either food was in place, and meals were terminated after 20 min. Findings replicate prior research (Piazza et al., 2002; Pizzo, Coyle, Seiverling, & Williams, 2012; Van Dalen & Penrod, 2010) in that consumption of the target food only increased in a final treatment phase where presentation of the preferred food was made contingent on consumption of the target food.

In the Whelan and Penrod (2018) study, participants' post-intervention SPAs demonstrated that preferences for target foods (i.e., nonpreferred foods) increased following treatment. Unlike results in prior research (Fernand et al., 2016; Penrod & Van Dalen, 2010), however, no generalization of treatment effects with nontarget foods occurred. While such a finding may be disappointing from a clinical standpoint, it provides beneficial information beyond what would be gathered by measuring more common dependent variables alone. Progress (or lack thereof) demonstrated in SPAs conducted before, after, and even during intervention can serve as a powerful, clinical decision-making tool to ensure treatment continues until a healthy variety of foods are readily consumed. Though additional research to determine the specific client characteristics, intervention components, and necessary treatment dosages required to establish such rep-

ertoires is needed, SPAs serve as an easy-to-implement, practical strategy to determine when intervention components can be removed for individual clients.

Thus, the use of pre- and post-intervention SPAs is recommended, both in research and in practice. Clinicians, however, should be aware of a potential difficulty they may encounter in assessing clients' food preferences. Since typical SPAs examine selection and consumption of items assessed as the dependent variable, this can sometimes be problematic when working with individuals with selective feeding repertoires as they may never select or consume any of the foods presented. Such an outcome would confirm that foods assessed are, in fact, nonpreferred and may be suitable as targets, yet no relative hierarchy of preference can be generated. It may also be assumed that all foods are equally nonpreferred although everyday experience dictates some nonpreferred foods are likely to be more aversive and less palatable than others. As such, additional methods to evaluate food preferences with highly selective clients may be necessary as assessing relative preferences may identify foods that are nonpreferred but less aversive and, therefore, more likely to be consumed than others.

Avoidance Assessments An alternative method of assessing preference when standard SPAs prove uninformative was outlined by Kunkel, Kozlowski, Taylor, and González (2018). In the first experiment, researchers conducted an avoidance assessment with foods never selected or consumed in a traditional SPA. The avoidance assessment procedure was adapted from avoidance assessments originally developed for use in the treatment of severe problem behavior (Call, Pabico, & Lomas, 2009; Fisher et al., 1994; Zarcone, Crosland, Fisher, Worsdell, & Herman, 1999) and involved repeated presentations of various foods on a plate with the instruction to "Take a bite." Participants could either independently consume all or a portion of the bite, or after 5 s without independent consumption, the bite was placed on a spoon and held at the participants' lips. If, after 30 s, the participant still had not consumed the food, the bite was removed and the next bite was presented. Each food was presented, in a randomized order, for a total of two, three-trial sessions, and data were collected on the duration of negative vocalizations, the number of grams consumed, and rate of combined avoidance responses per minute. Avoidance responses collectively included IMB such as head turns, covering mouth, throwing food, attempts to block the utensil or feeder's arm or hand, and expulsions, self-injurious, aggression, and disruptive behavior.

Foods evoking the highest rates of avoidant responses, the lowest levels of consumption, or both were hypothesized to be the most aversive foods, while foods with the lowest levels of avoidance responses, highest levels of consumption, or both were hypothesized as least aversive. This procedure was effective in producing a hierarchy of avoidance for all five participants. In Experiment 2 (Kunkel et al., 2018), researchers used a negative reinforcer assessment to determine if foods hypothesized to be most aversive functioned as negative reinforcers for the consumption of the least aversive foods. For two of three participants who completed Experiment 2, the food hypothesized to be most aversive in the avoidance assessment functioned as a negative reinforcer when presented contingent on refusal of the less aversive food. In other words, participants' consumption of the less aversive food increased when doing so allowed them to avoid the presentation of the more aversive food, validating the preference hierarchy established in the avoidance assessment.

This study demonstrated that nonpreferred foods are not equally nonpreferred and that adaptations of SPA procedures, such as avoidance assessments, may be an effective means of identifying relative preferences with highly selective clients. Alternative means of identifying preferences are needed, and research is currently underway to identify additional methods of establishing preference hierarchies by examining the level of interaction with various foods using the progressive demand fading steps outlined in Penrod et al. (2012). Development of such methods holds great potential to aid clinicians in identifying

foods that are less aversive and more likely to be consumed. By starting intervention with these foods, clinicians may effectively decrease IMB and establish new foods as primary reinforcers (Kunkel et al., 2018). This may allow clients to make treatment gains more quickly, remediating health risks associated with feeding disorders and promoting the acceptability of interventions.

Inappropriate Mealtime Behavior

While target behaviors related to food consumption are common within a behavioral intervention, behavioral intervention should also target IMB when it is present. IMB is a collective term used to encompass various topographies of problem behavior that accompany food refusal during mealtimes. IMB can include head turns; mouth covering; pushing food, utensils, or the feeder's hand away from the mouth; expulsions after acceptance; and negative vocalizations (Levin & Carr, 2001; Sharp, Jaquess, Morton, & Herzinger, 2010; Silbaugh et al., 2016). More serious topographies of problem behavior such as gagging, vomiting, aggression, property destruction, and self-injury (e.g., Borrero, England, Sarcia, & Woods, 2016; Borrero, Woods, Borrero, Masler, & Lesser, 2010; Piazza et al., 2003) may also be included in individualized operational definitions of IMB. Like any challenging behavior, IMB is problematic due to the disruption caused to daily routines and family context more generally. Moreover, depending on the specific topography of behavior, IMB may present danger to one's safety, health, and well-being.

For many families, mealtimes provide structure and an opportunity for positive parent-child interactions (Spagnola & Fiese, 2007). For families with children with IMB, on the other hand, this is often not the case. Curtin et al. (2015) found that children with food selectivity were more likely to demonstrate IMB and that both were associated with increases in parental stress. This finding held true for both typically developing participants and those with an ASD diagnosis. Ausderau and Juarez (2013) reported similar results when they interviewed six mothers of children with ASD who exhibited feeding difficulties. All six mothers stated that adapting mealtime routines in attempts to get their child to eat was an exhausting task that strained the entire family. Since parents are expected to ensure their child obtains proper nutrition, some may feel personally responsible for their child's chronic feeding difficulties (Craig, Scambler, & Spitz, 2003), and this can lead to emotional distress (Budd et al., 1992).

This parental stress may exacerbate feeding problems and IMB because it has been shown to affect the child as well as the parent (Abidin, 1992; Crnic & Greenberg, 1990). This is because high levels of parental stress have been associated with fewer positive parent-child interactions (Downey & Coyne, 1990; McKay, Pickens, & Stewart, 1996). Over time, parents of children with feeding difficulties may exhibit more frequent signs of anger and frustration during repeated attempts to feed their child (Greer, Gulotta, Masler, & Laud, 2008). For example, Polan and Ward (1994) found that parents of children with severe feeding disorders were much more physically intrusive during feedings than parents of children without such difficulties. These signs of parent frustration may cause further increases in IMB as the child attempts to avoid mealtimes (Didehbani, Kelly, Austin, & Wiechmann, 2011; Feldman, Keren, Gross-Rozval, & Tyano, 2004).

Antecedent Interventions

Antecedent interventions may be an effective method of mitigating this ineffective cycle of IMB and parental stress. This is because antecedent interventions tend to be less intrusive and, therefore, may be more acceptable to parents who have difficulty tolerating IMB. Common antecedent-based feeding interventions include using the high-probability instructional sequence (Dawson et al., 2003; Meier, Fryling, & Wallace, 2012; Patel et al., 2006, 2007), varying the order in which preferred and nonpreferred foods are presented (sequential versus simultaneous presentation; Ahearn, 2003; Piazza et al., 2002;

Van Dalen & Penrod, 2010; Whelan & Penrod, 2018), presenting the bolus on a flipped spoon (Dempsey, Piazza, Groff, & Kozisek, 2011; Rivas, Piazza, Kadey, Volkert, & Stewart, 2011), modeling consumption (Fu et al., 2015; Seiverling, Harclerode, & Williams, 2014; Sira & Fryling, 2012), providing choices (Fernand et al., 2016), fading across texture or flavor dimensions (Freeman & Piazza, 1998; Patel, Piazza, Kelly, Ochsner, & Santana, 2001; Reitman & Passeri, 2008; Sharp, Trumbull, & Lesack, 2015), and using noncontingent reinforcement (Reed et al., 2004; Wilder, Normand, & Atwell, 2005).

Although the effectiveness of antecedent interventions may depend on the severity of the feeding disorder and individual client history, antecedent-based strategies may facilitate improvements in IMB by devaluing escape as a reinforcer or making eating less effortful. These less-intensive presentation, preparation, and environmental enrichment strategies may facilitate treatment gains without the need for more intrusive, consequence-based interventions. In addition, a review by Seubert, Fryling, Wallance, Jimenez, and Meier (2014) found that even when antecedent interventions alone were ineffective in treating food selectivity and food refusal, they enhanced the efficacy of escape extinction in more than two thirds of the studies examined. Because it is important to treat the feeding problem as well as IMB, clinicians are encouraged to consider the use of antecedent interventions both alone and in combination with other intervention components to develop comprehensive treatment plans.

Functional Assessment of IMB

Should consequence-based procedures be required to achieve socially significant reductions in IMB, additional intervention components can be identified by initially assessing the environmental events surrounding its occurrence. Both descriptive assessments and functional analysis methodologies have proven effective in identifying the function of IMB (Borrero et al., 2016). Descriptive assessments of IMB may involve parents completing surveys or rating scales (e.g., Galensky, Miltenberger, Stricker, & Garlinghouse, 2001) and data collection on naturally occurring antecedent-behavior-consequence relations (Borrero et al., 2010; Galensky et al., 2001; Woods, Borrero, Laud, & Borrero, 2010). Alternatively, functional analyses of IMB involve the systematic manipulation of potential maintaining variables to isolate putative reinforcers (Bachmeyer et al., 2009; Piazza et al., 2003). Teaching parents to implement these functional assessment procedures may have several advantages.

Since researchers have demonstrated that functional analysis results may vary depending on who implements procedures (Ringdahl & Sellers, 2000), implementation by familiar individuals ensures relevant variables that maintain IMB in the natural environment are captured in the analysis (English & Anderson, 2004). Najdowski, Wallace, Doney, and Ghezzi (2003) successfully trained a mother to implement a functional analysis with her 5-year-old child with autism. The functional analysis indicated food refusal and IMB were maintained by negative reinforcement. The parent then implemented a function-based intervention consisting of differential reinforcement of an alternative behavior, escape extinction, and demand fading to effectively decrease refusal behavior. In a replication and extension of that study, Najdowski et al. (2008) taught six parents to implement functional analyses in a 1-hour training comprised of didactic instruction and modeling of procedures. This brief training and in vivo feedback during the functional analysis resulted in high levels of procedural fidelity and identification of function for all child participants.

Hodges et al. (2018) presented a promising initial demonstration of an alternative to the traditional functional analysis, the trial-based functional analysis (TBFA). In this study, researchers implemented both a traditional functional analysis (as described by Iwata, Dorsey, Slifer, Bauman, & Richmand, 1982\1994) and TBFA (as described by Rispoli et al., 2015) to identify the function of IMB demonstrated by two participants with developmental delays. When

results of the TBFA corresponded with those from the traditional functional analysis for both participants, an effective function-based intervention to decrease IMB was developed. Although the TBFA in this study was not conducted by a parent, prior research indicates caregivers and entry-level staff can readily be trained to implement TBFA procedures (e.g., Lambert, Bloom, Clay, Kunnavatana, & Collins, 2014; Lambert, Bloom, Kunnavatana, Collins, & Clay, 2013).

Since some have suggested that TBFA procedures, data collection, and analysis may be easier for those less familiar with behavioral interventions to implement (Rispoli, Neely, Healy, & Gregori, 2016), future research should explore the use of TBFAs for IMB with parents as implementers.

Although functional analysis has been considered the gold standard in functional assessment methodology and parents can be trained to implement the procedures with fidelity, a recent investigation by Borrero et al. (2016) indicates that descriptive assessments may be sufficient to generate a clinically useful hypothesis about the function of IMB. Researchers found that there was higher correspondence between descriptive assessments of IMB and results of functional analyses than has been reported in the assessment of other problem behaviors. This finding suggests that it may be more efficient to start by teaching parents to conduct descriptive assessments of IMB and proceed to functional analysis only when necessary. Regardless of how behavioral function is examined, including parents in the assessment of IMB not only ensures that data accurately reflects behavior in everyday mealtimes but also provides parents with knowledge and skills to better understand behavior more generally. While more research is needed in this important area, it is clear that functional assessment and analysis is fundamental to understanding intervention planning for IMB.

Thus far, we have considered two important factors to consider in behavioral feeding treatment, food preference, and IMB. In the following section, we describe research on self-feeding and the use of modeling contingencies.

Self-Feeding

For many individuals receiving feeding intervention, a long-term goal is to establish independent, self-feeding. For typically developing children, feeding often progresses from initially being fed by a caregiver to self-feeding before the age of 2 (Carruth, Ziegler, Gordon, & Hendricks, 2004). Self-feeding involves a large number of skills that may be overlooked as they may seem to develop somewhat seamlessly in many typically developing children. These include skills involved in gross motor development such as sitting independently and holding one's head up with their neck. In addition, self-feeding requires using a spoon or fork, bringing the food to the mouth, removing food from the utensil, drinking from a sippy and ultimately a regular cup, chewing, and swallowing. Many self-feeding skills are associated with corresponding developmental changes, such as the development of teeth in the child's mouth (Carruth et al., 2004). Unfortunately, less is understood about the development of self-feeding among children with feeding problems (Rivas et al., 2014). Indeed, while much research has been conducted on food consumption with therapist presented bites, relatively less research has been conducted on establishing self-feeding. Given the importance of developing this behavior, both for the individual to become more independent and to promote more long-term and generalizable treatment gains, the topic of self-feeding is deserving of more attention by researchers and clinicians. In what follows, we provide an overview of the behavior analytic literature on self-feeding.

Overview of the Research on Self-Feeding

As we mentioned, relatively little research has been conducted in the area of self-feeding. Much of the research that has been conducted centers around behavioral economics and the concept of response effort. For example, varying the number of responses or bites required and the amount of reinforcement have been explored as response

effort manipulations with the aim of shifting responding toward independent feeding (Kerwin, Ahearn, Eicher, & Burd, 1995). This type of effort manipulation might involve arranging for a single self-fed bite versus multiple caregiver-fed to increase self-feeding. Vaz, Volkert, and Piazza (2011) conducted a study using negative reinforcement (i.e., avoidance of nonpreferred foods) to increase self-feeding. In this study, target foods were identified for self-feeding as well as avoidance foods. Avoidance was arranged by increasing the response effort for being fed and decreasing the response effort for self-feeding. Response effort consisted of increasing the number of bites offered by the feeder (e.g., four bites of avoidance food) versus those required by self-feeding (e.g., one bite of target food). This arrangement resulted in a shift to self-feeding one bite of target food from being fed multiple bites of both the targeted and avoidance foods.

Similarly, Rivas et al. (2014) conducted response effort manipulations exploring whether the number of bites or the quality of avoidance food was responsible for the shift to self-feeding. The number of self-feeding bites remained constant, with a one bite requirement. The ratio of caregiver and self-fed bites was manipulated by gradually increasing the number of caregiver-fed bites (e.g., 1 self-fed to 1 caregiver-fed bites [1:1], then 1:2, then 1:3, etc.) until the participants chose to self-feed. This arrangement successfully increased self-feeding for two of the three participants. For the third participant, researchers identified a food that was least preferred (i.e., an avoidance food), which the caregiver presented if the child did not self-feed. When the effort manipulation failed, this procedure effectively increased self-feeding for this participant.

Volkert, Piazza, and Price (2016) conducted replications of the response effort manipulations described above to demonstrate how these procedures may be associated with the recurrence of previously treated behavior with children with a history of feeding problems. Two of the participants in this study engaged in packing, which involves a child storing food in their mouth (e.g., between gums and cheek). Packing may occur when an individual accepts but does not consume a food. Given that children with feeding disorders may have a history of reinforcement associated with IMB (including packing), these same behaviors may resurface when the target of intervention changes. This study replicated response effort manipulations conducted in previous studies (Rivas et al., 2014; Vaz et al., 2011) and also implemented procedures that were used to treat food refusal initially. These results suggest clinicians should be sensitive to a client's behavioral history, including previous interventions, when initiating intervention to increase self-feeding.

Feeding researchers have also evaluated interventions to improve self-drinking since children with feeding disorders may not progress to age typical drinking without formal intervention. Collins, Gast, Wolery, Holcombe, and Leatherby (1991) used physical guidance with a constant time delay procedure and descriptive praise to increase self-drinking for a child with an intellectual disability and visual impairment. A study by Peterson, Volkert, and Zeleny (2015) extended upon this by evaluating similar procedure with children with feeding disorders specifically. Peterson et al. (2015) increased self-drinking for two children with a history of feeding disorders utilizing differential reinforcement with tangible items. Intervention procedures consisted of instructions and access to an identified reinforcer with descriptive praise contingent on self-drinking. Both studies demonstrated that common behavior analytic interventions can effectively be used to increase a variety of independent feeding behaviors.

Additional intervention procedures have been used to treat more idiosyncratic issues with self-feeding. Jenkins, LeBlanc, and Lambert (2017) evaluated a treatment for a 6-year-old boy with autism who required parental approval or prompts for every bite of food. This investigation evaluated the use of specific instructions which were then followed by a gradual removal of prompts (i.e., fading) to increase independent feeding. Multiple-bite instructions, no instructions, and self-management procedures were all evaluated to identify the most effective treatment proce-

dures. Multiple-bite instructions consisted of a descriptive prompt to take a certain number of bites (e.g., "Take [#] bites of macaroni and cheese"), with the number of bites increasing contingent upon success with fewer bites. The no instructions condition consisted of failing to provide any prompts, while self-management consisted of teaching the child to follow a rule provided by the authors (e.g., "first 3 bites of corn then 3 bites of mac and cheese"). Multiple-bite instructions proved more effective than other intervention components, increasing self-feeding to near independent feeding.

While self-feeding often develops without specific intervention for typically developing individuals without feeding problems, individuals with feeding disorders may require specific intervention, in the absence of which self-feeding may not develop. The literature on self-feeding is limited, however, with most of these studies evaluating the effects of negative reinforcement by arranging a choice for the participants to avoid being fed and encourage self-feeding. The use of negative reinforcement procedures utilizes aversive control, which could potentially contribute to a more aversive mealtime context more generally, especially if procedures are poorly administered. Continued research may explore the use of fading and transitioning to alternative, less-invasive procedures contingent upon initial success.

Additional research should also evaluate positive reinforcement procedures for increasing self-feeding. Children with feeding disorders may lack motivation to self-feed even after acquiring the skills necessary to self-feed. Preference assessment procedures may be used to identify more preferred foods that may be used as reinforcers to increase self-feeding and presumably condition the entire feeding context to be less aversive. It is also possible that transfer of stimulus control and/or exposure-based interventions might increase self-feeding for individuals that already demonstrate self-feeding skills (e.g., Sasaki & Fryling, 2014). Self-feeding represents an important area for ongoing research and clinical work. A lack of self-feeding may prevent individuals from accessing many beneficial social interactions, as well as create additional caregiver stress, and therefore warrants intervention.

Modeling Contingencies

Modeling has been effective in teaching a wide variety of skills within the field of ABA and has been used more recently in the treatment of feeding problems. Modeling is an antecedent-based intervention and may be preferred by some parents because it is relatively noninvasive. Candidates for a modeling intervention should display a generalized imitative repertoire in which the client can readily copy the behavior of others. In this section, we review studies where modeling was incorporated in a treatment package with the model being an adult, a peer in the child's life, or characters in a video.

In an initial study on the use of modeling in feeding intervention, Greer, Dorow, Williams, McCorkle, and Asnes (1991) conducted two experiments using a peer modeling procedure. In Experiment 1, the participant was an 18-month-old child diagnosed with gastroesophageal reflux and nemaline myopathy (a muscular disease) with a history of feeding via a gastrostomy tube (G-tube). After the child was medically cleared and no longer required the G-tube, he failed to successfully transition to oral consumption. The authors reported that the participant was verbal and would imitate his older sister so the 5-year-old sister served as the model in the study. During baseline, the children were presented with the same sized portions simultaneously and sessions alternated with the presentation of liquids in a cup. During treatment, a single bite was placed on the model's plate, the model consumed the bite, and a token and praise was provided. A single bite was then placed on the participant's plate, and this rotation continued to occur until the model consumed ten bites. The model then exchanged the tokens for preferred items. The intervention was unsuccessful for seven consecutive sessions; therefore, the lead experimenter introduced a clear syringe to deposit the liquids. The syringe was used to deposit orange juice in the model's mouth. After observing the liquid

deposit, the participant picked up the syringe and deposited the orange juice in the model's mouth just as the lead experimenter had done. Reinforcement was provided to the model, and following this the participant deposited the orange juice in his own mouth. After this successful session, the participant began to consume both solid foods and liquids from a cup and no longer required the use of the G-tube.

In Experiment 2 (Greer et al., 1991), the participant was a 2-year-old male who did not have any medical issues related to feeding; however, he consumed very few foods. The models in this study were other students at a preschool the participant was attending. At first, lunches were presented simultaneously along with an instruction to eat, using the child's name. When this was unsuccessful, a modeling component was included in which a bite was presented to a peer, along with praise for consumption, and then a bite was presented to the participant. This intervention increased consumption for the participant. Collectively, the two experiments presented demonstrated that modeling can be an effective, antecedent intervention to increase consumption.

Another study using modeling was conducted in a preschool with 8 boys and 12 girls between the ages of 2- and 5-year-olds (Horne et al., 2011). The study consisted of a treatment package with the use of a video modeling of consumption of target foods, a statement in the video of the foods to be consumed that day, a follow-up letter regarding the group's performance, and a differential reinforcement system with increased reinforcement contingent upon how many target foods were consumed (i.e., more target foods consumed resulted in more reinforcement). This study was successful increasing vegetable and fruit consumption from a baseline of one to two foods per day to an average of eight fruits and vegetables per day for each child in the group. Generalization to nontarget fruits and vegetables was also observed. While the extent to which the individuals in the study were selective or picky eaters is unknown, it is clear that behavior change occurred as a function of the minimally invasive modeling and reinforcement-based intervention.

Sira and Fryling (2012) evaluated a peer modeling procedure with a 9-year-old child diagnosed with ASD. Like Greer et al. (1991), the model was the participant's sibling who did not have a history of feeding difficulties. During baseline, the participant was presented with one bite of nonpreferred food every 30 s with the instruction "Take a bite." During treatment, the model and the participant were seated directly across from one another and a single bite was presented to the model, and once the model consumed the bite, praise and access to a preferred item or edible was granted. A bite was then presented to the participant, who was also told he had 30 s to consume the bite. Contingent upon consumption, the participant also received access to a reinforcer. This procedure continued until the model consumed ten bites of the nonpreferred foods. The procedure was effective in increasing consumption of three nonpreferred foods.

More recently, a study conducted by Fu et al. (2015) evaluated the use of modeling with two male children diagnosed with autism. Both of the participants in the study consumed a limited variety of foods, and the model used in the study was an adult. During baseline, consumption of the nonpreferred foods was modeled without any consequences for consumption. During treatment phases, a rule describing the contingency was utilized to inform the participant of what would be occurring during that phase. During the first phase of the treatment, modeling of differential reinforcement was conducted in which the model received access to the participant's preferred items and edibles contingent upon consumption of the nonpreferred foods. This treatment was partially effective for one participant for two different foods and unsuccessful for the other participant. The second phase of the study consisted of modeling NRS. In this phase, the NRS procedure was implemented with the model while the participant observed. The model engaged in 30–60 s of IMB that was similar in topography to behaviors previously demonstrated by participants. After 30–60 s, the model opened their mouth and allowed the experimenter to deposit the bite of food. Modeling NRS was effective for increasing consumption with all foods for both

participants. This result demonstrates that, for clients with ability to understand that what happens to others can happen them, direct contact with NRS may not always be necessary to increase consumption.

It should be noted that in all of the aforementioned studies, modeling was conducted in a treatment package. Therefore, it is difficult to determine which intervention component was effective in increasing consumption. Fu et al. (2015) attempted to rule out that modeling alone was not effective in increasing consumption by including modeling alone in baseline. However, even in this study modeling contingencies was not the sole component in the treatment phases as a rule was utilized that described the contingency which may have been effective in increasing consumption for participants. Additional research is still needed to determine the effects of modeling contingencies in isolation and the necessity of modeling specific consequences for consumption. Moreover, future research should continue to examine the extent to which modeling may be effective for participants with more significant feeding problems. Given that parents and other caregivers are most likely responsible for managing mealtimes in natural settings, the involvement of caregivers in assessment and intervention for feeding problems is an important area for research and practice. The final section of this chapter provides a brief overview of this topic specifically.

Caregiver Involvement in the Treatment of Feeding Problems

Treatment integrity, or the reliable and accurate implementation of an intervention, is a vital component of behavioral interventions. This is especially so when caregivers are implementing behavioral interventions during mealtimes at home. However, there is relatively limited research on caregivers implementing feeding interventions, and many feeding studies are conducted in a clinic setting. In fact, research indicates that caregivers implemented behavior interventions in less than 20% behavioral articles and less than 60% mentioned a caregiver training component after successful intervention was implemented by a clinician (Cosbey & Muldoon, 2017).

While more research is certainly needed, the existent research on the topic suggests that parents and caregivers can readily learn to implement behavioral interventions for feeding problems. For example, in a study conducted by Mueller, Piazza, Moore, and Kelley (2003), caregivers were trained through vocal and written instructions, modeling, and review of a video of their session and were provided performance feedback on their implementation of the intervention. Depending on the needs of the children in the study, parents were trained to conduct noncontingent reinforcement or a differential reinforcement of alternative behavior procedure along with NRS. In a second experiment, parents were trained various combinations of training components used in Experiment 1 to determine which were necessary to produce acceptable treatment integrity. Results indicated that modeling and rehearsal were effective in producing high levels of accurate implementation of the interventions prescribed.

In another example, Pizzo, Williams, Paul, and Riegel (2009) demonstrated that caregiver-implemented treatment can increase food consumption in a relatively short amount of time (approximately 2 weeks). The intervention consisted of repeated taste exposure and fading procedures in which novel foods were rotated with one another and the portion size systematically increased from a pea size to a full spoon size. An experimenter conducted the first session, described the procedures, and modeled implementation for caregivers. Caregivers implemented the intervention in all subsequent sessions. Caregivers were also trained to conduct generalization meals with novel foods at home. Once caregivers learned how to implement the procedures, they reported during the follow-up probe that their children were eating nearly twice or three times as many foods as they were prior to treatment.

While additional research on the involvement of parents is necessary, we know that parents can

be trained to implement interventions, and implement them accurately, in a relatively short period of time (e.g., Anderson & McMillan, 2001; Najdowski et al., 2003; Penrod, Wallace, Reagon, Betz, & Higbee, 2010). Future research should explore preference for various parent training procedures, the long-term effects of parent training programs, and various methods of providing consultation in home-based settings.

Conclusion

A large body of research has demonstrated the efficacy of behavioral interventions to treat feeding problems (Addison et al., 2012; Piazza, 2008; Silbaugh et al., 2016; Taylor, Wernimont, Northstone, & Emmet, 2015; Volkert & Piazza, 2012). The present chapter focused on several topics that are important to consider when developing a feeding intervention and highlighted areas for further research. Feeding is an important topic and not only related to health and development but also to socialization and much of day-to-day life. We hope this chapter has shed light on this important topic and that it stimulates further research and practice.

References

Abidin, R. R. (1992). The determinants of parenting behavior. *Journal of Clinical Child Psychology, 21*, 407–412. https://doi.org/10.1207/s15374424jccp2104_12

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. https://doi.org/10.1901/jaba.2012.45-455

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

Anderson, C. M., & McMillan, K. (2001). Parental use of escape extinction and differential reinforcement to treat food selectivity. *Journal of Applied Behavior Analysis, 34*, 511–515. https://doi.org/10.1901/jaba.2001.34-511

Ausderau, K., & Juarez, M. (2013). The impact of Autism spectrum disorders and eating challenges on family mealtimes. *Infant, Child, & Adolescent Nutrition, 5*, 315–323. https://doi.org/10.1177/1941406413502808

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. https://doi.org/10.1901/jaba.2009.42-641

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. https://doi.org/10.1901/jaba.1968.1-91

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 analysis of pediatric food refusal and acceptance. *Journal of Applied Behavior Analysis, 43*, 71–88. https://doi.org/10.1901/jaba.2010.43-71

Budd, K. S., McGraw, T. E., Farbisz, R., Murphy, T. B., Hawkins, D., Heilman, N., & Werle, M. (1992). Psychosocial concomitants of children's feeding disorders. *Journal of Pediatric Psychology, 17*(1), 81–94. https://doi.org/10.1093/jpepsy/17.1.81

Call, N. A., Pabico, R. S., & Lomas, J. E. (2009). Use of latency to problem behavior to evaluate demands for inclusion in functional analyses. *Journal of Applied Behavior Analysis, 42*, 723–728. https://doi.org/10.1901/jaba.2009.42-723

Carruth, B. R., Ziegler, P. J., Gordon, A., & Hendricks, K. (2004). Developmental milestones and self-feeding behaviors in infants and toddlers. *Journal of the American Dietetic Association, 104*, 51–56. https://doi.org/10.1080/07315724.2002.10719199

Collins, B. C., Gast, D. L., Wolery, M., Holcombe, A., & Leatherby, J. G. (1991). Using constant time delay to teach self-feeding to you students with severe/profound handicaps: Evidence of limited effectiveness. *Journal of Developmental and Physical Disabilities, 3*, 157–179. https://doi.org/10.1007/bf01045931

Cosbey, J., & Muldoon, D. (2017). EAT-UP™ Family-centered feeding intervention to promote food acceptance and decrease challenging behaviors: A single-case experimental design replicated across three families of children with autism spectrum disorder. *Journal of Autism & Development Disorders, 47*, 564–578. https://doi.org/10.1007/s10803-016-2977-0

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. https://doi.org/10.1111/j.1469-8749.2003.tb00928.x

Crnic, K. A., & Greenberg, M. T. (1990). Minor parenting stresses with young children. *Child Development, 61*, 1628–1637. https://doi.org/10.2307/1130770

Curtin, C., Hubbard, K., Anderson, S. E., Mick, E., Must, A., & Bandini, L. G. (2015). Food selectivity, mealtime behavior problems, spousal stress, and family food choices in children with and without autism spectrum disorder. *Journal of Autism and Developmental Disorders, 45*, 3308–3315. https://doi.org/10.1007/s10803-015-2490-x

Dawson, J. E., Piazza, C. C., Sevin, B. M., Gulotta, C. S., Lerman, D., & Kelley, M. (2003). Use of the high-probability instructional sequence and escape extinction in a child with a feeding disorder. *Journal of Applied Behavior Analysis, 36*, 105–108. https://doi.org/10.1901/jaba.2003.36-105

DeLeon, I. G., & Iwata, B. A. (1996). Evaluation of a multiple stimulus presentation format for assessing reinforcer preferences. *Journal of Applied Behavior Analysis, 29*, 519–533. https://doi.org/10.1901/jaba.1996.29-519

Dempsey, J., Piazza, C. C., Groff, R. A., & Kozisek, J. M. (2011). A flipped spoon and chin prompt to increase mouth clean. *Journal of Applied Behavior Analysis, 44*, 961–965. https://doi.org/10.1901/jaba.2011.44-961

Didehbani, N., Kelly, K., Austin, L., & Wiechmann, A. (2011). Role of parental stress on pediatric feeding disorders. *Children's Health Care, 40*(2), 85–100. https://doi.org/10.1080/02739615.2011.564557

Downey, G., & Coyne, J. C. (1990). Children of depressed parents: An integrative review. *Psychology Bulletin, 108*, 50–76. https://doi.org/10.1037/0033-2909.108.1.50

English, C. L., & Anderson, C. M. (2004). Effects of familiar versus unfamiliar therapists on responding in the analog functional analysis. *Research in Developmental Disabilities, 25*, 39–55. https://doi.org/10.1016/j.ridd.2003.04.002

Feldman, R., Keren, M., Gross-Rozval, O., & Tyano, S. (2004). Mother–child touch patterns in infant feeding disorders: Relation to maternal, child, and environmental factors. *Journal of the American Academy of Child and Adolescent Psychiatry, 43*, 1089–1097. https://doi.org/10.1097/01.chi.0000132810.98922.83

Fernand, J. K., Penrod, B., Fu, S. B., Whelen, C. M., & Medved, S. (2016). The effects of choice between non-preferred foods on the food consumption of individuals with food selectivity. *Behavioral Interventions, 31*, 87–101. https://doi.org/10.1002/bin.1423

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. https://doi.org/10.1901/jaba.1992.25-491

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*, 447–457. https://doi.org/10.1901/jaba.1994.27-447

Freeman, K. A., & Piazza, C. C. (1998). Combining stimulus fading, reinforcement, and extinction to treat food refusal. *Journal of Applied Behavior Analysis, 31*, 691–694. https://doi.org/10.1901/jaba.1998.31-691

Fu, S. B., Penrod, B., Fernand, J. K., Whelan, C. M., Griffith, K., & Medved, S. (2015). The effects of modeling contingencies in the treatment of food selectivity in children with autism. *Behavior Modification, 39*, 771–784. https://doi.org/10.1177/0145445515592639

Galensky, T. L., Miltenberger, R. G., Stricker, J. M., & Garlinghouse, M. A. (2001). Functional assessment and treatment of mealtime behavior problems. *Journal of Positive Behavior Interventions, 3*, 211–224. https://doi.org/10.1177/109830070100300403

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

Greer, R. D., Dorow, L., Williams, G., McCorkle, N., & Asnes, R. (1991). Peer-Mediated procedures to induce swallowing and food acceptance in young children. *Journal of Applied Behavior Analysis, 24*, 783–790. https://doi.org/10.1901/jaba.1991.24-783

Hodges, A., Gerow, S., Davis, T. N., Radhakrishnan, S., Feind, A., O'Guinn, N., & Prawira, C. (2018). An initial evaluation of trial-based functional analyses of inappropriate mealtime behavior. *Journal of Developmental and Physical Disabilities, 30*, 391–408. https://doi.org/10.1007/s10882-018-9592-2

Horne, P. J., Greenhalgh, J., Erjavec, M., Lowe, C. F., Viktor, S., & Whitaker, C. J. (2011). Increasing preschool children's consumption of fruit and vegetables. A modelling and rewards intervention. *Appetite, 56*, 375–385. https://doi.org/10.1016/j.appet.2010.11.146

Iwata, B. A., Dorsey, M. F., Slifer, K. J., Bauman, K. E., & Richmand, 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). https://doi.org/10.1901/jaba.1994.27-197

Jenkins, S. R., LeBlanc, L. A., & Lambert, G. P. (2017). Treating food approval-seeking behavior: One bite at a time. *Behavior Analysis in Practice, 10*, 411–416. https://doi.org/10.1007/s40617-016-0170-8

Kerwin, M. E., Ahearn, W. H., Eicher, P. S., & Burd, D. M. (1995). The cost of eating: A behavioral economic analysis of food refusal. *Journal of Applied Behavior Analysis, 28*, 245–260. https://doi.org/10.1901/jaba.1995.28-245

Kunkel, K. R., Kozlowski, A. M., Taylor, T., & González, M. L. (2018). Validating a food avoidance assessment for children with food selectivity. *Behavioral Development, 23*(2), 89–105. https://doi.org/10.1037/bdb0000078

Lambert, J. M., Bloom, S. E., Clay, C. J., Kunnavatana, S. S., & Collins, S. D. (2014). Training residential staff and supervisors to conduct traditional functional analyses. *Research in Developmental Disabilities, 35*, 1757–1765. https://doi.org/10.1016/j.ridd.2014.02.014

Lambert, J. M., Bloom, S. E., Kunnavatana, S. S., Collins, S. D., & Clay, C. J. (2013). Training residential treatment staff to conduct trial-based functional analyses. *Journal of Applied Behavior Analysis, 46*, 296–300. https://doi.org/10.1002/jaba.17

Levin, L., & Carr, E. G. (2001). Food selectivity and problem behavior in children with developmental disabilities: Analysis and intervention. *Behavior Modification, 25*, 443–470. https://doi.org/10.1177/0145445501253004

McKay, J. M., Pickens, J., & Stewart, A. L. (1996). Inventoried and observed stress in parent- child interactions. *Current Psychology: Developmental, Learning, Personality, Social, 15*, 223–234. https://doi.org/10.1007/bf02686879

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

Mueller, M. M., Piazza, C. C., Moore, J. W., & Kelley, M. E. (2003). Training parents to implement pediatric feeding protocols. *Journal of Applied Behavior Analysis, 36*, 545–562. https://doi.org/10.1901/jaba.2003.36-545

Najdowski, A. C., Wallace, M. D., Doney, J. K., & Ghezzi, P. M. (2003). Parental assessment and treatment of food selectivity in natural settings. *Journal of Applied Behavior Analysis, 36*, 383–386. https://doi.org/10.1901/jaba.2003.36-383

Najdowski, A. C., Wallace, M. D., Penrod, B., Jonathan 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

Pace, G. M., Ivancic, M. T., Edwards, G. L., Iwata, B. A., & Page, T. J. (1985). Assessment of stimulus preference and reinforcer value with profoundly retarded individuals. *Journal of Applied Behavior Analysis, 18*, 249–255. https://doi.org/10.1901/jaba.1985.18-249

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. https://doi.org/10.1007/s40617-016-0143-y

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., Kelly, M. L., Ochsner, C. A., & Santana, C. M. (2001). Using a fading procedure to increase fluid consumption in a child with feeding problems. *Journal of Applied Behavior Analysis, 34*, 357–360. https://doi.org/10.1901/jaba.2001.34-357

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. https://doi.org/10.1016/j.ridd.2005.05.005

Penrod, B., Gardella, L., & Fernand, J. (2012). An evaluation of a progressive high-probability instructional sequence combined with low-probability demand fading in the treatment of food selectivity. *Journal of Applied Behavior Analysis, 45*, 527–537. https://doi.org/10.1901/jaba.2012.45-527

Penrod, B., & Van Dalen, K. H. (2010). An evaluation of emerging preference for non-preferred foods targeted in the treatment of food selectivity. *Behavioral Interventions, 25*, 239–251. https://doi.org/10.1002/bin.306

Penrod, B., Wallace, M. D., Reagon, K., Betz, A., & Higbee, T. S. (2010). A component analysis of a parent-conducted multi-component treatment for food selectivity. *Behavioral Interventions, 25*, 207–228. https://doi.org/10.1002/bin.307

Peterson, K. M., Volkert, V. M., & Zeleny, J. R. (2015). Increasing self-drinking for children with feeding disorders. *Journal of Applied Behavior Analysis, 48*, 436–441. https://doi.org/10.1002/jaba.210

Piazza, C. C. (2008). Feeding disorders and behavior: What have we learned? *Developmental Disabilities, 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 behaviors. *Journal of Applied Behavior Analysis, 36*, 187–204. https://doi.org/10.1901/jaba.2003.36-187

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 nonpreferred food to treat food selectivity. *Journal of Applied Behavior Analysis, 35*, 259–270. https://doi.org/10.1901/jaba.2002.35-259

Pizzo, B., Coyle, M., Seiverling, L., & Williams, K. (2012). Plate A–plate B: Use of sequential presentation in the treatment of food selectivity. *Behavioral Interventions, 27*, 175–184. https://doi.org/10.1002/bin.1347

Pizzo, B., Williams, K. E., Paul, C., & Riegel, K. (2009). Jump start exit criterion: Exploring a new model of service delivery for the treatment of childhood feeding problems. *Behavioral Interventions, 24*, 195–203. https://doi.org/10.1002/bin

Polan, H., & Ward, M. (1994). Role of the mother's touch in failure to thrive: A preliminary investigation. *Journal of the American Academy of Child and Adolescent Psychiatry, 33*, 1090–1105. https://doi.org/10.1097/00004583-199410000-00005

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. https://doi.org/10.1901/jaba.2004.37-27

Reitman, D., & Passeri, C. (2008). Use of stimulus fading and functional assessment to treat pill refusal with an 8-year-old boy diagnosed with ADHD. *Clinical Case Studies, 7*, 224–237. https://doi.org/10.1177/1534650107307476

Ringdahl, J. E., & Sellers, J. A. (2000). The effects of different adults as therapists during functional analyses. *Journal of Applied Behavior Analysis, 33*, 247–250. https://doi.org/10.1901/jaba.2000.33-247

Rispoli, M., Burke, M. D., Hatton, H., Ninci, J., Zaini, S., & Sanchez, L. (2015). Training head start teachers to conduct trial-based functional analysis of challenging behavior. *Journal of Positive Behavior Interventions, 17*, 235–244. https://doi.org/10.1177/1098300715577428

Rispoli, M., Neely, L., Healy, O., & Gregori, E. (2016). Training public school special educators to implement two functional analysis models. *Journal of Behavioral Education, 25*, 249–274. https://doi.org/10.1007/s10864-016-9247-2

Rivas, K. D., Piazza, C. C., Kadey, H. J., Volkert, V. M., & Stewart, V. (2011). Sequential treatment of a feeding problem using a pacifier and flipped spoon. *Journal of Applied Behavior Analysis, 44*, 387–391. https://doi.org/10.1901/jaba.2011.44-387

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

Sasaki, A. M., & Fryling, M. J. (2014). Cup distance fading to decrease inappropriate behavior in a child with autism. *Journal of Developmental and Physical Disabilities, 26*, 507–512. https://doi.org/10.1007/s10882-013-9355-z

Seiverling, L., Harclerode, W., & Williams, K. (2014). The effects of a modified treatment package with and without feeder modeling on one child's acceptance of novel foods. *Education and Treatment of Children, 37*, 477–494. https://doi.org/10.1353/etc.2014.0023

Seubert, C., Fryling, M. J., Wallance, 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

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. https://doi.org/10.1007/s10567-010-0079-7

Sharp, W. G., Trumbull, A., & Lesack, R. (2015). Blending to treat expulsion in a child with food refusal. *Behavioral Interventions, 30*, 247–255. https://doi.org/10.1002/bin.1413

Silbaugh, B. C., Penrod, B., Whelan, 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. *Review Journal of Autism and Developmental Disorders, 3*, 345–357. https://doi.org/10.1007/s40489-016-0087-8

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

Sira, B. K., & Fryling, M. J. (2012). Using peer modeling and differential reinforcement in the treatment of food selectivity. *Education & Treatment of Children, 35*, 91–100. https://doi.org/10.11353/etc.2012.0003

Spagnola, M., & Fiese, B. H. (2007). Family routines and rituals: A context for development in the lives of young children. *Infants & Young Children, 20*, 284–299. https://doi.org/10.1097/01.iyc.0000290352.32170.5a

Taylor, C. M., Wernimont, S. M., Northstone, K., & Emmet, P. M. (2015). Picky/fussy eating in children: Review of definitions, assessment, prevalence, and dietary intakes. *Appetite, 95*, 349–359. https://doi.org/10.1016/j.appet.2015.07.026

Van Dalen, K. H., & Penrod, B. (2010). A comparison of simultaneous versus sequential presentation of novel foods in the treatment of food selectivity. *Behavioral Interventions, 25*, 191–206. https://doi.org/10.1002/bin.310

Vaz, P. C. M., Volkert, V. M., & Piazza, C. C. (2011). Using negative reinforcement to increase self-feeding in a child with food selectivity. *Journal of Applied Behavior Analysis, 44*, 915–920. https://doi.org/10.1901/jaba.2011.44-915

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., & Price, R. R. (2016). Further manipulations in response effort or magnitude of an aversive consequence to increase self-feeding in children with feeding disorders. *Behavior Analysis in Practice, 9*, 103–113. https://doi.org/10.1007/s40617-016-0124-1

Whelan, C. M., & Penrod, B. (2018). An evaluation of sequential meal presentation with picky eaters. *Behavior Analysis in Practice*. Online first. https://doi.org/10.1007/s40617-018-00277-7

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. https://doi.org/10.1901/jaba.2005.132-04

Wolf, M. M. (1978). Social validity: The case for subjective measurement or how applied behavior analysis is finding its heart. *Journal of Applied Behavior Analysis, 11*, 203–214. https://doi.org/10.1901/jaba.1978.11-203

Woods, J. N., Borrero, J. C., Laud, R. B., & Borrero, C. S. W. (2010). Descriptive analyses of pediatric food refusal: The structure of parental attention. *Behavior Modification, 34*, 35–56. https://doi.org/10.1177/0145445509355646

Zarcone, J. R., Crosland, K., Fisher, W. W., Worsdell, A. S., & Herman, K. (1999). A brief method for conducting a negative-reinforcement assessment. *Research in Developmental Disabilities, 20*, 107–124. https://doi.org/10.1016/S0891-4222(98)00036-5

36

The Treatment of Dually Diagnosed Individuals with Sleep Disturbances and Intellectual Disabilities

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It is well-established that sleep disturbances are common in persons with intellectual disabilities. Caregivers report frequent sleep-related problems in both children and adults with intellectual disabilities and sleep assessments document this increased prevalence. Persons with intellectual disabilities are not only more likely to experience disturbed sleep, these disturbances are more frequent, more persistent, and more severe than those in the general population (Grigg-Damberger and Rawls, 2013; Stores, 2014). In addition, sleep disturbances increase the economic and emotional burdens on families (Heussler, 2016). Despite the well-documented need for therapies targeting sleep disturbances, research offers limited guidance for the treatment of persons with intellectual disabilities (Beresford et al., 2018; Buckles, Luckasson, & Keefe, 2013; Stores, 2014). To meet these challenges, clinicians should be well versed in the treatment of sleep disturbances and promote research to meet the needs of persons dually diagnosed with intellectual disabilities and sleep disturbances. This chapter will review the treatments for sleep disturbances encountered by mental health professionals and highlight the application of general treatment principles for persons with intellectual disabilities with a focus on clinical interventions for the mental health professional.

Clinical Vignette – The Treatment of Paul's Sleep Disturbance

Paul, a thirty-seven-year-old man with Down syndrome and moderate to severe intellectual disability, is referred to you for the assessment of sleep disturbances. Alexis, a group home staff, reports that he is waking during the night and roaming about the home. Recently he has been loud and has woken housemates. Alexis recalls that about 3 months ago Paul started having difficulty settling at bedtime and did not want to wear the continuous positive airway pressure (CPAP) mask that treats his sleep apnea. He has also been more irritable during the day and is beginning to make mistakes at the grocery store job he has held for 10 years.

A review of Paul's clinical file shows that he received the Alzheimer's Association–recommended baseline assessment of adult functioning 5 years ago. Reassessment including the Dementia Scale for Down syndrome (Gedye, 1995) and the Diagnostic Assessment for the Severely Handicapped-II (Sturmey, Matson,

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& Lott, 2004) does not indicate a decline in cognitive function or mental illness. With early onset dementia ruled out, Paul was referred for a medical evaluation. His electrocardiogram was unchanged and laboratory testing was normal. The primary care provider explains the health risks associated with Down syndrome and the contribution of diet and exercise to healthy sleep. Paul was referred to a dietician to address obesity and a physical therapist to guide a gradual increase in daily exercise. A referral was made to a sleep specialist to reassess his sleep apnea. Following a sleep study, the sleep specialist noted that REM percentage of total sleep time, a marker for dementia, was unchanged but made minor adjustments to Paul's CPAP settings to better treat his sleep apnea. The sleep specialist recommended behavioral interventions to address Paul's behavioral insomnia.

Before implementing the treatment for behavioral insomnia, you ask Paul and Alexis to complete a sleep diary for 2 weeks and an Epworth Pictorial Sleepiness Scale (EPSS) (Ghiassi, Murphy, Cummin, & Partridge, 2011). After reviewing Paul's daily activities, the EPSS was modified, replacing sleepiness during reading with watching others play a video game, a common activity for Paul. You explain the importance of completing the sleep diary each day and you ask Alexis to add notes to the diary regarding illness, challenging behaviors, and compliance with CPAP. You explain that sleep disturbances are correlated with behavioral difficulties, numerous health issues, and dementia. You thank Alexis for recognizing the importance of Paul's sleep health and bringing it to your attention. You schedule a follow-up appointment to address good sleep hygiene and review the sleep diary.

Models of Insomnia

The word "insomnia" is used broadly to indicate difficulty sleeping. Insomnia, whether referring to the diagnostic category, insomnia disorder, or behavioral insomnia, is the most common sleep disturbance in individuals with intellectual disabilities and the general population (Barnhill & Hollway, 2016). Sleep health is critical to physical and mental health. Poor sleep health contributes to disability from illness including cardiac, neurological, metabolic syndromes like diabetes, and most mental disorders (Institute of Medicine Committee on Sleep Medicine, 2006). Difficulties settling, excessive daytime sleepiness, night arousals, and other sleep disturbances may be early signs of a neurodegenerative disease, with disturbed sleep typically occurring prior to overt cognitive decline (Kim & Duffy, 2018). Sleep disturbances in adults with neurodevelopmental disorders warrants screening for Alzheimer's or other causes of dementia as persons with intellectual disabilities are reported to have equal or higher prevalence of dementia compared to the general population. The genetic predisposition to neurodegeneration in persons with Down syndrome which leads to Alzheimer's disease is well-documented (Mann et al., 2018).

While behavioral insomnia is not a DSM-5 diagnosis, it a common presenting complaint in clinical practice. Behavioral causes of insomnia include bedtime resistance due to inability to calm, anxiety, fears, or other factors (Esbensen & Schwichtenberg, 2016). Behavioral challenges are the most frequent sleep disturbance expressed by the caregivers of children with intellectual disability and are often chronic (Köse, Yılmaz, Ocakoğlu, & Özbaran, 2017). Poor settling in children may shift to delayed sleep or sleep anxiety with age (Deliens, Leproult, Schmitz, Destrebecqz, & Peigneux, 2015). Challenges may be precipitated by environmental factors such as the use of television or electronic equipment at bedtime as well as lights, noise, and temperature variations which may be especially disturbing to persons with sensory issues. Blue spectrum lighting, found in electronics, LEDs, and compact fluorescent bulbs, increases alertness and can disrupt circadian rhythms (Tosini, Ferguson, & Tsubota, 2016). Poor sleep health should be treated as it may increase the disability of mental and intellectual disorders (Tolaymat & Liu, 2017).

In the absence of biomarkers, interventions for insomnia have been guided by theoretical models. Numerous models have informed the

leading therapy for insomnia – the cognitive behavioral therapy for insomnia (CBT-I). The multicomponent CBT-I which is cited by the American Academy of Sleep Medicine and other authorities in the field as the first-line treatment for insomnia represents a variety of evidence-based interventions (Manber et al., 2012; Morgenthaler et al., 2006) (See Table 36.1). The strong behavioral focus of early models including the stimulus control model and three-factor model led to the most effective CBT-I treatments-stimulus control, and sleep restriction therapies. Treatment-oriented models with a strong cognitive focus, including Morin's microanalytic and Harvey's cognitive models, have informed components of CBT-I which challenge the thoughts and behaviors perpetuating insomnia. More biologically oriented models including the two-factor and neurobiological models incorporate neurophysiological and neural circuit research. Two integrated models of insomnia have recently been proposed – the pathophysiology of insomnia and the parallel process model.

Bootzin (1972) introduced the stimulus control model that promotes the concept of good stimulus control through strengthening the association between the bedroom and sleep. In this model, gaming, telephone, television, eating, and other distractions have no place in the bedroom as they promote stimulus dyscontrol. Eliminating factors that contribute to a complex conditioning history interfering with sleep allows a greater probability that the stimulus (bedroom) will elicit the desired response (sleep). Stimulus control therapy seeks to reduce internal and/or external cues for arousal and establish a strong discriminative stimuli for sleep (Bootzin, Epstein, & Wood, 1991; Bootzin & Perlis, 2011).

Spielman's three-factor model, or 3P model, describes insomnia via predisposing, precipitating and perpetuating factors. Predisposing and precipitating components characterize the stress-diathesis dynamic underlying the onset of insomnia which continues due to perpetuating factors which occur as a reaction to sleeplessness (Spielman, Caruso, & Glovinsky, 1987). It is well-documented that persons with intellectual disabilities are at increased risk for numerous factors that increase the risk of sleep disturbances including insomnia (Esbensen, 2016; Jang et al., 2013; Richdale & Baker, 2014; Stores, 2016). Table 36.2 summarizes predisposing and precipitating factors for persons with intellectual disabilities with an expanded conceptualization of perpetuating factors, beyond the traditional patient compensatory behaviors. The 3P model

Table 36.1 Common interventions for insomnia

Intervention	Description
Sleep hygiene	Sleep health education and instruction for patients and caregivers focusing on personal and environmental changes to improve sleep.
Stimulus control therapy	Promotes a healthy association between the bedroom and sleep to encourage a regular sleep-wake schedule and extinguish conditioned responses promoting poor sleep. Stimulus fading, also called graduated extinction, may be used to gradually decrease the reliance of young children on a caregiver's presence to fall asleep.
Relaxation therapy	An array of interventions designed to promote sleep by decreasing physical and cognitive arousal.
Cognitive therapy	Addresses cognitive distortions of patients and/or caregivers to establish realistic expectations about sleep.
Sleep restriction therapy	Improves the sleep drive by promoting improved sleep continuity. Sleep time is first restricted based on baseline sleep logs to encourage sleep consolidation, then gradually increased to the recommended sleep time. Variations include a faded bedtime, faded bedtime with response cost, and scheduled awakenings.
Multicomponent therapy without CBT	A multimodal approach combining sleep hygiene and behavioral therapies.
Paradoxical intention	A cognitive intervention used to decrease sleep anxiety by instructing the patient to gain control over sleep by staying awake.
Biofeedback	Decreases physical arousal at bedtime through feedback training to control a physiological response.

Stimulus Control Model
Three-Factor Model
Microanalytic Model
Neurocognitive Model
Two-Factor Model
Sleep Interfering-Interpreting Model
Psychobiologic Inhibition Model
Cognitive Model
Neurobiologic Model
Parallel Process Model
Triple-R Model

Fig. 36.1 Theoretical Models of Insomnia

Table 36.2 Factors of Spielman's 3P model common in persons with intellectual fisabilities

Predisposing	Precipitating	Perpetuating
Genetic factors	Acute medical conditions	Untreated medical conditions
Epigenetic factors	Pain/discomfort	Sensory issues
Neurophysiological factors	Environmental factors	Environmental factors
Medical conditions	Stress/trauma	Caregiver under identification
Mental disorders		Clinician under diagnosis
Obesity		Diagnostic overshadowing
Age-related factors		
Sensory issues		

Adapted from McCurry, Song, and Martin (2015)

was later expanded to recognize the contribution of Pavlovian conditioning as a perpetuating factor and to include a temporal component, which recognizes that all three factors may vary over time (Perlis, Ellis, Kloss, & Riemann, 2017). The three-factor model informs the treatment of insomnia through sleep restriction therapy, which first aims to increase sleep continuity by limiting sleep to an allotted time based on baseline sleep diaries, and then gradually increasing sleep time (Spielman, Saskin, & Thorpy, 1987). Sleep restriction therapy (SRT) is a well-established component of CBT-I and multicomponent therapies for insomnia. Well-researched measurable components of SRT include sleep window generation, minimum time-in-bed, sleep efficiency titration criteria, sleep window modification, and positioning of the sleep window; however, there is no uniformity among studies in reporting on these measures. Kyle et al. (2015) proposed a standardized method for reporting SRT parameters which were further refined by Maurer, Espie, and Kyle (2018) as the SRT Triple-R model. The Triple-R model of SRT builds on Spielman's work, incorporating advances in sleep science into a paradigm facilitating systematic study to promote the understanding of the mechanism of action of SRT (Maurer et al., 2018).

The microanalytic model hypothesizes a multidirectional relationship between dysfunctional cognition, arousal, maladaptive habits, and the consequences of impaired sleep as factors which must be addressed for the successful treatment of insomnia (Morin, 1996). This multicomponent treatment approach was detailed in Morin's 1993 *Insomnia: Psychological Assessment and Management*, the first treatment manual for insomnia.

Harvey's cognitive model of insomnia is grounded in the cognitive models of anxiety dis-

orders. As such, the cognitive model proposes that distorted perceptions and beliefs, selective attention and monitoring, and safety behaviors fuel negatively toned cognitions regarding sleep, leading to insomnia (Harvey, 2002). These cognitions may take the form of worry, rumination, or catastrophizing (Harvey & Tang, 2012; Hiller, Lovato, Gradisar, Oliver, & Slater, 2014). A strength of Harvey's model is that the elements can be measured with instruments including the Daytime Insomnia Symptom Response Scale, the Dysfunctional Beliefs and Attitudes About Sleep, the Sleep Related Behaviors Questionnaire, and the Stroop Color Task (Hiller, Johnston, Dohnt, Lovato, & Gradisar, 2015). [See Chap. 23 for a brief overview of sleep science.]

The two-factor model of sleep describes the interaction of process C, the circadian process, and process S, the neurochemical physiological or homeostatic process. Process S regulates sleep intensity while process C regulates the timing of sleep. These processes are interrelated and may be intentionally overridden. When Process S is at a minimum, it triggers awakening; at its maximum level it triggers sleep, if Process C is in the correct circadian phase (Borbely, Daan, Wirz-Justice, & Deboer, 2016; Schwartz & Kilduff, 2015).

The neurobiological model of Buysse and colleagues incorporates neuroscience research regarding neurophysiology and brain circuitry to explain sleep/wake functions in normal sleep, insomnia, and the behavioral and pharmacological treatments of insomnia (Buysse, Germain, Hall, Monk, & Nofzinger, 2011). This model highlights central sleep regulatory systems (specific brain regions) which are triggered by local (neuronal) processes. [See chap. 23 for an overview of sleep science.] The neurobiological model has been used to investigate patient reports of poor sleep that cannot be documented by polysomnogram called subjective-objective sleep discrepancies. The activation of brain regions responsible for self-awareness during NREM sleep may contribute to insomnia promoting cognitions which can be treated with CBT-I and/or medication (Kay et al., 2017).

Levenson, Kay, and Buysse (2015) have offered the pathophysiology of insomnia model, which assimilates behavioral, cognitive, and neurobiological sleep data using the research domain criteria (RDoC) taxonomy with hyperarousal as the key factor. The pathophysiology of insomnia model specifically highlights genetic vulnerability and abnormalities in neurobiological processes, critical elements for understanding insomnia in persons with intellectual disabilities. The hyperarousal focus of this model has been a highlighted in the study of sleep disturbances in persons with autism (Souders et al., 2017). The parallel process model illustrates how the cognitive and behavioral spheres are parallel to the neurocognitive and neurobiological spheres (Perlis et al., 2017).

Clinical Vignette – Paul's Baseline Data

Paul and Alexis bring the completed sleep dairy and EPSS to his follow-up appointment (See Table 36.3). When reviewing the two-week sleep diary, you notice intraindividual sleep/wake variability. Paul goes to sleep faster on Tuesdays, Thursdays, Saturday, and Sunday and wakes easily the following morning. Awakening/roaming only occurred twice on these nights. Other days he has long sleep onset latency, night awakening with roaming, and is difficult to wake the following morning. The EPSS notes mild excessive daytime sleepiness on Tuesdays and Thursdays with higher than normal daytime sleepiness other days. Notes on the sleep dairy say Paul tried to nap on a couple of occasions on weekends and nocturnal enuresis has occurred twice. Alexis tells you that staff have engaged him in a walk to avoid napping. Paul volunteers that he works on Monday, Wednesday, and Friday and likes to nap in the van on the way home. You discuss good sleep hygiene and a medical evaluation for enuresis with Paul and Alexis. Alexis was aware that caffeine and napping could impact sleep health but was surprised to learn that napping can be beneficial for some but may cause confusion in persons with Down syndrome. She took notes as

Table 36.3 Sleep diary parameters

Parameter	Definition	Parameter goal	Paul's sleep dairy		
			Baseline	Follow-up	Final
Bedtime (BT)	Time person gets in bed	Varies by individual and age	10 pm	10 pm	10 pm
Lights out (OT)	Time light is turned out	Ideally, same as BT	10 pm	10 pm	10 pm
Sleep onset (SO)	Time person is asleep	Ideally, same each night	Varies	10:20 pm	10:15 pm
Sleep latency (SL)	BT minus SO	Less than 30 minutes/night	Varies	20 minutes	15 minutes
Number of awakenings/duration	Time and length of each awake time	Ideally, 1–2× or less, each less than 5 minutes	4×/week	Week 1 3× Week 2 4×	Once in 2 weeks
Wake after sleep onset (WASO)	Sum of wake times from SO to WT	Less than 30 minutes/night	1–2 hours	1.5 hours	10 minutes
Total wake time (TWT)	SL + WASO	Less than 30 minutes/night	Varies	110 minutes	25 minutes
Wake time (WT)	Time person rises for the day	Ideally, same time each morning	7 am	7 am	7 am
Total sleep time (TST)	TIB − (SL + WASO)	More than 6.5 hours	Varies	7.2 hours	8.6 hours
Total time spent in bed (TIB)	BT to WT	Ideally, near or equals TST	9 hours	9 hours	9 hours
Sleep efficiency (SE)	TST divided by TIB times 100	More than 85%	Varies	80%	95%
Nap time/frequency/duration	Daytime sleeping	Varies by age	3 days/week	None	None

you explained how other lifestyle factors and environmental factors like temperature, exercise, and relaxation impact Paul's sleep. Paul lets you know that he cannot sleep if he is hot. Alexis thanks you for helping her understand sleep hygiene and agrees to help Paul avoid napping and to investigate environmental and lifestyle factors that might impact Paul's sleep. She invites you to the group home's monthly staff meeting to discuss sleep health and good sleep hygiene practices.

Collecting Baseline Treatment Data

Reviewing medical history, interviewing the patient and caregivers, and collecting baseline sleep information before starting treatment are critical in order to target interventions effectively and monitor progress. Numerous questionnaires are designed to screen and inform diagnostic decisions and treatment planning but cannot replace a sleep study for the diagnosis and management of sleep apnea. [See Chap. 23 for a discussion of sleep assessment.] Because few instruments have been validated for persons with intellectual disabilities, modifications to existing instruments reflecting the abilities, activities, and medical issues of an individual client may be helpful, for example, noting seizure activity in a sleep diary for persons with a seizure disorder. Seizures occur in over 20% of persons with intellectual disabilities, with a higher prevalence among the aging and those with more severe disability (Robertson, Hatton, Emerson, & Baines, 2015). Poorly controlled epilepsy is associated with daytime sleepiness and alterations in sleep architecture, and may mimic a sleep disorder (Unterberger et al., 2015). Because reciprocal relationships exist between intellectual disabilities and sleep disturbances and medical/mental disorders, the later must be monitored during the treatment of sleep disturbances (Adams, Matson, Cervantes, & Goldin, 2014; Schutte-Rodin, Broch, Buysse, Dorsey, & Sateia, 2008).

Treatment for sleep disturbances in non-ID populations typically occurs when daytime sleepiness interferes with daily activities. In contrast, persons with intellectual disabilities are typically referred for treatment when behavioral disturbances challenge others. It is well-documented that the sleep disturbances of persons with intellectual disabilities impact the health and daytime performance and often disrupts the sleep of other family members (Micsinszki, Ballantyne, Cleverley, Green, & Stremler, 2018; Simard-Tremblay, Constantin, Gruber, Brouillette, & Shevell, 2011; Wayte, McCaughey, Holley, Annaz, & Hill, 2012). In a study of persons with dementia and their caregivers, caregivers had clinically significant sleep disturbances which improved dramatically when the client was placed outside the home (Lee, Morgan, & Lindesay, 2007). McCurry et al. (2015) have adapted Spielman's 3P model to capture factors predisposing, precipitating, and perpetuating insomnia in caregivers. Because caregiver health is critical to the well-being of persons with disabilities, the American Psychological Association provides a *Family Caregiver Briefcase* at https://www.apa.org/pi/about/publications/caregivers/index.aspx. In planning for treatment, the clinician must address the sleep health of the client as well as caregiver concerns, keeping in mind that there may be multiple caregivers and care settings. Acknowledging caregiver concerns and promoting the caregiver's perception of competence may lessen the burden of implementing therapeutic recommendations and improve compliance (Mol, Monbaliu, Ven, Vergote, & Prinzie, 2012). Successful treatment planning and implementation necessitates including clients, caregivers, and all treatment providers in the process.

Successful behavioral interventions begin with characterizing baseline sleep in a sleep diary. A sleep diary captures bedtime, sleep onset latency, total sleep time, total wake time, and wake after sleep onset as well as daytime napping (See Table 36.3). The mean of these variables as well as intraindividual variability should be considered (Bei, Wiley, Trinder, & Manber, 2016). Intraindividual variability in sleep-wake patterns are the changes that occur from night to night. Intraindividual variability is common in young children and persons with neurodevelopmental disorders. It may also be influenced by socioeco-

nomic status, race, health, cultural, and environmental factors (Becker, Sidol, Van Dyk, Epstein, & Beebe, 2017). For persons with intellectual disability, particularly in group settings, bedtime may be imposed rather than a choice and may not align with an individual's natural circadian rhythm. A careful history may help identify night owls or morning larks as shifting the natural body clock can be difficult. Sleep onset latency is the time between bedtime and sleep onset. While an ideal value is not defined, a sleep onset latency of less than 30 minutes is considered normal (Schutte-Rodin et al., 2008). Recommended total sleep times vary by age with the National Sleep Foundation recommending 14–17 hours for newborns, 12–15 hours for infants, 11–14 hours for toddlers, and between 10 and 13 hours for preschoolers including daytime sleeping. By school-age, recommended sleep duration is 9–11 hours for school-aged children, 8–10 hours for teenagers, 7–9 hours for young adults and adults, and 7–8 hours for older adults (Hirshkowitz et al., 2015). Waketimes after sleep onset should be brief and typically reflect arousal during light sleep or due to toileting needs. Difficulty returning to sleep after night wakening is a common concern reported by parents. When consulting with group settings on sleep issues, staff should be queried about toileting schedules that may include staff rousing residents from sleep to prevent toileting accidents. Ideally, total time in bed should be near the total sleep time. When total time in bed significantly exceeds total sleep time, forced times to go to bed and rise may be a concern. Periods of napping should be recorded including the time of day, duration, and situations where drowsiness or napping occurs.

Napping is typical of young children and is common with aging (Cooke, Delalot, & Werner, 2015; Staton, Smith, Hurst, Pattinson, & Thorpe, 2017). Excessive daytime sleepiness is common in persons with some syndromes, such as Prader-Willi and Williams (Esbensen & Schwichtenberg, 2016; Richdale & Baker, 2014). When treating sleep disturbances, factors beyond insomnia contributing to daytime sleepiness should be considered. After ruling out medical conditions, acute illness, medications, and/or pain, behavioral reasons including escape, avoidance, boredom, and caregiver convenience should be considered. Numerous methods for assessing and monitoring sleepiness are available. Average sleep propensity, a measure of daytime sleepiness, can be assessed using an Epworth Sleepiness Scale (ESS). The ESS rates the tendency to fall asleep during common activities on a scale of 0–3 (Johns, 1991). The Karolinska Sleepiness Scale (KSS) captures subjective, situational sleepiness on a 10-point scale ranging from extremely alert to extremely sleepy (Åkerstedt & Gillberg, 1990). A pictorial version is available (Maldonado, Bentley, & Mitchell, 2004). Drowsiness may be measured directly using the Optalert® technology and the Johns Drowsiness Scale™ which are based on the speed of eyelid opening and closing during blinking (Johns, 1991; Johns et al., 2006).

Sleep Hygiene Education

Sleep hygiene education is the component of CBT-I that helps the patient and caregivers understand how sleep health is impacted by lifestyle and the environment. A systematic review and meta-analysis of 15 studies found sleep hygiene education was less effective than CBT-I and mindfulness interventions (Chung et al., 2018). Still, systematically addressing life style and environmental factors in residential facilities has been shown to benefit persons dually diagnosed with intellectual disabilities and sleep disturbances (McCurry, LaFazia, Pike, Logsdon, & Teri, 2012). While there is not a standardized approach to sleep hygiene education, recommendations from national organizations abound, including the Centers for Disease Control and Prevention, National Sleep Foundation, and Autism Speaks (Autism Speaks, 2018; CDC, 2016; NSF, 2018). School-based sleep education programs targeting sleep health in teens have highlighted the importance of student and teacher involvement, the use of engaging and interactive learning platforms, and assessing readiness for change (Blunden & Rigney, 2015). For persons with intellectual disabilities, caregiver involvement, multimodal teaching, and readiness for

change in the patient and caregiver should be considered. Commonly addressed lifestyle factors include diet, exercise, and regular sleep schedules. Environmental factors include lighting, noise, and temperature. Addressing these factors through sleep hygiene education is not a standalone intervention but complements other CBT-I modalities with recommendations the therapist targets to an individual patient's needs based on the sleep history (Irish, Kline, Gunn, Buysse, & Hall, 2015). McCurry et al., 2012 have reported that adult family home staff have high interest in learning sleep hygiene interventions and are able to successfully implement individualized resident behavioral sleep plans after four training sessions. Sleep hygiene education and behavioral interventions for sleep disturbances delivered in workshop and lecture formats to residential facility staff have been shown to improve sleep efficiency of persons with intellectual disabilities (Hylkema, Petitiaux, & Vlaskamp, 2011). Addressing lifestyle and environmental factors is critical in improving sleep health and quality of life.

Lifestyle Factors

The daily cycle of wakefulness and sleep is governed by internal and external elements in a complex interrelationship that can be enhanced with attention to diet, physical activity, and regular sleep-wake schedules. Persons with intellectual disabilities may have limited autonomy over these factors. They may also have less insight into the impact of these factors on sleep efficiency and sleep health. Communication challenges are common, requiring the clinician to communicate effectively with both the patient and the caregiver. Diagnostic overshadowing (attributing difficulties to intellectual disability without considering other causes) and time constraints in clinical settings may limit queries and education regarding lifestyle factors. These factors can contribute to health care disadvantages for persons with intellectual disability. Caregiver advocacy and participation should be encouraged as they can provide valuable assistance in relating the details of daily activities and assisting in the implementation of lifestyle changes. Additional research is needed to promote greater personalization of sleep lifestyle recommendations.

Lifestyle Factors – Diet

To understand the impact of diet on sleep, what is ingested and the timing of intake must be considered. Some substances including caffeine, alcohol, and nicotine are known to interfere with sleep. Caffeine is found in coffee, tea, energy drinks, many soft drinks, chocolate, and some medications. The impact of caffeine on sleep is regulated by adenosine A_{2A} receptors in the brain. The variant of the adenosine A_{2A} gene an individual possesses determines caffeine sensitivity or insensitivity (Rétey et al., 2007). While caffeine-insensitive individuals do not experience sleep disruption after consuming caffeine, sensitive individuals may experience difficulty falling asleep up to 6 hours after ingestion (Drake, Roehrs, Shambroom, & Roth, 2013). The effect may be mitigated by regular use; however, additional research is needed to fully understand caffeine metabolism in persons with intellectual disabilities (Irish et al., 2015). Sleep hygiene protocols typically recommend abstaining from caffeine 6–7 hours before bedtime.

Nicotine is a stimulant that disrupts sleep when ingested via traditional cigarettes, patches, vaping, pills, or as second-hand smoke. Nicotine impairs sleep efficiency with delay in sleep onset, shortened total sleep time, and poorer quality sleep which typically persists for weeks after smoking cessation (Cohrs et al., 2014; Jaehne et al., 2015). While care setting may impact nicotine use, adults with intellectual disabilities have reported to smoke at higher rates than the general population (Krahn, Walker, & Correa-De-Araujo, 2015). Unfortunately, they are less likely to be offered smoking cessation services (Cooper et al., 2018). The impact of indirect nicotine ingestion is similar to direct ingestion. A study of over 30,000 youth exposed to second-hand smoke found increased difficul-

ties with sleep onset, sleep maintenance, and a shortened total sleep time (Morioka et al., 2018). Sleep hygiene protocols recommend nicotine abstinence.

Alcohol has an initial sedating effect in non-habitual users; however, with chronic use sleep onset is delayed. Alcohol disrupts sleep architecture, resulting in an overall decrease in sleep quality. Sleep disturbances during withdrawal after prolonged use is nearly universal and may last for a year after abstinence in heavy users (Angarita, Emadi, Hodges, & Morgan, 2016). A review conducted by Carroll-Chapman and Wu (2012) found substance use, including alcohol, to be less prevalent among persons with intellectual disabilities, but, among those who do use, the risk of a substance use problem is significant and intervention is less likely. Limited research is available regarding the effects of alcohol on persons with intellectual disabilities; however, sleep hygiene education should include abstinence from alcohol including medications containing alcohol as some preparations contain up to 25% alcohol.

In addition to specific substances that are known to interfere with sleep, education should also address food quality and the timing of food intake and should be tailored to fit an individual's dietary needs. A diet high in fats has been associated with poorer sleep quality, changes in sleep architecture, and more arousals during sleep (St-Onge, Mikic, & Pietrolungo, 2016). For persons with gastrointestinal disturbances, fatty and spicy foods are discouraged, as is eating or drinking within 3 hours before bedtime. Conversely, hypoglycemia, low blood sugar, may cause night wakening. Persons with Cori's disease (glycogen storage disease type III) are prone to nighttime hypoglycemia as are persons with diabetes. For these individuals, a small snack before bedtime may be prescribed by the primary care provider (Kinsey & Ormsbee, 2015). The impact of diet, timing of intake, and the contribution of the gut microbiome are important areas for future research (Garcia, McPherson, Patel, & Burns, 2017).

Lifestyle Factors – Physical Activity

Regular physical activity promotes good health including good sleep health. Decreased sleep onset and subjective reports of improved sleep have been noted for persons with insomnia disorder and subthreshold insomnia who undertake a program of regular exercise (Lowe et al., 2018). While exercise has immediate and long-term benefits on sleep, the mechanisms are not fully understood, with limited research focusing on persons with intellectual disabilities. It is known that the acute effects of exercise include raising the core body temperature, endocrine effects on growth hormone and insulin, and decreasing the overall arousal of the autonomic nervous system. In addition to these effects, regular exercise decreases inflammation, helps regulate circadian rhythms, and improves symptoms of depression and anxiety (Chennaoui, Arnal, Sauvet, & Léger, 2015). The timing and intensity of activity should be considered when recommending exercise as part of a sleep health program. In the 2013 National Sleep Foundation Sleep in America Poll, 97% of respondents reported no change or improved sleep after evening exercise (defined as within 4 hours of bedtime) (Buman, Phillips, Youngstedt, Kline, & Hirshkowitz, 2014). Timing of exercise for children and persons with sleep disorders is less clear. It is generally recommended that vigorous exercise be avoided within 2 hours of bedtime to avoid increased arousal interfering with calming and sleep onset (Hill et al., 2008; Markwald, Iftikhar, & Youngstedt, 2018).

In a study of adults with mild–to-severe intellectual disability, only 10% met the World Health Organization recommendations for daily physical activity when measured using actigraphy (Oviedo, Travier, & Guerra-Balic, 2017). A sedentary, low-stimulation lifestyle contributes to poor sleep. Aerobic, resistance, and stretching exercises may be incorporated into a sleep treatment plan for insomnia (D'Aurea et al., 2018; Markwald et al., 2018). Aerobic exercises might include running, brisk walking, swimming, cycling, or dancing. Resistance exercises could include weight training, Pilates, resistance bands,

or calisthenics. Stretching exercises that improve balance and flexibility, such as yoga, are beneficial. While daily exercise should be encouraged, a goal of moderate exercise at least three times a week for 1 hour should be discussed with patients and caregivers. Because persons with intellectual disabilities engage in less physical activity and are at greater risk for obesity and heart disease, exercise should be introduced gradually after clearance from a primary care provider (Krahn et al., 2015).

Lifestyle Factors – Sleep-Wake Scheduling

Sleep-wake scheduling is addressed as a component of sleep hygiene education, with recommendations for set sleep and wake times and calming or low stimulation activities before bed. Regular sleep and wake times are associated with improved synchronization between circadian rhythm and nighttime sleep (Irish et al., 2015). Successful sleep is closely linked to an individual's circadian rhythm with consistent sleep and wake times associated with improved subjective reports of sleep quality (Monk et al., 2011).

Instruction regarding bedtime routines is a standard intervention in pediatric practice (Honaker & Meltzer, 2016). While an ideal bedtime routine has not been defined, preschool children show improved sleep onset and total sleep time with fewer night time awakenings when a bedtime routine is followed at least 3–4 nights a week. Starting the bedtime routine during infancy predicted better sleep outcomes (Mindell, Li, Sadeh, Kwon, & Goh, 2015). A bedtime routine that included a consistent bedtime, brushing teeth, and parent-child reading time but not snacks or media time was associated with improved school readiness, executive functioning, and dental health (Kitsaras, Goodwin, Allan, Kelly, & Pretty, 2018).

Bedtime routines have also been effective with adult residential populations. Relaxing activities at bedtime such as massage have been successful in reducing pain and requests for sleep medications in long-term care settings (McFeeters, Pront, Cuthbertson, & King, 2016; Ye & Richards, 2018). Acupressure, the massaging of sleep promoting acupoints on the ears, hands, and feet, has shown promising results (Capezuti et al., 2018; Waits, Tang, Cheng, Tai, & Chien, 2018). While most research on sleep scheduling in adults has focused on shift workers, there are studies addressing the needs of persons with intellectual disabilities. Hylkema and Vlaskamp (2009) found that adults in residential settings may spend half the day in bed, reducing sleep efficiency. Although limited by staff routines, sleep scheduling did improve sleep efficiency in their study. Facilities should implement sleep scheduling to accommodate intraindividual variability if possible. Intraindividual variability in sleep-wake patterns is an emerging area of study with limited research addressing the high sleep variability of persons with intellectual disability (Becker et al., 2017). While ideal scheduling varies among individuals, a mathematical unified model of performance has been developed to guide sleep wake scheduling for optimal performance on a psychomotor vigilance task (Ramakrishnan, Wesensten, Balkin, & Reifman, 2016). Sleep scheduling recommendations will need to accommodate both the patient and caregivers.

Sleep-wake scheduling should also address the timing and duration of napping. Napping is typical in preschool children (Staton et al., 2017). Naps have been linked to memory consolidation in typically developing children, but this benefit was not found in children with Down syndrome. Altered sleep architecture with decreased REM sleep in children with Down syndrome is associated with decreased recall on a word learning task (Spanò et al., 2018).

Positive effects of napping include increased pain tolerance, attention, and reaction time as well as stress reduction with effects of stress, pain tolerance, and health found only in persons who nap many times a week (Faraut, Andrillon, Vecchierini, & Leger, 2017). Short napping, less than 30 minutes, has not been shown to disrupt sleep in persons without sleep disturbances; however, additional research is needed to understand the effects of napping on individuals with intel-

lectual disabilities (Irish et al., 2015). Longer naps, greater than 1 hour in the elderly, have been associated with increased risk of heart disease and diabetes, while napping less than 30 minutes may be beneficial (Faraut et al., 2017). The acute onset of napping or excessive daytime sleepiness in persons with intellectual disability signals the need for a medical evaluation with possible referral to a sleep specialist. Additional study is needed to understand the optimal timing and duration of naps in persons with intellectual disabilities.

Sleep Hygiene – Environmental Factors

Addressing environmental factors is critical to addressing sleep disturbances. Noise, light, temperature, and bedding can be altered to improve sleep. For persons with sensory issues, environmental interventions can improve sleep dramatically. In addition to sensory issues, seeing items associated with daytime activities may cue daytime behaviors. For example, seeing clothes may trigger dressing. Phones or electronics near the bed may promote gaming. Caregivers should be instructed to pay careful attention to the impact of environmental factors on the sleep of persons with intellectual disabilities.

Environmental Factors – Noise

Noise at levels as low as 40 decibels (quiet conversation) can impact sleep architecture and sleep quality, with children and the elderly more sensitive to noises (Halperin, 2014). The brain perceives and reacts to noise during sleep with arousal and, over time, a negative impact on health (Basner et al., 2014). To limit cardiovascular and other health effects, the World Health Organization (2012) has recommended that sleep environments limit noise to 30–40 dBA (sound level as perceived by the ear). In addition to sound level, individuals may be sensitive to sound duration, frequency, and pitch and respond to sounds at lower levels if they have personal significance (Halperin, 2014). Vibrations caused by certain frequencies of traffic noise have been shown to disturb sleep (Caddick, Gregory, Arsintescu, & Flynn-Evans, 2018).

Hyperacusis, sound sensitivity, is well documented in persons with autism and with Williams syndrome. It may trigger pain, fear, or annoyance in addition to causing sleep disturbances. While individual variation in sound sensitivity is common among persons with autism, it occurs in 95% of persons with Williams syndrome and may be due to the deletion of genes which contribute to inner ear hair cell motility (Tyler Richard et al., 2014). Amir, Lamerton, and Montague (2018) reported successful interventions for hyperacusis in a retrospective 5-year study. Most children in their sample were diagnosed with developmental disability including 60% with autism and an additional 20% with neurodevelopmental disabilities including ADHD, Down syndrome, Fetal Alcohol Spectrum Disorder, and cerebral palsy. Behavioral interventions including the development of personalized coping strategies and relaxation training were combined with an instruction on using a programmable sound ball for auditory desensitization. Ear maskers that resemble hearing aids but produce low level white noise were helpful as well.

Noise impacts sleep as well as overall quality of life in acute and long-term settings (Garre-Olmo et al., 2012; Halperin, 2014). If noise is a concern, caregivers can be asked to measure sound with a decibel meter. Many acceptable smartphone apps are available for this purpose, but selection should be researched before downloading as quality varies widely (Kardous & Shaw, 2014). If necessary, sound masking with white noise, noise cancelling head phones, and ear plugs may be used to modulate environmental sounds (Xyrichis, Wynne, Mackrill, Rafferty, & Carlyle, 2018). Architectural modifications including noise reduction panels, sound reducing doors and windows, heavy window coverings, and wall hangings that reduce noise can also be considered (Caddick et al., 2018).

Environmental Factors – Light

The timing, duration, frequency, and intensity of light exposure impact sleep quality and architecture through effects on circadian rhythm and the homeostatic sleep drive (Wams et al., 2017). Sleep architecture can be disrupted by exposure to a dim nightlight light (5 lux) even when the eyes are closed and during sleep (Cho et al., 2016). Bright home lighting can delay evening melatonin production and release (Burgess & Molina, 2014). Blue light such as that emitted by many electronics is more disruptive to sleep than red light (Caddick et al., 2018). Inexpensive glasses can block blue light. Electronic devices can be switched to night mode – emitting less blue light. Outdoor lights visible through windows during the night is associated with poorer sleep quality (Ohayon & Milesi, 2016). Light intensity is measured with a lux meter. Available smartphone technology does not provide the accuracy of commercially available light meters (Cerqueira, Carvalho, & Melo, 2018). Fortunately, well-calibrated meters are available for under 40 dollars. The sleep-disrupting effects of light can be minimized in residential settings by using infrared lighting in sleep areas, closing light blocking window coverings, and using eye masks during sleep.

Exposure to white light of at least moderate intensity (1000 lux, comparable to an overcast day) for at least 30 minutes during the morning is associated with increased alertness during the day and improved sleep at night (Xu & Lang, 2018; Ye & Richards, 2018). This benefit of light is used in chronotherapy to treat insomnia and circadian rhythm sleep disorders as well as sleep disturbances in persons with dementia and is well documented though interventions described vary (van Maanen, Meijer, van der Heijden, & Oort, 2016). Residential facilities often fail to meet light requirements for sleep health (Joseph, Choi, & Quan, 2015). For persons with limited mobility or those with limited opportunity to spend time outdoors, a light box may provide chronotherapy (Dutt, Roduta-Roberts, & Brown, 2015).

Environmental Factors – Temperature

Sleep is prompted by a decrease in body temperature due to vasodilation of the hands and feet which is coupled with the circadian rhythm and linked to melatonin secretion (Kräuchi, 2007; Okamoto-Mizuno & Mizuno, 2012). Most people prefer sleeping in a cool room; however, preferences vary between individuals and for a given individual at different times of the year. Temperature around 90° F impacts sleep architecture, an affect not seen with cold exposure (Okamoto-Mizuno & Mizuno, 2012). An individual's sleep microclimate is created by bedding, bed clothes, air movement, room temperature, and humidity. Each of these may trigger sensitivity issues or breathing difficulties in susceptible persons. Increased temperature is associated with exacerbation of sleep-disordered breathing (Weinreich et al., 2015). Temperature, humidity, and air movement can have a mediating effect on odors which may stimulate or irritate some individuals. The scent of lavender was shown to have a relaxing effect with decreases in skin temperature, blood pressure, and heart rate similar to the body's natural pre-sleep state in one study (Sayorwan et al., 2012) but overall results have been mixed (Lillehei & Halcon, 2014). A systematic review of aromatherapy as a sleep aid for children with autism found that it was not effective (McLay & France, 2016).

Warming of hands and feet with massage or foot baths to cause vasodilation and mimic normal pre-sleep has been shown to decrease sleep onset in some studies, but results are mixed (Hwang & Shin, 2016; Okamoto-Mizuno & Mizuno, 2012). Very small increases in skin temperature, not body temperature, have been associated with deeper sleep and a decrease in early morning awakening (Raymann, Swaab, & Van Someren, 2008). While this study used sophisticated equipment to control skin temperature, the authors suggest low-cost sensors be used to measure skin temperature, and if values are low, preheating the bed might shorten sleep onset and an electric blanket on a timer and a very low setting might decrease early morning awakening.

Persons with intellectual disabilities may fail to maintain optimal skin temperature because of clothing or bedding choices. Mobility may limit adjusting bed coverings. The association between skin temperature and sleep in persons with intellectual disability merits further study.

Environmental Factors – Bedding

Bedding, including bed, pillows, and bed coverings as well as clothing, contributes to comfort and impact sleep quality. Special bedding includes weighted blankets, cooling or heating mattress covers, bed alarms, and alternating pressure mattresses. Special pillows abound including the Dreampad Pillow® that plays music to promote relaxation and others designed for persons with sleep apnea. In small studies, the Dreampad Pillow®has been shown to reduce sleep onset and night awakenings and improve overall sleep quality (Gutman et al., 2017; Ho & Siu, 2018). While preferred by many study participants, weighted blankets have shown inconsistent results for sleep (Gringras et al., 2014; Gutman et al., 2017). For persons with a history of roaming at night, bed or door alarms are helpful signals for caregivers. Alternating pressure mattresses are designed to minimize the risk of pressure ulcers in persons who are bedridden or have limited mobility. Persons with cerebral palsy (CP) may require night orthoses to prevent contractures and/or sleep positioning systems to prevent hip migration. Night orthoses were not found to cause sleep disturbance in a study of 82 children (Mol et al., 2012). Sleep positioning systems have not been shown to improve sleep quality or reduce pain in randomized studies (Blake et al., 2015). Effective pain management is critical to improve the sleep of persons with CP (Lelis, Cardoso, & Hall, 2016).

Vignette – Follow-Up

Paul volunteers that he now has a fan in his room and is using his CPAP most nights. Paul and Alexis report that Paul is falling asleep more easily and has experienced less daytime sleepiness since naps have been eliminated. This is confirmed by the sleep diary documenting shortened sleep onset latency and the EPSS noting decreased daytime sleepiness. Nocturnal enuresis continues three or four nights a week around 1 am. Roaming continues. On some nights this appears to occur when Paul wakes, the bed is wet, and he becomes upset. With medical causes for enuresis ruled out, you suggest scheduled awakenings for toileting. A later bedtime is discussed to increase sleep pressure and decrease awakenings. Alexis feels a later bedtime for Paul would create difficulties with his roommate and could be met with staff resistance. You explain to Paul and Alexis that staff should wake Paul at midnight and request that he use the toilet and then return to bed. You praise Paul's good decision to use his CPAP and discuss ways to reinforce CPAP use.

Interventions for Sleep Disturbances

While sleep hygiene education is a common component of CBT-I, it must be combined with other interventions to adequately address insomnia. For persons with intellectual disabilities, multicomponent approaches without CBT are common. Interventions beyond sleep hygiene education include relaxation, sleep restriction therapy, stimulus control, mindfulness, and intensive sleep retraining.

Relaxation

Relaxation to address behavioral, cognitive, and neurological hyperarousal is one of the most recommended interventions for sleep disturbances (Murawski, Wade, Plotnikoff, Lubans, & Duncan, 2018). Progressive relaxation was the first intervention for insomnia (Jacobson, 1929). Common practices include autogenic training, progressive muscle relaxation, guided imagery, and abdominal breathing. Twenty minutes of slow-paced breathing at 10 second intervals decreased sleep onset latency, time to onset of slow wave sleep,

and total wake time with improved report of sleep quality in a small, controlled study (Tsai, Kuo, Lee, & Yang, 2015). The authors suggest that persons with insomnia suffer autonomic dysfunction which is modulated by slow deep breathing exercises which decrease heart rate variability. Heart rate variability has been suggested as a biomarker for insomnia; however, a systematic review of 22 studies suggested additional research is necessary for confirmation (Dodds, Miller, Kyle, Marshall, & Gordon, 2017). In a prospective, cohort study of 173 persons completing autogenic training, 73% reported sleep improvement (Bowden, Lorenc, & Robinson, 2012). Progressive muscle relaxation has been shown to improve sleep quality as measured by the Pittsburg Sleep Quality Index and the Epworth Sleepiness Scale (Sun, Kang, Wang, & Zeng, 2013). A controlled multisensory environment, such as Snoezelen®, has shown efficacy as a means of relaxation for persons with intellectual disabilities (Bergstrom, O'Brien-Langer, & Marsh, 2018; Lotan & Gold, 2009). Given the contribution of sensory over-responsivity to sleep disturbances in persons with autism (Mazurek & Petroski, 2015), decreasing arousal with a controlled multisensory environment may be helpful. Additional research is needed to define the relaxation parameters necessary to provide benefit for sleep disturbances.

While often recommended, relaxation strategies are rarely implemented consistently. In the 2007 National Health Survey of over 4000 adults who reported insomnia, only 23% used relaxation to address sleep difficulty, with deep breathing being the most common technique employed (Bertisch, Wells, Smith, & McCarthy, 2012). Relaxation was used by less than 1% in a study of 402 persons in Hong Kong with sleep difficulties (Yeung et al., 2014). In general populations, patients who believed their behavior could significantly impact health, a high internal health locus of control, were more likely to use relaxation strategies (Cramer, Lauche, Langhorst, Dobos, & Paul, 2013). While all individuals benefit from detailed relaxation instruction and practice, modifications may be necessary for persons with intellectual disabilities. Careful selection of relaxation strategy is required as well. For example, progressive muscle relaxation and deep breathing may challenge some with neuromuscular disorders or cerebral palsy. The risks and benefits of relaxation techniques for persons with seizures have been questioned and require further study. Treatment team input should be considered before undertaking sleep interventions including relaxation. Groden, Weidenman, and Diller (2016) detail the process of teaching relaxation to persons with developmental disabilities, highlighting the patience necessary during an extended skill acquisition phase including the use of personalized cues and reinforcement and the importance of attention to generalizing the relaxation skills to other environments.

Sleep Restriction Therapy (SRT)

Spielman, Saskin, and Thorpy (1987) developed sleep restriction therapy (SRT) as a treatment to ameliorate the prolonged sleep onset latency, awakenings, and poor sleep quality experienced by persons with insomnia. SRT is hypothesized to increase sleep pressure and circadian control while decreasing hyperarousal and time in bed, a perpetuating factor under the 3P model (Spielman, Caruso, & Glovinsky, 1987; Spielman, Yang, & Glovinsky, 2011) The mechanism of action of SRT has not been determined (Maurer et al., 2018). The AASM recommends SRT as a standalone treatment of chronic insomnia or as a component of CBT-I or multicomponent therapy without CBT (Morgenthaler, Kramer, et al., 2006). Shorter sleep latency, sleep consolidation/compression, and improved sleep quality have been widely reported benefits of SRT (Cheung, Jarrin, Ballot, Bharwani, & Morin, 2019; Maurer et al., 2018; Miller et al., 2014). In clinical trials the instructions for SRT and sleep parameter cited vary widely, leading to recommendations for standardization (Kyle et al., 2015). Maurer et al. (2018) have proposed the Triple-R model of SRT to address the need for standardization to facilitate research regarding efficacy and mechanism of action. The Triple-R

model focuses on the restricting, regulating, and reconditioning components of SRT.

Individualized directions for SRT are based on average sleep time data from a two-week sleep diary. Initially, the total time in bed is restricted to the average sleep time. No napping is allowed. A consistent wake time is selected, with bedtime determined by counting back from the wake time, the hours allowed in bed. As sleep onset latency and awakenings decrease, time in bed is increased by 15 minutes until any daytime sleepiness is eliminated (Spielman, Saskin, & Thorpy, 1987). In practice, an average sleep time of 6 hours and a daily wake time of 7 am would result in an initial bedtime of 1 am. The patient should be cautioned that this will result in daytime sleepiness which could impact driving or work performance (Spielman et al., 2011). Other common SRT side effects include fatigue, extreme sleepiness, reduced motivation, headache, agitation/irritability, and euphoria (Kyle, Morgan, Spiegelhalder, & Espie, 2011). Elements of SRT have been implemented for persons with ID and sleep disturbances such as limiting time in bed and eliminating napping (McCurry et al., 2012). Studies implementing a full SRT protocol in persons with intellectual disability were not identified in a PubMed search. SRT should be approached with caution in persons with seizures, mental disorders, or behavioral disturbances as sleep restriction may exacerbate these conditions.

Stimulus Control Therapy (SCT)

Richard Bootzin (1972) draws on the principles of operant and classical (Pavlovian) conditioning as a basis for stimulus control therapy (SCT) for insomnia, with an emphasis on addressing sleep onset difficulties. Using operant principles, SCT targets insufficient stimulus control by strengthening the association between the internal and external cues for falling asleep. The classical conditioning element addresses discriminative stimuli including physical cues for sleep onset distress or anxiety and internal signals promoting arousal (Bootzin & Perlis, 2011). In SCT the therapist promotes improved sleep by individualizing six instructions:

1. Lie down to go to sleep only when you are sleepy.
2. Do not use your bed for anything except sleep.
3. If you find yourself unable to fall asleep, get up and go into another room. Stay up as long as you wish and then return to the bedroom to sleep.
4. If you still cannot fall asleep, repeat step 3.
5. Set your alarm and get up at the same time every morning irrespective of how much sleep you got during the night.
6. Do not nap during the day (Bootzin & Epstein, 2011, p. 443).

While SCT is recommended by the American Academy of Sleep Medicine, backed by randomized controlled studies, these studies did not include persons with intellectual disabilities (Morgenthaler, Kramer, et al., 2006). Case reports indicate that STC is successful for insomnia in persons with intellectual disability (Gunning & Espie, 2003). SCT allows for modification of these instructions for persons with intellectual disability but does not recommend specific adaptations (Bootzin & Perlis, 2011). Modifications should be person-centered. To implement instruction number one, caregivers should help with the recognition of sleepiness cues such as yawning, psychomotor slowing, or eyelids drooping and give verbal feedback suggesting readiness for sleep. Caregivers should also monitor for anxiety or agitation at bedtime. Further assessment may be necessary to identify the source of bedtime arousal to allow for the implementation of targeted calming strategies. Limiting bed/bedroom activity to sleep may require modification to remove distractions from sight. Instructions number three and four may be difficult to implement in residential settings. Additional staff may be required to assist with rising and returning to bed. Caution must be taken to avoid the association of waking with getting out of bed and leaving the room. Regular sleep and wake times should not be confused with bedtime and rising time. Bedtimes and rising times should be monitored

to limit the wake time in bed as the goal of SCT is to strengthen the association of the bed with sleep. Napping should generally be avoided, but individual needs should be considered as napping may be beneficial for some (Faraut et al., 2015).

Multicomponent Approaches Without CBT

Multicomponent approaches combine the elements of CBT-I that are clinically indicated for an individual's sleep disturbance. Multicomponent sleep intervention without CBT is a common approach to the treatment of sleep disturbances in persons with intellectual disabilities as CBT is less effective for persons with moderate–to-profound ID and may not be suitable for all persons with mild ID (Jennings & Hewitt, 2015; Roberts & Kwan, 2018; Stott, Charlesworth, & Scior, 2017). Multicomponent therapy may be delivered individually or in a group format and is initiated after sleep assessment defines an individual's sleep issues and rules out the need for medical interventions. Typical components are sleep health education and the development of an individualized treatment plan that may include any combination of CBT-I elements. Hanley (2005) has developed a functional behavioral assessment format, the Sleep Assessment and Treatment Tool (SATT), to inform multicomponent interventions for children. The SATT has been used by studies to develop treatments for children with autism and other developmental disabilities (Jin, Hanley, & Beaulieu, 2013; McLay, France, Knight, Blampied, & Hastie, 2018).

Multicomponent therapy delivered in group format has allowed parents and caregivers to successfully address sleep disturbances in persons with intellectual disability. Johnson et al. (2013) reported high parent satisfaction with a five-session manualized behavioral parent management training program for addressing sleep challenges in children with autism. Parents participating in two workshops as part of the *Sleepwise* parent education program for addressing sleep disturbances in youth with developmental disabilities reported decreased sleep disturbances in children and less stress in the home (Moss, Gordon, & O'Connell, 2014). The *Sleepwise* program includes a home visit/assessment before the workshops and telephone support after the workshops. The Sleep Education Program for caregivers in adult family homes includes workshops addressing sleep health, the development of an individualized sleep plan with the A-B-C behavioral approach, problem solving, and development of a maintenance sleep plan (McCurry, LaFazia, Pike, Logsdon, & Teri, 2009; McCurry et al., 2012). Care staff developed successful plans to address sleep/wake scheduling issues, nighttime agitation and disruptive behaviors, and night fears with sleep hygiene, sleep restriction, stimulus control, and cognitive interventions targeting caregiver beliefs and expectations. Specific interventions included the introduction of new daytime activities, sleep schedule changes, increased exercise and light exposure, and reducing environmental cues for daytime behaviors (McCurry et al., 2012). A systematic review of 33 studies reporting on parent-delivered behavioral interventions for sleep in children with neurodevelopmental disorders found the practice to be effective (Rigney et al., 2018). Caregivers participating in multicomponent sleep therapy programs found the information and skills empowering and successfully implemented strategies for sleep improvement. The use of telecommunication to support and reinforce caregivers problem solving and to further refine sleep behavior programs is a promising practice.

Intensive Sleep Retraining (ISR)

Intensive sleep retraining (ISR) for initial onset insomnia increases the association of the bed as a discriminative stimuli for sleep similar to sleep restriction therapy (SRT) but compresses the treatment time into 1 day rather than the weeks required by SRT (Harris, Lack, Kemp, Wright, & Bootzin, 2012; Harris, Lack, Wright, Gradisar, & Brooks, 2007). In laboratory-based ISR, patients arrive in the evening after a night of restricted

sleep. They are allowed to fall asleep for 2–3 minutes, then woken, allowing a brief period of stage 1 sleep followed by 20 minutes of wake time. As this cycle is repeated over 24 hours, sleep pressure increases, leading to shorter sleep onset times (Lack, Scott, Micic, & Lovato, 2017). In Harris' randomized controlled trial (2012), 47% of patients receiving ISR achieved sleep onset in 30 minutes or less compared to 38% of those receiving SRT. Combined ISR and SRT resulted in 61% achieving success with decreased sleep onset time and awakenings and increased total sleep time. Improvements were maintained over a 6-month follow-up period. The Sleep on Cue smartphone app, available at sleeponq.com, uses the principles of ISR to guide at home treatment. A small study evaluating the Sleep on Cue app in 12 university students in a sleep lab found the application promising but in need of further study (Scott, Lack, & Lovato, 2018).

No ISR studies involving persons with intellectual disability or autism or children and adolescents were found in a PubMed search conducted during the preparation of this chapter. ISR should be approached with caution in persons with intellectual disability as sleep restriction may increase irritability and behavioral disturbances. In addition, it may induce seizure activity in persons with seizures and exacerbate preexisting mental disorders.

Mindfulness

Shallcross, Visvanathan, Sperber, and Duberstein (2018) have developed an integrative etiological model of sleep disturbance to explain how the components of mindfulness address cognitive behavioral elements of sleep disturbance. Mindfulness components include experiential awareness, attention control, and acceptance. Cognitive behavioral elements of sleep disturbance addressed through mindfulness practices are rumination, primary arousal, secondary arousal, distorted perceptions, selective attention/sleep monitoring, and effort. Shallcross et al. (2018) have presented an argument for the addition of mindfulness practices to CBT-I with a systematic review identifying mindfulness as a promising addition to CBT-I (Rusch et al., 2018). Meta-analyses of randomized controlled studies found that mindfulness practices decreased time to sleep onset and night awakenings while improving sleep efficiency and quality but the effect on total sleep time was not significant (Gong et al., 2016; W Kanen, Nazir, Sedky, & K Pradhan, 2015). Lindsay and Creswell (2017) have developed Monitor and Acceptance Theory to explain attention monitoring and acceptance as the mechanisms of action for mindfulness practices. Lau, Leung, Wing, and Lee (2018) have confirmed the contributions of awareness and acceptance to the sleep enhancement through mindfulness practices using Baer's Five Facet Mindfulness Questionnaire (2008). A group of leading scientists have called attention to the lack of a universal definition of mindfulness and challenged the scientific community to define the concept further (Van Dam et al., 2017).

Behavioral Interventions for Children

Sleep hygiene, relaxation, and caregiver education are common interventions for sleep disturbances across all ages; however, few of the adult interventions for insomnia have been explored in children and persons with intellectual disability. The AASM has issued a practice parameter identifying interventions for young children with sleep disturbances (Morgenthaler, Owens, & Alessi, 2006). The AAP offers parents age-specific guidance at healthychildren.org. Consulting a sleep specialist and implementing behavioral interventions for sleep disturbances are common themes across sources.

As with adults, the goals of interventions are to improve sleep quality and efficacy by decreasing arousal, strengthening the association between the bed and sleep, increasing the sleep pressure, consolidating sleep, and strengthening circadian sleep associations (Owens & Moore, 2017). Calming bedtime routines, limiting daytime sleeping, and exercise can decrease evening arousal. Avoiding time in bed unless sleeping

serves to strengthen the association between the bed and sleep. Faded bedtimes, temporarily setting a later bedtime, is a strategy that increases sleep pressure and promotes consolidated sleep (Meltzer, 2010; Piazza & Fisher, 1991). Extinction, ignoring the undesired behaviors (crying or calling out to the parent), removes parental attention as a perpetuating factor for insomnia and may encourage the child to use calming strategies. Variations include graduated extinction, brief, scheduled reassurance by the parent at set intervals that are gradually increased and then stopped, and extinction with stimulus fading, the parent being near and then over several nights moving further away (Corkum, Davidson, Tan-MacNeill, & Weiss, 2014). With extinction strategies an extinction burst or temporary increase in challenging behavior may occur. In addition, a token economy that rewards success with extinction strategies or compliance with an established bedtime routine may be implemented. For night awakenings, a bedtime pass allowing one visit to the parent during the night may encourage the use of self-calming strategies (Moore, Friman, Fruzzetti, & MacAleese, 2007). Spruyt and Curfs (2015) and Corkum et al. (2014) provide detailed information regarding the behavioral management of sleep challenges in children with developmental disabilities.

Medications for Sleep Disturbances

When first-line behavioral and cognitive interventions do not fully ameliorate sleep challenges, pharmacological interventions may be considered (See Fig. 36.2). Pharmacological interventions include prescribed medications and over-the-counter sleep aids including complementary and alternative (CAM) products. The National Ambulatory Medical Care Survey reported a 293% increase in prescriptions for sleep between 1999 and 2010 (Ford et al., 2014). Medication prescriptions for sleep disorders for indications outside the FDA-approved uses, or "off-label" prescribing, ranks highest among off-label prescriptions, with the tricyclic antidepressants being the most common (Vijay, Becker, & Ross, 2018; Wong et al., 2017). Interestingly, a systematic review and meta-analysis of studies found a significant placebo effect for sleep medications on perceived sleep onset latency, total sleep time, and global sleep quality (Yeung, Sharpe, Glozier, Hackett, & Colagiuri, 2018).

Antihistamines
Diphenhydramine (Benadryl)
Doxlyamine (Unisom)
Antidepressants
Doxepin (Silenor, Sinequan)
Trazodone (Desyrel)*
Amitriptyline (Elavil)*
Mirtazapine (Remeron)*
Melatonin Receptor Agonist
Ramelteon (Rozerem)
Orexin Receptor Antagonist
Suvorexant (Belsomra)
Nonbenzodiazepines
Eszopiclone (Lunesta)
Zaleplon (Sonata)
Zolpidem (Ambien, Intermezzo)
Benzodiazepines
Estazolam (ProSom)
Flurazepam (Dalmane)
Lorazepam (Ativan)*
Quazepam (Doral)
Triazolam (Halcion)
Temazepam (Restoril)
Barbituates
Butabarbital (Butisol)
Secobarbital (Seconal)
***Not FDA Approved**

Fig. 36.2 Medications for Insomnia

There are no medications that are approved by the U.S. Food and Drug administration for insomnia in children. Prolonged release melatonin, Slenyto®(PedPRM), has been approved by the European Medicines Agency (2018) to treat insomnia in youth with autism and Smith-Magenis syndrome after behavioral interventions have failed. A survey of child and adolescent psychiatrists (n = 1273) found that 96% had prescribed medication for sleep and 88% had recommended an over-the-counter sleep aid in the last month (Owens, Rosen, Mindell, & Kirchner, 2010). In a study of children with ASD between the ages of 4 and 10, a third were prescribed medication for sleep (Malow et al., 2016). In the National Institutes of Mental Health

Evolution Pathways to Insomnia Cohort (EPIC)Study of over 900 individuals meeting DSM-5 criteria for insomnia, over 20% reported taking prescription medication for sleep, with about a third also using over-the-counter preparations (Pillai et al., 2016). Epidemiological studies reporting on psychotropic medication use in persons with developmental disabilities typically report medication category without a clear indication if the medication was prescribed for sleep or another mental disorder. Additional studies are needed regarding the prevalence of sleep medication use in persons with developmental disabilities with detailed information on specific medication for each neurodevelopmental disorder.

To guide clinical decision making, professional medical organizations have published guidelines including the *Clinical Practice Guideline for the Pharmacological Treatment of Adults* and *Management of Chronic Insomnia Disorder in Adults: An American Academy of Sleep Medicine* and the *Clinical Practice Guideline A Clinical Practice Guideline from the American College of Physicians*; however, the *Guidelines* note that the reviews of scientific evidence for the medications and OTC products reviewed were limited (Qaseem et al., 2016; Sateia, Buysse, Krystal, Neubauer, & Heald, 2017). For a comprehensive review of the literature, see the AASM*Guideline* (2017). In formulating recommendations, the AASM *Guideline* considers the totality of evidence from the viewpoint of the patient, answering the question: "[would the] well-informed patient use [a given treatment] over no treatment?" (Sateia et al., 2017). The AASM and ACP guidelines do not address special populations including children and persons with developmental disabilities. *The Frith Prescribing Guidelines for People with Intellectual Disability* includes a brief review of medication strategies for the treatment of sleep disorders in persons with ID (Bhaumik, Gangadharan, Branford, & Barrett, 2015). All three publications highlight the pressing need for additional research on the management of sleep disorders in children and persons with developmental disabilities.

Many patients will have experienced limited success with over-the-counter (OTC) sleep aids prior to seeking professional guidance for sleep issues. The most common OTC products are the antihistamines – diphenhydramine and doxylamine succinate, the hormone - melatonin, and the herbal products – valerian, chamomile, and glycine (Zhou, Gardiner, & Bertisch, 2017). Diphenhydramine and doxylamine are available without a prescription as FDA-regulated formulations. Melatonin is available in FDA-regulated (Circadin) and FDA-unregulated formulations. The dietary supplements valerian, L-tryptophan, chamomile, glycine are unregulated formulations. The FDA reviews product labeling but not safety or quality data for dietary supplements. The NIH National Center for Complementary and Integrative Health provides reliable information on the use of CAM for sleep disorders (NCCIH, 2017). The AASM*Clinical Practice Guideline for the Pharmacological Treatment of Adults* and *Management of Chronic Insomnia Disorder in Adults* do not recommend OTC sleep aids for the treatment of chronic insomnia citing a "relative lack of safety and efficacy data" (Sateia et al., 2017).

Melatonin is a circadian rhythm regulating hormone that is naturally produced by many plants and most animals (Cipolla-Neto & Amaral, 2018). Millions of adults and nearly half a million children in the United States took melatonin in 2012 with use increasing dramatically between 2007 and 2012 (Black, Clarke, Barnes, Stussman, & Nahin, 2015; Clarke, Black, Stussman, Barnes, & Nahin, 2015). While the AASM cites evidence for the treatment of some circadian rhythm sleep-wake disorders with melatonin, it does not recommend melatonin for sleep onset or sleep maintenance insomnia (Auger et al., 2015; Sateia et al., 2017). Still melatonin is commonly used to treat sleep disturbances in persons with intellectual disabilities and some studies have shown efficacy (Abdelgadir, Gordon, & Akobeng, 2018; Schwichtenberg & Malow, 2015; Ward, Nanjappa, Hinder, & Roy, 2015). Melatonin is commonly used to regulate circadian rhythms in persons with Smith-Magenis syndrome, neurological syndromes with irregular sleep-wake

rhythm disorder, and persons with blindness who have non-24-hour sleep-wake rhythm disorder (Auger et al., 2015; Barboni et al., 2018). A randomized, double-blind study of 125 youth with ASD (n = 121) or Smith-Magenis syndrome (n = 4) found prolonged release melatonin to be safe and effective (Gringras, Nir, Breddy, Frydman-Marom, & Findling, 2017).

Melatonin is generally well tolerated and considered to be safe, but there are some important caveats to its use (Abdelgadir et al., 2018; Culpepper & Wingertzahn, 2015). The content of melatonin in OTC products has been found to vary widely from brand to brand, from −83% to +478%, and between lots, up to 465%, for a given brand (Erland & Saxena, 2017). In addition, tryptophan and serotonin have been found in many preparations (Cerezo et al., 2016; Erland & Saxena, 2017). While melatonin production is stable day-to-day for an individual, there can be substantial variation from person to person; the optimal time for melatonin dosing can be determined by testing the dim light melatonin onset (DLMO) in blood or saliva samples or, as a less than optimal substitute, given one hour before the usual bedtime when testing is not feasible (Cipolla-Neto & Amaral, 2018). Dosing guidelines vary as well (Auger et al., 2015; Bruni et al., 2015; Malow et al., 2012; Sateia et al., 2017). Finally, it is unclear if seasonal dosing adjustments are necessary (Cipolla-Neto & Amaral, 2018). These caveats may help explain the mixed results regarding melatonin efficacy and highlight the importance of additional well-controlled studies.

Additional Sleep Disturbances Common in Clinical Practice

Common sleep disturbances encountered in clinical practice include nightmares, night terrors, sleep-walking, sleep talking, and sleep-related aggression. Nightmares are more common and typically transient in children but have been found to be more persistent in persons with neurodevelopmental disabilities (Angriman, Caravale, Novelli, Ferri, & Bruni, 2015). Nightmares are a nearly universal experience. Nightmare disorder is experienced by 4% of adults and may be idiopathic, related to medications or substance use, or to a comorbid mental disorder such as PTSD (Morgenthaler et al., 2018). Sleep-walking and night terrors are NREM sleep arousal disorders and typically occur early in the nightly sleep cycle (Bollu, Goyal, Thakkar, & Sahota, 2018). Sleep-walking and sleep terrors are common in youth with neurofibromatosis type 1 (Licis et al., 2013); Stores, 2014). Night terrors and fearful dreams are common in Angelman syndrome (Spruyt, Braam, & Curfs, 2018). Sleep talking may occur in REM or NREM sleep and is considered a normal variant unless it causes serious disruption (Alfonsi, D'Atri, Scarpelli, Mangiaruga, & De Gennaro, 2019). Disruption of the muscle atonia normally associated with REM sleep may be responsible for REM sleep behavior disorders which may include minor movements or complex and aggressive movements, and/or vocalizations (Bollu et al., 2018). [See Chap. 22 for *The Assessment of Sleep Disorders: Dual Diagnosis with Intellectual Disability.*]

The AASM*Position Paper for the Treatment of Nightmare Disorder in Adults: An American Academy of Sleep Medicine Position Paper* reviews the medications and behavioral interventions for nightmares in detail but does not address interventions for persons dually diagnosed with intellectual disability and nightmares (Morgenthaler et al., 2018). For nightmare disorder the *Guideline* recommends and reviews cognitive behavioral therapy (CBT); exposure, relaxation, and rescripting therapy (ERRT); hypnosis; lucid dreaming therapy; progressive deep muscle relaxation; sleep dynamic therapy; self-exposure therapy; systematic desensitization; and testimony method. The AASM has also published *Practice Parameters for Behavioral Treatment and Bedtime Problems and Night Wakenings in Infants and Young Children* (Morgenthaler, Kramer, et al., 2006). Research regarding the treatment of nightmare disorder in persons with intellectual disability is very limited (Gilderthorp, 2014; Willner, 2004).

Common approaches to addressing nightmares include practicing good sleep hygiene, reassurance, and discussing the dream, if remembered the next day. In addition to the lifestyle and environmental factors addressed previously, persons with nightmares should avoid exposure to violent or frightening media as this has been linked to nightmare content (Stephan, Schredl, Henley-Einion, & Blagrove, 2012; Van den Bulck, Çetin, Terzi, & Bushman, 2016). The contribution of stressors or medications to nightmares should be considered. Sleep architecture may be altered by many medications including antidepressants, antipsychotics, nonbenzodiazepine hypnotics, stimulants, some medications for allergies and asthma, narcotic analgesics, most seizure medications, and some cardiac medications (Doghramji & Jangro, 2016; Schutte-Rodin et al., 2008; van de Wouw, Evenhuis, & Echteld, 2012). When a person wakes with nightmares, reassurances of safety and that the disturbance was a dream and not real can be helpful as well as staying with the person and offering comfort items such as a nightlight, favorite blanket, or stuffed animal. If recalled, the dream may be discussed the next day and additional reassurances offered.

As opposed to nightmares which typically occur during the second half of sleep, somnambulism, or sleep-walking, and night terrors typically occur during the first few hours of sleep during the transitioning of sleep phases and often near the same time each night. Treatment is directed altering this transition. A systematic review of treatments for sleepwalking synthesized information from case studies as no randomized controlled trials were found, concluding that scheduled waking was the most promising approach (Stallman & Kohler, 2017; Stallman, Kohler, & White, 2018). This approach is also used for sleep terrors (Galbiati, Rinaldi, Giora, Ferini-Strambi, & Marelli, 2015). In implementing scheduled waking, also called anticipatory awakening, the caregiver is instructed to wake the patient 15–20 minutes before the typical time the disturbance occurs and assist the patient in returning to sleep (Frank, Spirito, Stark, & Owens-Stively, 1997).

Sleep talking, or somniloquy, typically does not require treatment and resolves spontaneously. If sleep talking becomes disruptive in residential settings, caregivers should be reassured and lifestyle and environmental sleep hygiene should be addressed (Alfonsi et al., 2019). If sleep talking is part of an aggressive REM sleep behavior disorder, a bed or door alarm may be helpful to alert caregivers and prevent injury. Modifications may need to be made to the environment to prevent injury. Medications for REM sleep behavior disorders include melatonin and clonazepam (Dauvilliers et al., 2018; McGrane, Leung, St. Louis, & Boeve, 2015).

Vignette

After 2 months of regular CPAP use, good sleep hygiene, and scheduled awakenings, Paul began to wake himself for toileting. He no longer experiences daytime sleepiness and is doing well at his job. Group home staff have decreased napping among residents by increasing daytime activities, increased morning light exposure by opening bedroom curtains each morning, and placed white noise machines in bedrooms near the night staff work station.

Concluding Remarks

Mental health professionals must be versed in a wide range of behavioral interventions to address the challenges faced by persons dually diagnosed with intellectual disability and sleep disturbances; however, additional research is necessary to establish a strong evidence base for describing healthy sleep and best practices for intervention in dually diagnosed persons (Meltzer, 2017; Priday, Byrne, & Totsika, 2017). Before implementing behavioral treatments for sleep disturbances, a thorough medical evaluation is necessary. A referral to a sleep specialist should be strongly considered for all persons who are dually diagnosed with sleep disturbance and intellectual disability. Medication should be used with caution and only after behavioral interven-

tions fail to ameliorate the sleep disturbance. To provide further guidance, the American Academy of Sleep Medicine will release an updated clinical practice guideline on the *Behavioral and Psychological Treatments for Chronic Insomnia Disorder in Adults* in late 2020.

References

Abdelgadir, I. S., Gordon, M. A., & Akobeng, A. K. (2018). Melatonin for the management of sleep problems in children with neurodevelopmental disorders: A systematic review and meta-analysis. *Archives of Disease in Childhood, 103*(12), 1155. Retrieved from http://adc.bmj.com/content/103/12/1155.abstract. https://doi.org/10.1136/archdischild-2017-314181

Adams, H. L., Matson, J. L., Cervantes, P. E., & Goldin, R. L. (2014). The relationship between autism symptom severity and sleep problems: Should bidirectionality be considered? *Research in Autism Spectrum Disorders, 8*(3), 193–199. Retrieved from http://www.sciencedirect.com/science/article/pii/S1750946713002365. https://doi.org/10.1016/j.rasd.2013.11.008

Åkerstedt, T., & Gillberg, M. (1990). Subjective and objective sleepiness in the active individual. *International Journal of Neuroscience, 52*(1–2), 29–37. https://doi.org/10.3109/00207459008994241

Alfonsi, V., D'Atri, A., Scarpelli, S., Mangiaruga, A., & De Gennaro, L. (2019). Sleep talking: A viable access to mental processes during sleep. *Sleep Medicine Reviews, 44*, 12–22. Retrieved from http://www.sciencedirect.com/science/article/pii/S1087079218300807. https://doi.org/10.1016/j.smrv.2018.12.001

Amir, I., Lamerton, D., & Montague, M.-L. (2018). Hyperacusis in children: The Edinburgh experience. *International Journal of Pediatric Otorhinolaryngology, 112*, 39–44. Retrieved from http://www.sciencedirect.com/science/article/pii/S0165587618302593. https://doi.org/10.1016/j.ijporl.2018.06.015

Angarita, G. A., Emadi, N., Hodges, S., & Morgan, P. T. (2016). Sleep abnormalities associated with alcohol, cannabis, cocaine, and opiate use: A comprehensive review. *Addiction Science & Clinical Practice, 11*(1), 9. https://doi.org/10.1186/s13722-016-0056-7

Angriman, M., Caravale, B., Novelli, L., Ferri, R., & Bruni, O. (2015). Sleep in children with neurodevelopmental disabilities. *Neuropediatrics, 46*(3), 199–210. https://doi.org/10.1055/s-0035-1550151

Auger, R. R., Burgess, H. J., Emens, J. S., Deriy, L. V., Thomas, S. M., & Sharkey, K. M. (2015). Clinical practice guideline for the treatment of intrinsic circadian rhythm sleep-wake disorders: Advanced sleep-wake phase disorder (ASWPD), delayed sleep-wake phase disorder (DSWPD), non-24-hour sleep-wake rhythm disorder (N24SWD), and irregular sleep-wake rhythm disorder (ISWRD). An update for 2015. *Journal of Clinical Sleep Medicine, 11*(10), 1199–1236.

Autism Speaks. (2018). Strategies to improve sleep in children with autism spectrum disorders. In A. Speaks (Ed.), Retrieved from https://www.autismspeaks.org/tool-kit/atnair-p-strategies-improve-sleep-children-autism.

Baer, R. A., Smith, G. T., Lykins, E., Button, D., Krietemeyer, J., Sauer, S., … Williams, J. M. G. (2008). Construct validity of the five facet mindfulness questionnaire in meditating and nonmeditating samples. *Assessment, 15*(3), 329–342.

Barboni, M. T. S., Bueno, C., Nagy, B. V., Maia, P. L., Vidal, K. S. M., Alves, R. C., … Ventura, D. F. (2018). Melanopsin system dysfunction in smith-magenis syndrome patients. *Investigative Ophthalmology & Visual Science, 59*(1), 362–369. https://doi.org/10.1167/iovs.17-22612

Barnhill, S., & Hollway. (2016). *Sleep disorders*. In R. Fletcher (Ed.), *Diagnostic manual-intellectual disability: A textbook of diagnosis of mental disorders in persons with intellectual disability*. Kingston, NY: NADD.

Basner, M., Babisch, W., Davis, A., Brink, M., Clark, C., Janssen, S., & Stansfeld, S. (2014). Auditory and non-auditory effects of noise on health. *Lancet, 383*(9925), 1325–1332. https://doi.org/10.1016/s0140-6736(13)61613-x

Becker, S. P., Sidol, C. A., Van Dyk, T. R., Epstein, J. N., & Beebe, D. W. (2017). Intraindividual variability of sleep/wake patterns in relation to child and adolescent functioning: A systematic review. *Sleep Medicine Reviews, 34*, 94–121. Retrieved from http://www.sciencedirect.com/science/article/pii/S1087079216300661. https://doi.org/10.1016/j.smrv.2016.07.004

Bei, B., Wiley, J. F., Trinder, J., & Manber, R. (2016). Beyond the mean: A systematic review on the correlates of daily intraindividual variability of sleep/wake patterns. *Sleep Medicine Reviews, 28*, 108–124. Retrieved from http://www.sciencedirect.com/science/article/pii/S1087079215000908. https://doi.org/10.1016/j.smrv.2015.06.003

Beresford, B., McDaid, C., Parker, A., Scantlebury, A., Spiers, G., Fairhurst, C., … Thomas, M. (2018). Pharmacological and non-pharmacological interventions for non-respiratory sleep disturbance in children with neurodisabilities: A systematic review. *Health technology assessment (Winchester, England), 22*(60), 1–296. Retrieved from https://www.ncbi.nlm.nih.gov/pubmed/30382936. Retrieved from https://www.ncbi.nlm.nih.gov/pmc/PMC6231061/. https://doi.org/10.3310/hta22600

Bergstrom, V. N. Z., O'Brien-Langer, A., & Marsh, R. (2018). Supporting children with fetal alcohol spectrum disorder: Potential applications of a Snoezelen multisensory room. *Journal of Occupational Therapy, Schools, & Early Intervention, 12*(1), 98–114. https://doi.org/10.1080/19411243.2018.1496869

Bertisch, S. M., Wells, R. E., Smith, M. T., & McCarthy, E. P. (2012). Use of relaxation techniques and complementary and alternative medicine by American adults with insomnia symptoms: Results from a national survey. *Journal of Clinical Sleep Medicine: Official Publication of the American Academy of Sleep Medicine, 8*(6), 681–691. Retrieved from https://www.ncbi.nlm.nih.gov/pubmed/23243402. Retrieved from https://www.ncbi.nlm.nih.gov/pmc/PMC3501665/. https://doi.org/10.5664/jcsm.2264

Bhaumik, S., Gangadharan, S. K., Branford, D., & Barrett, M. (2015). *The Frith prescribing guidelines for people with intellectual disability*. Chichester, UK: Wiley.

Black, L. I., Clarke, T. C., Barnes, P. M., Stussman, B. J., & Nahin, R. L. (2015). Use of complementary health approaches among children aged 4–17 years in the United States: National Health Interview Survey, 2007–2012. *National Health Statistics Reports, 78*, 1.

Blake, S. F., Logan, S., Humphreys, G., Matthews, J., Rogers, M., Thompson-Coon, J., … Morris, C. (2015). Sleep positioning systems for children with cerebral palsy. *Cochrane Database of Systematic Reviews*, (11), Cd009257. https://doi.org/10.1002/14651858.CD009257.pub2

Blunden, S., & Rigney, G. (2015). Lessons learned from sleep education in schools: A review of dos and don'ts. *Journal of Clinical Sleep Medicine: Official Publication of the American Academy of Sleep Medicine, 11*(6), 671–680. Retrieved from https://www.ncbi.nlm.nih.gov/pubmed/25766709. Retrieved from https://www.ncbi.nlm.nih.gov/pmc/PMC4442228/. https://doi.org/10.5664/jcsm.4782

Bollu, P. C., Goyal, M. K., Thakkar, M. M., & Sahota, P. (2018). Sleep medicine: Parasomnias. *Missouri Medicine, 115*(2), 169–175. Retrieved from https://www.ncbi.nlm.nih.gov/pubmed/30228711. Retrieved from https://www.ncbi.nlm.nih.gov/pmc/PMC6139852/

Bootzin, R. (1972). Proceedings of the 80th Annual Convention of the American Psychological Association (vol. 7, pp. 395–396).

Bootzin, R. R., Epstein, D., & Wood, J. M. (1991). Stimulus control instructions. In P. J. Hauri (Ed.), *Case studies in insomnia* (pp. 19–28). Boston, MA: Springer.

Bootzin, R. R., & Epstein, D. R. (2011). Understanding and treating insomnia. *Annual Review of Clinical Psychology, 7*(1), 435–458. Retrieved from. https://doi.org/10.1146/annurev.clinpsy.3.022806.091516

Bootzin, R. R., & Perlis, M. L. (2011). Chapter 2 – Stimulus control therapy. In M. Perlis, M. Aloia, & B. Kuhn (Eds.), *Behavioral treatments for sleep disorders* (pp. 21–30). San Diego: Academic Press.

Borbely, A. A., Daan, S., Wirz-Justice, A., & Deboer, T. (2016). The two-process model of sleep regulation: A reappraisal. *Journal of Sleep Research, 25*, 131–143.

Bowden, A., Lorenc, A., & Robinson, N. (2012). Autogenic training as a behavioural approach to insomnia: A prospective cohort study. *Primary Health Care Research & Development, 13*(2), 175–185. Retrieved from https://www.cambridge.org/core/article/autogenic-training-as-a-behavioural-approach-to-insomnia-a-prospective-cohort-study/6844B1C62564AED69BCD89A4995070C5. https://doi.org/10.1017/S1463423611000181

Bruni, O., Alonso-Alconada, D., Besag, F., Biran, V., Braam, W., Cortese, S., … Curatolo, P. (2015). Current role of melatonin in pediatric neurology: Clinical recommendations. *European Journal of Paediatric Neurology, 19*(2), 122–133. Retrieved from http://www.sciencedirect.com/science/article/pii/S1090379814002086. https://doi.org/10.1016/j.ejpn.2014.12.007

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. https://doi.org/10.1080/19315864.2011.651682

Buman, M. P., Phillips, B. A., Youngstedt, S. D., Kline, C. E., & Hirshkowitz, M. (2014). Does nighttime exercise really disturb sleep? Results from the 2013 National Sleep Foundation Sleep in America Poll. *Sleep Medicine, 15*(7), 755–761. Retrieved from http://www.sciencedirect.com/science/article/pii/S1389945714000458. https://doi.org/10.1016/j.sleep.2014.01.008

Burgess, H. J., & Molina, T. A. (2014). Home lighting before usual bedtime impacts circadian timing: A field study. *Photochemistry and Photobiology, 90*(3), 723–726. https://doi.org/10.1111/php.12241

Buysse, D. J., Germain, A., Hall, M., Monk, T. H., & Nofzinger, E. A. (2011). A neurobiological model of insomnia. *Drug Discovery Today: Disease Models, 8*(4), 129–137. https://doi.org/10.1016/j.ddmod.2011.07.002

Caddick, Z. A., Gregory, K., Arsintescu, L., & Flynn-Evans, E. E. (2018). A review of the environmental parameters necessary for an optimal sleep environment. *Building and Environment, 132*, 11–20. Retrieved from http://www.sciencedirect.com/science/article/pii/S0360132318300325. https://doi.org/10.1016/j.buildenv.2018.01.020

Capezuti, E., Sagha Zadeh, R., Pain, K., Basara, A., Jiang, N. Z., & Krieger, A. C. (2018). A systematic review of non-pharmacological interventions to improve nighttime sleep among residents of long-term care settings. *BMC Geriatrics, 18*(1), 143. https://doi.org/10.1186/s12877-018-0794-3

Carroll-Chapman, S. L., & Wu, L.-T. (2012). Substance abuse among individuals with intellectual disabilities. *Research in Developmental Disabilities, 33*(4), 1147–1156. Retrieved from http://www.sciencedirect.com/science/article/pii/S0891422212000376. https://doi.org/10.1016/j.ridd.2012.02.009

CDC. (2016). *Tips for better sleep*. Retrieved from https://www.cdc.gov/sleep/about_sleep/sleep_hygiene.html

Cerezo, A. B., Leal, A., Alvarez-Fernandez, M. A., Hornedo-Ortega, R., Troncoso, A. M., & Garcia-Parrilla, M. C. (2016). Quality control and determi-

nation of melatonin in food supplements. *Journal of Food Composition and Analysis, 45*, 80–86. Retrieved from Go to ISI:WOS:000366536200011. https://doi.org/10.1016/j.jfca.2015.09.013

Cerqueira, D., Carvalho, F., & Melo, R. B. (2018). *Is it smart to use smartphones to measure illuminance for occupational health and safety purposes?* Paper presented at the Advances in Safety Management and Human Factors, Cham.

Chennaoui, M., Arnal, P. J., Sauvet, F., & Léger, D. (2015). Sleep and exercise: A reciprocal issue? *Sleep Medicine Reviews, 20*, 59–72. Retrieved from http://www.sciencedirect.com/science/article/pii/S1087079214000720. https://doi.org/10.1016/j.smrv.2014.06.008

Cheung, J. M. Y., Jarrin, D. C., Ballot, O., Bharwani, A., & Morin, C. M. (2019). A systematic review of cognitive behavioral therapy for insomnia implemented in primary care and community settings. *Sleep Medicine Reviews, 44*, 23–36. Retrieved from http://www.sciencedirect.com/science/article/pii/S1087079217302046. https://doi.org/10.1016/j.smrv.2018.11.001

Cho, C.-H., Lee, H.-J., Yoon, H.-K., Kang, S.-G., Bok, K.-N., Jung, K.-Y., … Lee, E.-I. (2016). Exposure to dim artificial light at night increases REM sleep and awakenings in humans. *Chronobiology International, 33*(1), 117–123. https://doi.org/10.3109/07420528.2015.1108980

Chung, K.-F., Lee, C.-T., Yeung, W.-F., Chan, M.-S., Chung, E. W.-Y., & Lin, W.-L. (2018). Sleep hygiene education as a treatment of insomnia: A systematic review and meta-analysis. *Family Practice, 35*(4), 365–375. https://doi.org/10.1093/fampra/cmx122

Cipolla-Neto, J., & Amaral, F. G. D. (2018). Melatonin as a hormone: New physiological and clinical insights. *Endocrine Reviews, 39*(6), 990–1028. https://doi.org/10.1210/er.2018-00084

Clarke, T. C., Black, L. I., Stussman, B. J., Barnes, P. M., & Nahin, R. L. (2015). Trends in the use of complementary health approaches among adults: United States, 2002–2012. *National Health Statistics Reports, 10*(79), 1–16. Retrieved from https://www.ncbi.nlm.nih.gov/pubmed/25671660. Retrieved from https://www.ncbi.nlm.nih.gov/pmc/PMC4573565/.

Cohrs, S., Rodenbeck, A., Riemann, D., Szagun, B., Jaehne, A., Brinkmeyer, J., … Winterer, G. (2014). Impaired sleep quality and sleep duration in smokers—Results from the German Multicenter Study on Nicotine Dependence. *Addiction Biology, 19*(3), 486–496. https://doi.org/10.1111/j.1369-1600.2012.00487.x

Cooke, B. K., Delalot, D., & Werner, T. L. (2015). Hall v. florida: Capital punishment, IQ, and persons with intellectual disabilities. *Journal of the American Academy of Psychiatry and the Law, 43*(2), 230–234. Retrieved from Go to ISI: WOS:000356175700015.

Cooper, S.-A., Hughes-McCormack, L., Greenlaw, N., McConnachie, A., Allan, L., Baltzer, M., … Morrison, J. (2018). Management and prevalence of long-term conditions in primary health care for adults with intellectual disabilities compared with the general population: A population-based cohort study. *Journal of Applied Research in Intellectual Disabilities, 31*(S1), 68–81. https://doi.org/10.1111/jar.12386

Corkum, P., Davidson, F. D., Tan-MacNeill, K., & Weiss, S. K. (2014). Sleep in children with neurodevelopmental disorders: A focus on insomnia in children with ADHD and ASD. *Sleep Medicine Clinics, 9*(2), 149–168.

Cramer, H., Lauche, R., Langhorst, J., Dobos, G., & Paul, A. (2013). Characteristics of patients with internal diseases who use relaxation techniques as a coping strategy. *Complementary Therapies in Medicine, 21*(5), 481–486. Retrieved from http://www.sciencedirect.com/science/article/pii/S0965229913001234. https://doi.org/10.1016/j.ctim.2013.08.001

Culpepper, L., & Wingertzahn, M. A. (2015). Over-the-counter agents for the treatment of occasional disturbed sleep or transient insomnia: A systematic review of efficacy and safety. *The Primary Care Companion for CNS Disorders, 17*(6). https://doi.org/10.4088/PCC.4015r01798. Retrieved from https://www.ncbi.nlm.nih.gov/pubmed/27057416. Retrieved from https://www.ncbi.nlm.nih.gov/pmc/PMC4805417/

D'Aurea, C. V., Passos, G. S., Santana, M. G., Youngstedt, S. D., Poyares, D., De Souza, A. A., … De Mello, M. T. (2018). 0383 effects of resistance exercise and stretching on sleep of patients with chronic insomnia. *Sleep, 41*(suppl_1), A146–A146. https://doi.org/10.1093/sleep/zsy061.382

Dauvilliers, Y., Schenck, C. H., Postuma, R. B., Iranzo, A., Luppi, P.-H., Plazzi, G., … Boeve, B. (2018). REM sleep behaviour disorder. *Nature Reviews Disease Primers, 4*(1), 19. https://doi.org/10.1038/s41572-018-0016-5

Deliens, G., Leproult, R., Schmitz, R., Destrebecqz, A., & Peigneux, P. (2015). Sleep disturbances in autism spectrum disorders. *Review Journal of Autism and Developmental Disorders, 2*(4), 343–356. https://doi.org/10.1007/s40489-015-0057-6

Dodds, K. L., Miller, C. B., Kyle, S. D., Marshall, N. S., & Gordon, C. J. (2017). Heart rate variability in insomnia patients: A critical review of the literature. *Sleep Medicine Reviews, 33*, 88–100. Retrieved from http://www.sciencedirect.com/science/article/pii/S1087079216300533. https://doi.org/10.1016/j.smrv.2016.06.004

Doghramji, K., & Jangro, W. C. (2016). Adverse effects of psychotropic medications on sleep. *Sleep Medicine Clinics, 11*(4), 503–514. Retrieved from http://www.sciencedirect.com/science/article/pii/S1556407X16300637. https://doi.org/10.1016/j.jsmc.2016.08.001

Drake, C., Roehrs, T., Shambroom, J., & Roth, T. (2013). Caffeine effects on sleep taken 0, 3, or 6 hours before going to bed. *Journal of Clinical Sleep Medicine: Official Publication of the American Academy of Sleep Medicine, 9*(11), 1195–1200. Retrieved from https://www.ncbi.nlm.nih.gov/pubmed/24235903. Retrieved from https://www.ncbi.nlm.nih.gov/pmc/PMC3805807/. https://doi.org/10.5664/jcsm.3170

Dutt, R., Roduta-Roberts, M., & Brown, A. C. (2015). Sleep and children with cerebral palsy: A review of current evidence and environmental non-pharmacological interventions. *Children, 2*(1), 78–88. https://doi.org/10.3390/children2010078

Erland, L. A., & Saxena, P. K. (2017). Melatonin natural health products and supplements: Presence of serotonin and significant variability of melatonin content. *Journal of Clinical Sleep Medicine, 13*(2), 275–281. Retrieved from https://www.ncbi.nlm.nih.gov/pubmed/27855744. https://doi.org/10.5664/jcsm.6462

Esbensen, A. J. (2016). Sleep problems and associated comorbidities among adults with Down syndrome. *Journal of Intellectual Disability Research, 60*(1), 68–79. https://doi.org/10.1111/jir.12236

Esbensen, A. J., & Schwichtenberg, A. J. (2016). Sleep in neurodevelopmental disorders. *International Review of Research in Developmental Disabilities, 51*, 153–191. Retrieved from https://www.ncbi.nlm.nih.gov/pubmed/28503406. https://doi.org/10.1016/bs.irrdd.2016.07.005

European Medicines Agency. (2018, 10/10/2018). *Slenyto*. Retrieved from https://www.ema.europa.eu/en/medicines/human/EPAR/slenyto.

Faraut, B., Andrillon, T., Vecchierini, M.-F., & Leger, D. (2017). Napping: A public health issue. From epidemiological to laboratory studies. *Sleep Medicine Reviews, 35*, 85–100. Retrieved from http://www.sciencedirect.com/science/article/pii/S1087079216300946. https://doi.org/10.1016/j.smrv.2016.09.002

Faraut, B., Léger, D., Medkour, T., Dubois, A., Bayon, V., Chennaoui, M., & Perrot, S. (2015). Napping reverses increased pain sensitivity due to sleep restriction. *PLoS One, 10*(2), e0117425. https://doi.org/10.1371/journal.pone.0117425

Ford, E. S., Wheaton, A. G., Cunningham, T. J., Giles, W. H., Chapman, D. P., & Croft, J. B. (2014). Trends in outpatient visits for insomnia, sleep apnea, and prescriptions for sleep medications among US adults: Findings from the National Ambulatory Medical Care Survey 1999–2010. *Sleep, 37*(8), 1283–1293. Retrieved from. https://doi.org/10.5665/sleep.3914

Frank, N. C., Spirito, A., Stark, L., & Owens-Stively, J. (1997). The use of scheduled awakenings to eliminate childhood sleepwalking. *Journal of Pediatric Psychology, 22*(3), 345–353. Retrieved from. https://doi.org/10.1093/jpepsy/22.3.345

Galbiati, A., Rinaldi, F., Giora, E., Ferini-Strambi, L., & Marelli, S. (2015). Behavioural and cognitive-behavioural treatments of parasomnias. *Behavioural Neurology, 2015*(1), 786928–786928.

Garcia, M. J., McPherson, P., Patel, S. Y., & Burns, C. O. (2017). Diet and supplementation targeted for autism spectrum disorder. In J. L. Matson (Ed.), *Handbook of treatments for autism spectrum disorder* (pp. 397–425). Cham, Switzerland: Springer International Publishing.

Garre-Olmo, J., López-Pousa, S., Turon-Estrada, A., Juvinyà, D., Ballester, D., & Vilalta-Franch, J. (2012). Environmental determinants of quality of life in nursing home residents with severe dementia. *Journal of the American Geriatrics Society, 60*(7), 1230–1236. Retrieved from. https://doi.org/10.1111/j.1532-5415.2012.04040.x

Gedye, A. (1995). *Dementia scale for Down syndrome: Manual*. Vancouver: C. B. A. Gedye.

Ghiassi, R., Murphy, K., Cummin, A. R., & Partridge, M. R. (2011). Developing a pictorial Epworth Sleepiness Scale. *Thorax, 66*(2), 97. Retrieved from http://thorax.bmj.com/content/66/2/97.abstract. https://doi.org/10.1136/thx.2010.136879

Gilderthorp, R. C. (2014). Is EMDR an effective treatment for people diagnosed with both intellectual disability and post-traumatic stress disorder? *Journal of Intellectual Disabilities, 19*(1), 58–68. Retrieved from. https://doi.org/10.1177/1744629514560638

Gong, H., Ni, C.-X., Liu, Y.-Z., Zhang, Y., Su, W.-J., Lian, Y.-J., … Jiang, C.-L. (2016). Mindfulness meditation for insomnia: A meta-analysis of randomized controlled trials. *Journal of Psychosomatic Research, 89*, 1–6. Retrieved from http://www.sciencedirect.com/science/article/pii/S0022399916303579. https://doi.org/10.1016/j.jpsychores.2016.07.016

Grigg-Damberger, M., & Ralls, F. (2013). Treatment strategies for complex behavioral insomnia in children with neurodevelopmental disorders. *Current Opinion in Pulmonary Medicine, 19*(6), 616–625.

Gringras, P., Green, D., Wright, B., Rush, C., Sparrowhawk, M., Pratt, K., … Zaiwalla, Z. (2014). Weighted blankets and sleep in autistic children—A randomized controlled trial. *Pediatrics, 134*(2), 298–306.

Gringras, P., Nir, T., Breddy, J., Frydman-Marom, A., & Findling, R. L. (2017). Efficacy and safety of pediatric prolonged-release melatonin for insomnia in children with autism spectrum disorder. *Journal of the American Academy of Child and Adolescent Psychiatry, 56*(11), 948–957. e944. https://doi.org/10.1016/j.jaac.2017.09.414

Groden, J., Weidenman, L., & Diller, A. (2016). *Relaxation: A comprehensive manual for children and adults with autism and other developmental disabilities*. Champaign, IL: Research Press.

Gunning, M. J., & Espie, C. A. (2003). Psychological treatment of reported sleep disorder in adults with intellectual disability using a multiple baseline design. *Journal of Intellectual Disability Research, 47*(3), 191–202. Retrieved from. https://doi.org/10.1046/j.1365-2788.2003.00461.x

Gutman, S. A., Gregory, K. A., Sadlier-Brown, M. M., Schlissel, M. A., Schubert, A. M., Westover, L. A., & Miller, R. C. (2017). Comparative effectiveness of three occupational therapy sleep interventions: A randomized controlled study. *OTJR: Occupation, Participation and Health, 37*(1), 5–13. Retrieved from http://europepmc.org/abstract/MED/27760887. https://doi.org/10.1177/1539449216673045

Halperin, D. (2014). Environmental noise and sleep disturbances: A threat to health? *Sleep Science, 7*(4), 209–212. Retrieved from http://www.sciencedirect.

com/science/article/pii/S1984006314000601. https://doi.org/10.1016/j.slsci.2014.11.003

Hanley, G. P. (2005). Sleep assessment and treatment tool [Measurement Instrument].

Harris, J., Lack, L., Kemp, K., Wright, H., & Bootzin, R. (2012). A randomized controlled trial of intensive sleep retraining (ISR): A brief conditioning treatment for chronic insomnia. *Sleep, 35*(1), 49–60. Retrieved from. https://doi.org/10.5665/sleep.1584

Harris, J., Lack, L., Wright, H., Gradisar, M., & Brooks, A. (2007). Intensive Sleep Retraining treatment for chronic primary insomnia: A preliminary investigation. *Journal of Sleep Research, 16*(3), 276–284. Retrieved from. https://doi.org/10.1111/j.1365-2869.2007.00595.x

Harvey, A. G. (2002). A cognitive model of insomnia. *Behaviour Research and Therapy, 40*(8), 869–893.

Harvey, A. G., & Tang, N. K. Y. (2012). (Mis)perception of sleep in insomnia: A puzzle and a resolution. *Psychological Bulletin, 138*(1), 77–101. Retrieved from https://www.ncbi.nlm.nih.gov/pubmed/21967449. Retrieved from https://www.ncbi.nlm.nih.gov/pmc/PMC3277880/. https://doi.org/10.1037/a0025730

Heussler, H. S. (2016). Management of sleep disorders in neurodevelopmental disorders and genetic syndromes. *Current Opinion in Psychiatry, 29*(2), 138–143.

Hill, E. E., Zack, E., Battaglini, C., Viru, M., Viru, A., & Hackney, A. C. (2008). Exercise and circulating cortisol levels: The intensity threshold effect. *Journal of Endocrinological Investigation, 31*(7), 587–591. https://doi.org/10.1007/bf03345606

Hiller, R. M., Johnston, A., Dohnt, H., Lovato, N., & Gradisar, M. (2015). Assessing cognitive processes related to insomnia: A review and measurement guide for Harvey's cognitive model for the maintenance of insomnia. *Sleep Medicine Reviews, 23*, 46–53. https://doi.org/10.1016/j.smrv.2014.11.006

Hiller, R. M., Lovato, N., Gradisar, M., Oliver, M., & Slater, A. (2014). Trying to fall asleep while catastrophising: What sleep-disordered adolescents think and feel. *Sleep Medicine, 15*(1), 96–103. https://doi.org/10.1016/j.sleep.2013.09.014

Hirshkowitz, M., Whiton, K., Albert, S. M., Alessi, C., Bruni, O., DonCarlos, L., … Adams Hillard, P. J. (2015). National Sleep Foundation's sleep time duration recommendations: Methodology and results summary. *Sleep Health, 1*(1), 40–43. Retrieved from http://www.sciencedirect.com/science/article/pii/S2352721815000157. https://doi.org/10.1016/j.sleh.2014.12.010

Ho, E., & Siu, A. (2018). Occupational therapy practice in sleep management: A review of conceptual models and research evidence. *Occupational Therapy International, 2018*, 1–12.

Honaker, S. M., & Meltzer, L. J. (2016). Sleep in pediatric primary care: A review of the literature. *Sleep Medicine Reviews, 25*, 31–39. Retrieved from http://www.sciencedirect.com/science/article/pii/S1087079215000167. https://doi.org/10.1016/j.smrv.2015.01.004

Hwang, E., & Shin, S. (2016). Effectiveness of non-pharmacological intervention for insomnia: A systematic review and meta-analysis. *Indian Journal of Science and Technology, 9*(9), 1–9.

Hylkema, T., Petitiaux, W., & Vlaskamp, C. (2011). Utility of staff training on correcting sleep problems in people with intellectual disabilities living in residential settings. *Journal of Policy and Practice in Intellectual Disabilities, 8*(2), 85–91. Retrieved from. https://doi.org/10.1111/j.1741-1130.2011.00294.x

Hylkema, T., & Vlaskamp, C. (2009). Significant improvement in sleep in people with intellectual disabilities living in residential settings by non-pharmaceutical interventions. *Journal of Intellectual Disability Research, 53*(8), 695–703. https://doi.org/10.1111/j.1365-2788.2009.01177.x

Irish, L. A., Kline, C. E., Gunn, H. E., Buysse, D. J., & Hall, M. H. (2015). The role of sleep hygiene in promoting public health: A review of empirical evidence. *Sleep Medicine Reviews, 22*, 23–36. Retrieved from http://www.sciencedirect.com/science/article/pii/S1087079214001002. https://doi.org/10.1016/j.smrv.2014.10.001

Jacobson, E. (1929). *Progressive relaxation a physiological and clinical investigation of muscular states and their significance in psychology and medical practice*. Chicago & London: The University of Chicago Press.

Jaehne, A., Unbehaun, T., Feige, B., Cohrs, S., Rodenbeck, A., Schütz, A.-L., … Riemann, D. (2015). Sleep changes in smokers before, during and 3 months after nicotine withdrawal. *Addiction Biology, 20*(4), 747–755. Retrieved from. https://doi.org/10.1111/adb.12151

Jang, J., Matson, J. L., Williams, L. W., Tureck, K., Goldin, R. L., & Cervantes, P. E. (2013). Rates of comorbid symptoms in children with ASD, ADHD, and comorbid ASD and ADHD. *Research in Developmental Disabilities, 34*(8), 2369–2378. Retrieved from https://www.ncbi.nlm.nih.gov/pubmed/23708709. https://doi.org/10.1016/j.ridd.2013.04.021

Jennings, C., & Hewitt, O. (2015). The use of cognitive behaviour therapy to treat depression in people with learning disabilities: A systematic review. *Tizard Learning Disability Review, 20*(2), 54–64.

Jin, C. S., Hanley, G. P., & Beaulieu, L. (2013). An individualized and comprehensive approach to treating sleep problems in young children. *Journal of Applied Behavior Analysis, 46*(1), 161–180. Retrieved from. https://doi.org/10.1002/jaba.16

Johns, M. W. (1991). A new method for measuring daytime sleepiness: The Epworth sleepiness scale. *Sleep, 14*(6), 540–545.

Johns, M. W., Tucker, A. J., Chapman, R. J., Michael, N. J., Beale, C. A., & Stephens, M. N. (2006). A new scale of drowsiness based on multiple characteristics of eye and eyelid movements: The Johns Drowsiness Scale. *Sleep & Biological Rhythms, 4*, A37–A38.

Johnson, C. R., Turner, K. S., Foldes, E., Brooks, M. M., Kronk, R., & Wiggs, L. (2013). Behavioral parent training to address sleep disturbances in young chil-

dren with autism spectrum disorder: A pilot trial. *Sleep Medicine, 14*(10), 995–1004. https://doi.org/10.1016/j.sleep.2013.05.013

Joseph, A., Choi, Y.-S., & Quan, X. (2015). Impact of the physical environment of residential health, care, and support facilities (RHCSF) on staff and residents: A systematic review of the literature. *Environment and Behavior, 48*(10), 1203–1241. Retrieved from. https://doi.org/10.1177/0013916515597027

Kardous, C. A., & Shaw, P. B. (2014). Evaluation of smartphone sound measurement applications. *The Journal of the Acoustical Society of America, 135*(4), EL186–EL192. Retrieved from. https://doi.org/10.1121/1.4865269

Kay, D. B., Karim, H. T., Soehner, A. M., Hasler, B. P., James, J. A., Germain, A., … Buysse, D. J. (2017). Subjective-objective sleep discrepancy is associated with alterations in regional glucose metabolism in patients with insomnia and good sleeper controls. *Sleep, 40*(11), zsx155. https://doi.org/10.1093/sleep/zsx155

Kim, J. H., & Duffy, J. F. (2018). Circadian rhythm sleep-wake disorders in older adults. *Sleep Medicine Clinics, 13*, 39–50.

Kinsey, A. W., & Ormsbee, M. J. (2015). The health impact of nighttime eating: Old and new perspectives. *Nutrients, 7*(4), 2648–2662. Retrieved from https://www.ncbi.nlm.nih.gov/pubmed/25859885. Retrieved from https://www.ncbi.nlm.nih.gov/pmc/PMC4425165/. https://doi.org/10.3390/nu7042648

Kitsaras, G., Goodwin, M., Allan, J., Kelly, M. P., & Pretty, I. A. (2018). Bedtime routines child well-being & development. *BMC Public Health, 18*(1), 386–386. Retrieved from https://www.ncbi.nlm.nih.gov/pubmed/29562892. Retrieved from https://www.ncbi.nlm.nih.gov/pmc/PMC5861615/. https://doi.org/10.1186/s12889-018-5290-3

Köse, S., Yılmaz, H., Ocakoğlu, F. T., & Özbaran, N. B. (2017). Sleep problems in children with autism spectrum disorder and intellectual disability without autism spectrum disorder. *Sleep Medicine, 40*, 69–77. Retrieved from http://www.sciencedirect.com/science/article/pii/S1389945717303842. https://doi.org/10.1016/j.sleep.2017.09.021

Krahn, G. L., Walker, D. K., & Correa-De-Araujo, R. (2015). Persons with disabilities as an unrecognized health disparity population. *American Journal of Public Health, 105*(S2), S198–S206. Retrieved from. https://doi.org/10.2105/AJPH.2014.302182

Kräuchi, K. (2007). The thermophysiological cascade leading to sleep initiation in relation to phase of entrainment. *Sleep Medicine Reviews, 11*(6), 439–451. Retrieved from http://www.sciencedirect.com/science/article/pii/S1087079207000950. https://doi.org/10.1016/j.smrv.2007.07.001

Kyle, S. D., Aquino, M. R. J., Miller, C. B., Henry, A. L., Crawford, M. R., Espie, C. A., & Spielman, A. J. (2015). Towards standardisation and improved understanding of sleep restriction therapy for insomnia disorder: A systematic examination of CBT-I trial content. *Sleep Medicine Reviews, 23*, 83–88. Retrieved from http://www.sciencedirect.com/science/article/pii/S1087079215000301. https://doi.org/10.1016/j.smrv.2015.02.003

Kyle, S. D., Morgan, K., Spiegelhalder, K., & Espie, C. A. (2011). No pain, no gain: An exploratory within-subjects mixed-methods evaluation of the patient experience of sleep restriction therapy (SRT) for insomnia. *Sleep Medicine, 12*(8), 735–747. Retrieved from http://www.sciencedirect.com/science/article/pii/S1389945711002061. https://doi.org/10.1016/j.sleep.2011.03.016

Lack, L., Scott, H., Micic, G., & Lovato, N. (2017). Intensive sleep re-training: From bench to bedside. *Brain Sciences, 7*(4), 33. Retrieved from https://www.ncbi.nlm.nih.gov/pubmed/28346384. Retrieved from https://www.ncbi.nlm.nih.gov/pmc/PMC5406690/. https://doi.org/10.3390/brainsci7040033

Lau, W. K. W., Leung, M.-K., Wing, Y.-K., & Lee, T. M. C. (2018). Potential mechanisms of mindfulness in improving sleep and distress. *Mindfulness, 9*(2), 547–555. Retrieved from. https://doi.org/10.1007/s12671-017-0796-9

Lee, D., Morgan, K., & Lindesay, J. (2007). Effect $$. *Journal of the American Geriatrics Society, 55*(2), 252–258. Retrieved from. https://doi.org/10.1111/j.1532-5415.2007.01036.x

Lelis, A. L., Cardoso, M. V., & Hall, W. A. (2016). Sleep disorders in children with cerebral palsy: An integrative review. *Sleep Medicine Reviews, 30*, 63–71. https://doi.org/10.1016/j.smrv.2015.11.008

Levenson, J. C., Kay, D. B., & Buysse, D. J. (2015). The pathophysiology of insomnia. *Chest, 147*(4), 1179–1192. Retrieved from https://www.ncbi.nlm.nih.gov/pubmed/25846534. Retrieved from https://www.ncbi.nlm.nih.gov/pmc/PMC4388122/. https://doi.org/10.1378/chest.14-1617

Licis, A. K., Vallorani, A., Gao, F., Chen, C., Lenox, J., Yamada, K. A., … Gutmann, D. H. (2013). Prevalence of sleep disturbances in children with neurofibromatosis type 1. *Journal of Child Neurology, 28*(11), 1400–1405. Retrieved from https://www.ncbi.nlm.nih.gov/pubmed/24065580. Retrieved from https://www.ncbi.nlm.nih.gov/pmc/PMC3805763/. https://doi.org/10.1177/0883073813500849

Lillehei, A. S., & Halcon, L. L. (2014). A systematic review of the effect of inhaled essential oils on sleep. *The Journal of Alternative and Complementary Medicine, 20*(6), 441–451. Retrieved from. https://doi.org/10.1089/acm.2013.0311

Lindsay, E. K., & Creswell, J. D. (2017). Mechanisms of mindfulness training: Monitor and Acceptance Theory (MAT). *Clinical Psychology Review, 51*, 48–59. Retrieved from http://www.sciencedirect.com/science/article/pii/S0272735816302008. https://doi.org/10.1016/j.cpr.2016.10.011

Lotan, M., & Gold, C. (2009). Meta-analysis of the effectiveness of individual intervention in the controlled multisensory environment (Snoezelen®) for individuals with intellectual disability. *Journal*

of Intellectual & Developmental Disability, 34(3), 207–215. Retrieved from. https://doi.org/10.1080/13668250903080106

Lowe, H., Haddock, G., Mulligan, L. D., Gregg, L., Carter, L.-A., Fuzellier-Hart, A., & Kyle, S. D. (2018). Does exercise improve sleep for adults with insomnia? A systematic review with quality appraisal. *Clinical Psychology Review, 68*, 1–12. Retrieved from http://www.sciencedirect.com/science/article/pii/S0272735817303306. https://doi.org/10.1016/j.cpr.2018.11.002

Maldonado, C. C., Bentley, A. J., & Mitchell, D. (2004). A pictorial sleepiness scale based on cartoon faces. *Sleep, 27*(3), 541–548. Retrieved from. https://doi.org/10.1093/sleep/27.3.541

Malow, B. A., Byars, K., Johnson, K., Weiss, S., Bernal, P., Goldman, S. E., … Glaze, D. G. (2012). A practice pathway for the identification, evaluation, and management of insomnia in children and adolescents with autism spectrum disorders. *Pediatrics, 130*, S106–S124. Retrieved from Go to ISI: WOS:000209485500009. https://doi.org/10.1542/peds.2012-0900I

Malow, B. A., Katz, T., Reynolds, A. M., Shui, A., Carno, M., Connolly, H. V., … Bennett, A. E. (2016). Sleep difficulties and medications in children with autism spectrum disorders: A registry study. *Pediatrics, 137*(Suppl 2), S98–s104. https://doi.org/10.1542/peds.2015-2851H

Manber, R., Carney, C., Edinger, J., Epstein, D., Friedman, L., Haynes, P. L., … Trockel, M. (2012). Dissemination of CBTI to the non-sleep specialist: Protocol development and training issues. *Journal of Clinical Sleep Medicine: Official Publication of the American Academy of Sleep Medicine, 8*(2), 209–218. Retrieved from https://www.ncbi.nlm.nih.gov/pubmed/22505869. Retrieved from https://www.ncbi.nlm.nih.gov/pmc/PMC3311421/. https://doi.org/10.5664/jcsm.1786

Mann, D. M. A., Davidson, Y. S., Robinson, A. C., Allen, N., Hashimoto, T., Richardson, A., … Potier, M.-C. (2018). Patterns and severity of vascular amyloid in Alzheimer's disease associated with duplications and missense mutations in APP gene, Down syndrome and sporadic Alzheimer's disease. *Acta Neuropathologica, 136*(4), 569–587.

Markwald, R. R., Iftikhar, I., & Youngstedt, S. D. (2018). Behavioral strategies, including exercise, for addressing insomnia. *ACSM's Health & Fitness Journal, 22*(2), 23. Retrieved from https://journals.lww.com/acsm-healthfitness/Fulltext/2018/03000/BEHAVIORAL_STRATEGIES,_INCLUDING_EXERCISE,_FOR.8.aspx.

Maurer, L. F., Espie, C. A., & Kyle, S. D. (2018). How does sleep restriction therapy for insomnia work? A systematic review of mechanistic evidence and the introduction of the Triple-R model. *Sleep Medicine Reviews, 42*, 127–138. Retrieved from http://www.sciencedirect.com/science/article/pii/S1087079218300790. https://doi.org/10.1016/j.smrv.2018.07.005

Mazurek, M. O., & Petroski, G. F. (2015). Sleep problems in children with autism spectrum disorder: Examining the contributions of sensory over-responsivity and anxiety. *Sleep Medicine, 16*(2), 270–279. Retrieved from http://www.sciencedirect.com/science/article/pii/S1389945714004705. https://doi.org/10.1016/j.sleep.2014.11.006

McCurry, S. M., LaFazia, D. M., Pike, K. C., Logsdon, R. G., & Teri, L. (2009). Managing sleep disturbances in adult family homes: Recruitment and implementation of a behavioral treatment program. *Geriatric Nursing, 30*(1), 36–44. Retrieved from http://www.sciencedirect.com/science/article/pii/S0197457208001729. https://doi.org/10.1016/j.gerinurse.2008.05.001

McCurry, S. M., LaFazia, D. M., Pike, K. C., Logsdon, R. G., & Teri, L. (2012). Development and evaluation of a sleep education program for older adults with dementia living in adult family homes. *The American Journal of Geriatric Psychiatry, 20*(6), 494–504. Retrieved from http://www.sciencedirect.com/science/article/pii/S1064748112620636. https://doi.org/10.1097/JGP.0b013e318248ae79

McCurry, S. M., Song, Y., & Martin, J. L. (2015). Sleep in caregivers: What we know and what we need to learn. *Current Opinion in Psychiatry, 28*(6), 497–503. Retrieved from https://journals.lww.com/co-psychiatry/Fulltext/2015/11000/Sleep_in_caregivers___what_we_know_and_what_we.18.aspx.

McFeeters, S., Pront, L., Cuthbertson, L., & King, L. (2016). Massage, a complementary therapy effectively promoting the health and well-being of older people in residential care settings: A review of the literature. *International Journal of Older People Nursing, 11*(4), 266–283. Retrieved from. https://doi.org/10.1111/opn.12115

McGrane, I. R., Leung, J. G., St. Louis, E. K., & Boeve, B. F. (2015). Melatonin therapy for REM sleep behavior disorder: A critical review of evidence. *Sleep Medicine, 16*(1), 19–26. Retrieved from http://www.sciencedirect.com/science/article/pii/S1389945714004122. https://doi.org/10.1016/j.sleep.2014.09.011

McLay, L. K., France, K. G., Knight, J., Blampied, N. M., & Hastie, B. (2018). The effectiveness of function-based interventions to treat sleep problems, including unwanted co-sleeping, in children with autism. *Behavioral Interventions, 34*(1), 30–51. Retrieved from. https://doi.org/10.1002/bin.1651

McLay, L.-L. K., & France, K. (2016). Empirical research evaluating non-traditional approaches to managing sleep problems in children with autism. *Developmental Neurorehabilitation, 19*(2), 123–134. Retrieved from. https://doi.org/10.3109/17518423.2014.904452

Meltzer, L. J. (2010). Clinical management of behavioral insomnia of childhood: Treatment of bedtime problems and night wakings in young children. *Behavioral Sleep Medicine, 8*(3), 172–189.

Meltzer, L. J. (2017). Future directions in sleep and developmental psychopathology. *Journal of Clinical Child*

& Adolescent Psychology, 46(2), 295–301. Retrieved from http://search.ebscohost.com/login.aspx?direct=true&db=pbh&AN=121550202&site=ehost-live. https://doi.org/10.1080/15374416.2016.1236727

Micsinszki, S. K., Ballantyne, M., Cleverley, K., Green, P., & Stremler, R. (2018). Sleep outcomes for parents of children with neurodevelopmental disabilities: A systematic review. *Journal of Family Nursing, 24*(2), 217–249. Retrieved from. https://doi.org/10.1177/1074840718773381

Miller, C. B., Espie, C. A., Epstein, D. R., Friedman, L., Morin, C. M., Pigeon, W. R., … Kyle, S. D. (2014). The evidence base of sleep restriction therapy for treating insomnia disorder. *Sleep Medicine Reviews, 18*(5), 415–424. Retrieved from http://www.sciencedirect.com/science/article/pii/S1087079214000161. https://doi.org/10.1016/j.smrv.2014.01.006

Mindell, J. A., Li, A. M., Sadeh, A., Kwon, R., & Goh, D. Y. T. (2015). Bedtime routines for young children: A dose-dependent association with sleep outcomes. *Sleep, 38*(5), 717–722. Retrieved from https://www.ncbi.nlm.nih.gov/pubmed/25325483. Retrieved from https://www.ncbi.nlm.nih.gov/pmc/PMC4402657/. https://doi.org/10.5665/sleep.4662

Mol, E. M., Monbaliu, E., Ven, M., Vergote, M., & Prinzie, P. (2012). The use of night orthoses in cerebral palsy treatment: Sleep disturbance in children and parental burden or not? *Research in Developmental Disabilities, 33*(2), 341–349. https://doi.org/10.1016/j.ridd.2011.10.026

Monk, T. H., Buysse, D. J., Billy, B. D., Fletcher, M. E., Kennedy, K. S., Schlarb, J. E., & Beach, S. R. (2011). Circadian type and bed-timing regularity in 654 retired seniors: Correlations with subjective sleep measures. *Sleep, 34*(2), 235–239. Retrieved from https://www.ncbi.nlm.nih.gov/pubmed/21286245. Retrieved from https://www.ncbi.nlm.nih.gov/pmc/PMC3022945/

Moore, B. A., Friman, P. C., Fruzzetti, A. E., & MacAleese, K. (2007). Brief report: Evaluating the bedtime pass program for child resistance to bedtime—A randomized, controlled trial. *Journal of Pediatric Psychology, 32*(3), 283–287. Retrieved from. https://doi.org/10.1093/jpepsy/jsl025

Morgenthaler, T., Kramer, M., Alessi, C., Friedman, L., Boehlecke, B., Brown, T., … American Academy of Sleep Medicine. (2006). Practice parameters for the psychological and behavioral treatment of insomnia: An update. An american academy of sleep medicine report. *Sleep, 29*(11), 1415–1419. Retrieved from https://www.ncbi.nlm.nih.gov/pubmed/17162987

Morgenthaler, T. I., Auerbach, S., Casey, K. R., Kristo, D., Maganti, R., Ramar, K., … Kartje, R. (2018). Position paper for the treatment of nightmare disorder in adults: An american academy of sleep medicine position paper. *Journal of Clinical Sleep Medicine, 14*(6), 1041–1055. https://doi.org/10.5664/jcsm.7178

Morgenthaler, T. I., Owens, J., & Alessi, C. (2006). Practice parameters for behavioral treatment of bedtime problems and night wakings in infants and young children. *Sleep, 29*(10), 1277–1281.

Morin, C. M. (1996). *Insomnia: Psychological assessment and management*. New York, UK: Guilford Publications.

Morioka, H., Jike, M., Kanda, H., Osaki, Y., Nakagome, S., Otsuka, Y., … Ohida, T. (2018). The association between sleep disturbance and second-hand smoke exposure: A large-scale, nationwide, cross-sectional study of adolescents in Japan. *Sleep Medicine, 50*, 29–35. Retrieved from http://www.sciencedirect.com/science/article/pii/S1389945718302041. https://doi.org/10.1016/j.sleep.2018.04.014

Moss, A. H. B., Gordon, J. E., & O'Connell, A. (2014). Impact of sleepwise: An intervention for youth with developmental disabilities and sleep disturbance. *Journal of Autism and Developmental Disorders, 44*(7), 1695–1707. Retrieved from. https://doi.org/10.1007/s10803-014-2040-y

Murawski, B., Wade, L., Plotnikoff, R. C., Lubans, D. R., & Duncan, M. J. (2018). A systematic review and meta-analysis of cognitive and behavioral interventions to improve sleep health in adults without sleep disorders. *Sleep Medicine Reviews, 40*, 160–169. Retrieved from http://www.sciencedirect.com/science/article/pii/S1087079217301661. https://doi.org/10.1016/j.smrv.2017.12.003

National Center for Complementary and Integrative Health. (2017). Sleep disorders: In *Depth*. NCCIH Pub No. D437.

National Institute of Medicine Committee on Sleep Medicine. (2006). The National Academies Collection: Reports funded by National Institutes of Health. In H. R. Colten & B. M. Altevogt (Eds.), *Sleep disorders and sleep deprivation: An unmet public health problem*. Washington, DC: National Academies Press.

National Science Foundation. (2018). *What is sleep hygiene?* Retrieved from https://www.sleepfoundation.org/sleep-topics/sleep-hygiene.

Ohayon, M. M., & Milesi, C. (2016). Artificial outdoor nighttime lights associate with altered sleep behavior in the American general population. *Sleep, 39*(6), 1311–1320. Retrieved from. https://doi.org/10.5665/sleep.5860

Okamoto-Mizuno, K., & Mizuno, K. (2012). Effects of thermal environment on sleep and circadian rhythm. *Journal of Physiological Anthropology, 31*(1), 14–14. Retrieved from https://www.ncbi.nlm.nih.gov/pubmed/22738673. Retrieved from https://www.ncbi.nlm.nih.gov/pmc/PMC3427038/. https://doi.org/10.1186/1880-6805-31-14

Oviedo, R. G., Travier, N., & Guerra-Balic, M. (2017). Sedentary and physical activity patterns in adults with intellectual disability. *International Journal of Environmental Research and Public Health, 14*(9), 1027. https://doi.org/10.3390/ijerph14091027

Owens, J. A., & Moore, M. (2017). Insomnia in infants and young children. *Pediatric Annals, 46*(9), e321–e326.

Owens, J. A., Rosen, C. L., Mindell, J. A., & Kirchner, H. L. (2010). Use of pharmacotherapy for insomnia in child psychiatry practice: A national survey. *Sleep Medicine, 11*(7), 692–700. Retrieved from

http://www.sciencedirect.com/science/article/pii/S138994571000211X. https://doi.org/10.1016/j.sleep.2009.11.015

Perlis, M. L., Ellis, J. G., Kloss, J. D., & Riemann, D. W. (2017). Chapter 82 etiology and pathophysiology of insomnia. In *Principles and practice of sleep medicine* (pp. 769–784.e764). Philadelphia, PA: Elsevier.

Piazza, C. C., & Fisher, W. (1991). A faded bedtime with response cost protocol for treatment of multiple sleep problems in children. *Journal of Applied Behavior Analysis, 24*(1), 129–140.

Pillai, V., Cheng, P., Kalmbach, D. A., Roehrs, T., Roth, T., & Drake, C. L. (2016). Prevalence and predictors of prescription sleep aid use among individuals with DSM-5 insomnia: The role of hyperarousal. *Sleep, 39*(4), 825–832. Retrieved from. https://doi.org/10.5665/sleep.5636

Priday, L. J., Byrne, C., & Totsika, V. (2017). Behavioural interventions for sleep problems in people with an intellectual disability: A systematic review and meta-analysis of single case and group studies. *Journal of Intellectual Disability Research, 61*(1), 1–15. https://doi.org/10.1111/jir.12265

Qaseem, A., Kansagara, D., Forciea, M. A., Cooke, M., Denberg, T. D., & Clinical Guidelines Committee of the American College of Physicians. (2016). Management of chronic insomnia disorder in adults: A clinical practice guideline from the American college of physicians. *Annals of Internal Medicine, 165*(2), 125–133. Retrieved from https://www.ncbi.nlm.nih.gov/pubmed/27136449. https://doi.org/10.7326/M15-2175

Ramakrishnan, S., Wesensten, N. J., Balkin, T. J., & Reifman, J. (2016). A unified model of performance: Validation of its predictions across different sleep/wake schedules. *Sleep, 39*(1), 249–262. Retrieved from. https://doi.org/10.5665/sleep.5358

Raymann, R. J. E. M., Swaab, D. F., & Van Someren, E. J. W. (2008). Skin deep: Enhanced sleep depth by cutaneous temperature manipulation. *Brain, 131*(2), 500–513. Retrieved from. https://doi.org/10.1093/brain/awm315

Rétey, J. V., Adam, M., Khatami, R., Luhmann, U. F. O., Jung, H. H., Berger, W., & Landolt, H. P. (2007). A genetic variation in the adenosine A2A receptor gene (ADORA2A) contributes to individual sensitivity to caffeine effects on sleep. *Clinical Pharmacology & Therapeutics, 81*(5), 692–698. Retrieved from. https://doi.org/10.1038/sj.clpt.6100102

Richdale, A. L., & Baker, E. K. (2014). Sleep in individuals with an intellectual or developmental disability: Recent research reports. *Current Developmental Disorders Reports, 1*, 74–85. https://doi.org/10.1007/s40474-014-0010-x

Rigney, G., Ali, N. S., Corkum, P. V., Brown, C. A., Constantin, E., Godbout, R., … Weiss, S. K. (2018). A systematic review to explore the feasibility of a behavioural sleep intervention for insomnia in children with neurodevelopmental disorders: A transdiagnostic approach. *Sleep Medicine Reviews, 41*, 244–254. Retrieved from http://www.sciencedirect.com/science/article/pii/S1087079218300017. https://doi.org/10.1016/j.smrv.2018.03.008

Roberts, L., & Kwan, S. (2018). Putting the C into CBT: Cognitive challenging with adults with mild to moderate intellectual disabilities and anxiety disorders. *Clinical Psychology & Psychotherapy, 25*(5), 662–671. Retrieved from. https://doi.org/10.1002/cpp.2196

Robertson, J., Hatton, C., Emerson, E., & Baines, S. (2015). Prevalence of epilepsy among people with intellectual disabilities: A systematic review. *Seizure, 29*, 46–62. Retrieved from http://www.sciencedirect.com/science/article/pii/S1059131115000862. https://doi.org/10.1016/j.seizure.2015.03.016

Rusch, H. L., Rosario, M., Levison, L. M., Olivera, A., Livingston, W. S., Wu, T., & Gill, J. M. (2018). The effect of mindfulness meditation on sleep quality: A systematic review and meta-analysis of randomized controlled trials. *Annals of the New York Academy of Sciences, 1445*(1), 5–16. Retrieved from. https://doi.org/10.1111/nyas.13996

Sateia, M. J., Buysse, D. J., Krystal, A. D., Neubauer, D. N., & Heald, J. L. (2017). Clinical practice guideline for the pharmacologic treatment of chronic insomnia in adults: An american academy of sleep medicine clinical practice guideline. *Journal of Clinical Sleep Medicine, 13*(2), 307–349. Retrieved from https://www.ncbi.nlm.nih.gov/pubmed/27998379. https://doi.org/10.5664/jcsm.6470

Sayorwan, W., Siripornpanich, V., Piriyapunyaporn, T., Hongratanaworakit, T., Kotchabhakdi, N., & Ruangrungsi, N. (2012). The effects of lavender oil inhalation on emotional states, autonomic nervous system, and brain electrical activity. *Journal of the Medical Association of Thailand, 95*(4), 598–606.

Schutte-Rodin, S., Broch, L., Buysse, D., Dorsey, C., & Sateia, M. (2008). Clinical guideline for the evaluation and management of chronic insomnia in adults. *Journal of Clinical Sleep Medicine: Official Publication of the American Academy of Sleep Medicine, 4*(5), 487–504. Retrieved from https://www.ncbi.nlm.nih.gov/pubmed/18853708. Retrieved from https://www.ncbi.nlm.nih.gov/pmc/PMC2576317/

Schwartz, M. D., & Kilduff, T. (2015). The neurobiology of sleep and wakefulness. *Psychiatric Clinics of North America, 38*, 615–644. Retrieved from. https://doi.org/10.1016/j.psc.2015.07.002

Schwichtenberg, A. J., & Malow, B. A. (2015). Melatonin treatment in children with developmental disabilities. *Sleep Medicine Clinics, 10*(2), 181–187. https://doi.org/10.1016/j.jsmc.2015.02.008

Scott, H., Lack, L., & Lovato, N. (2018). A pilot study of a novel smartphone application for the estimation of sleep onset. *Journal of Sleep Research, 27*(1), 90–97. Retrieved from. https://doi.org/10.1111/jsr.12575

Shallcross, A. J., Visvanathan, P. D., Sperber, S. H., & Duberstein, Z. T. (2018). Waking up to the problem of sleep: Can mindfulness help? A review of theory and evidence for the effects of mindfulness for sleep. *Current Opinion in Psychology, 28*, 37–41.

Simard-Tremblay, E., Constantin, E., Gruber, R., Brouillette, R. T., & Shevell, M. (2011). Sleep in children with cerebral palsy: A review. *Journal of Child Neurology, 26*(10), 1303–1310. Retrieved from. https://doi.org/10.1177/0883073811408902

Souders, M. C., Zavodny, S., Eriksen, W., Sinko, R., Connell, J., Kerns, C., … Pinto-Martin, J. (2017). Sleep in children with autism spectrum disorder. *Current Psychiatry Reports, 19*(6), 34. https://doi.org/10.1007/s11920-017-0782-x

Spanò, G., Gómez, R. L., Demara, B. I., Alt, M., Cowen, S. L., & Edgin, J. O. (2018). REM sleep in naps differentially relates to memory consolidation in typical preschoolers and children with Down syndrome. *Proceedings of the National Academy of Sciences of the United States of America, 115*(46), 11844. Retrieved from http://www.pnas.org/content/115/46/11844.abstract. https://doi.org/10.1073/pnas.1811488115

Spielman, A. J., Caruso, L. S., & Glovinsky, P. B. (1987). A behavioral perspective on insomnia treatment. *The Psychiatric Clinics of North America, 10*(4), 541–553.

Spielman, A. J., Saskin, P., & Thorpy, M. J. (1987). Treatment of chronic insomnia by restriction of time in bed. *Sleep, 10*(1), 45–56.

Spielman, A. J., Yang, C.-M., & Glovinsky, P. B. (2011). Chapter 1 – Sleep restriction therapy. In M. Perlis, M. Aloia, & B. Kuhn (Eds.), *Behavioral treatments for sleep disorders* (pp. 9–19). San Diego: Academic Press.

Spruyt, K., Braam, W., & Curfs, L. M. G. (2018). Sleep in Angelman syndrome: A review of evidence. *Sleep Medicine Reviews, 37*, 69–84. Retrieved from http://www.sciencedirect.com/science/article/pii/S1087079217300175. https://doi.org/10.1016/j.smrv.2017.01.002

Spruyt, K., & Curfs, L. M. G. (2015). Non-pharmacological management of problematic sleeping in children with developmental disabilities. *Developmental Medicine & Child Neurology, 57*(2), 120–136. Retrieved from. https://doi.org/10.1111/dmcn.12623

Stallman, H. M., & Kohler, M. (2017). Systematic review of treatments for sleepwalking: 100 years of case studies. *Sleep and Hypnosis (Online), 19*(2), 21.

Stallman, H. M., Kohler, M., & White, J. (2018). Medication induced sleepwalking: A systematic review. *Sleep Medicine Reviews, 37*, 105–113. Retrieved from http://www.sciencedirect.com/science/article/pii/S1087079217300205. https://doi.org/10.1016/j.smrv.2017.01.005

Staton, S. L., Smith, S. S., Hurst, C., Pattinson, C. L., & Thorpe, K. J. (2017). Mandatory nap times and group napping patterns in child care: An observational study. *Behavioral Sleep Medicine, 15*(2), 129–143. https://doi.org/10.1080/15402002.2015.1120199

Stephan, J., Schredl, M., Henley-Einion, J., & Blagrove, M. (2012). TV viewing and dreaming in children: The UK library study. *International Journal of Dream Research, 5*(2), 130–133.

St-Onge, M.-P., Mikic, A., & Pietrolungo, C. E. (2016). Effects of diet on sleep quality. *Advances in Nutrition, 7*(5), 938–949. Retrieved from https://www.ncbi.nlm.nih.gov/pubmed/27633109. Retrieved from https://www.ncbi.nlm.nih.gov/pmc/PMC5015038/. https://doi.org/10.3945/an.116.012336

Stores, G. (2014). *Sleep and its disorders in children and adolescents with a neurodevelopmental disorder: A review and clinical guide*. Cambridge: Cambridge University Press.

Stores, G. (2016). Multifactorial influences, including comorbidities, contributing to sleep disturbance in children with a neurodevelopmental disorder. *CNS Neuroscience & Therapeutics, 22*(11), 875–879. Retrieved from Go to ISI: WOS:000387805000002. https://doi.org/10.1111/cns.12574

Stott, J., Charlesworth, G., & Scior, K. (2017). Measures of readiness for cognitive behavioural therapy in people with intellectual disability: A systematic review. *Research in Developmental Disabilities, 60*, 37–51. Retrieved from http://www.sciencedirect.com/science/article/pii/S0891422216302463. https://doi.org/10.1016/j.ridd.2016.11.003

Sturmey, P., Matson, J. L., & Lott, J. D. (2004). The factor structure of the DASH-II. *Journal of Developmental and Physical Disabilities, 16*(3), 247–255. Retrieved from. https://doi.org/10.1023/B:JODD.0000032300.05833.d1

Sun, J., Kang, J., Wang, P., & Zeng, H. (2013). Self-relaxation training can improve sleep quality and cognitive functions in the older: A one-year randomised controlled trial. *Journal of Clinical Nursing, 22*(9–10), 1270–1280. Retrieved from. https://doi.org/10.1111/jocn.12096

Tolaymat, A., & Liu, Z. (2017). Sleep disorders in childhood neurological diseases. *Children, 4*(10), 84.

Tosini, G., Ferguson, I., & Tsubota, K. (2016). Effects of blue light on the circadian system and eye physiology. *Molecular Vision, 22*, 61–72. Retrieved from https://www.ncbi.nlm.nih.gov/pubmed/26900325. Retrieved from https://www.ncbi.nlm.nih.gov/pmc/PMC4734149/

Tsai, H. J., Kuo, T. B. J., Lee, G.-S., & Yang, C. C. H. (2015). Efficacy of paced breathing for insomnia: Enhances vagal activity and improves sleep quality. *Psychophysiology, 52*(3), 388–396. Retrieved from. https://doi.org/10.1111/psyp.12333

Tyler Richard, S., Pienkowski, M., Roncancio Eveling, R., Jun Hyung, J., Brozoski, T., Dauman, N., … Moore Brian, C. J. (2014). A review of hyperacusis and future directions: Part I. Definitions and manifestations. *American Journal of Audiology, 23*(4), 402–419. Retrieved from. https://doi.org/10.1044/2014_AJA-14-0010

Unterberger, I., Gabelia, D., Prieschl, M., Chea, K., Hofer, M., Högl, B., … Frauscher, B. (2015). Sleep disorders and circadian rhythm in epilepsy revisited: A prospective controlled study. *Sleep Medicine, 16*(2), 237–242. Retrieved from http://www.sciencedirect.com/science/article/pii/S1389945714004985. https://doi.org/10.1016/j.sleep.2014.09.021

Van Dam, N. T., van Vugt, M. K., Vago, D. R., Schmalzl, L., Saron, C. D., Olendzki, A., … Meyer, D. E. (2017). Mind the hype: A critical evaluation and prescriptive agenda for research on mindfulness and meditation. *Perspectives on Psychological Science, 13*(1), 36–61. Retrieved from. https://doi.org/10.1177/1745691617709589

van de Wouw, E., Evenhuis, H. M., & Echteld, M. A. (2012). Prevalence, associated factors and treatment of sleep problems in adults with intellectual disability: A systematic review. *Research in Developmental Disabilities, 33*(4), 1310–1332. Retrieved from http://www.sciencedirect.com/science/article/pii/S0891422212000583. https://doi.org/10.1016/j.ridd.2012.03.003

Van den Bulck, J., Çetin, Y., Terzi, Ö., & Bushman, B. J. (2016). Violence, sex, and dreams: Violent and sexual media content infiltrate our dreams at night. *Dreaming, 26*(4), 271.

van Maanen, A., Meijer, A. M., van der Heijden, K. B., & Oort, F. J. (2016). The effects of light therapy on sleep problems: A systematic review and meta-analysis. *Sleep Medicine Reviews, 29*, 52–62. Retrieved from http://www.sciencedirect.com/science/article/pii/S1087079215001136. https://doi.org/10.1016/j.smrv.2015.08.009

Vijay, A., Becker, J. E., & Ross, J. S. (2018). Patterns and predictors of off-label prescription of psychiatric drugs. *PLoS One, 13*(7), e0198363. https://doi.org/10.1371/journal.pone.0198363

W Kanen, J., Nazir, R., Sedky, K., & K Pradhan, B. (2015). The effects of mindfulness-based interventions on sleep disturbance: A meta-analysis. *Adolescent Psychiatry, 5*(2), 105–115.

Waits, A., Tang, Y.-R., Cheng, H.-M., Tai, C.-J., & Chien, L.-Y. (2018). Acupressure effect on sleep quality: A systematic review and meta-analysis. *Sleep Medicine Reviews, 37*, 24–34. Retrieved from http://www.sciencedirect.com/science/article/pii/S1087079216301605. https://doi.org/10.1016/j.smrv.2016.12.004

Wams, E. J., Woelders, T., Marring, I., van Rosmalen, L., Beersma, D. G. M., Gordijn, M. C. M., & Hut, R. A. (2017). Linking light exposure and subsequent sleep: A field polysomnography study in humans. *Sleep, 40*(12), zsx165–zsx165. Retrieved from. https://doi.org/10.1093/sleep/zsx165

Ward, F., Nanjappa, M., Hinder, S. A. J., & Roy, M. (2015). Use of melatonin for sleep disturbance in a large intellectual disability psychiatry service. *International Journal of Developmental Disabilities, 61*(3), 182–187. Retrieved from. https://doi.org/10.1179/2047387714Y.0000000051

Wayte, S., McCaughey, E., Holley, S., Annaz, D., & Hill, C. M. (2012). Sleep problems in children with cerebral palsy and their relationship with maternal sleep and depression. *Acta Paediatrica, 101*(6), 618–623. Retrieved from. https://doi.org/10.1111/j.1651-2227.2012.02603.x

Weinreich, G., Wessendorf, T. E., Pundt, N., Weinmayr, G., Hennig, F., Moebus, S., … Hoffmann, B. (2015). Association of short-term ozone and temperature with sleep disordered breathing. *European Respiratory Journal, 46*(5), 1361–1369. Retrieved from http://erj.ersjournals.com/content/early/2015/07/09/13993003.02255-2014.abstract. https://doi.org/10.1183/13993003.02255-2014

Willner, P. (2004). Brief cognitive therapy of nightmares and post-traumatic ruminations in a man with a learning disability. *British Journal of Clinical Psychology, 43*(4), 459–464.

Wong, J., Motulsky, A., Abrahamowicz, M., Eguale, T., Buckeridge, D. L., & Tamblyn, R. (2017). Off-label indications for antidepressants in primary care: Descriptive study of prescriptions from an indication based electronic prescribing system. *BMJ, 356*, j603. https://doi.org/10.1136/bmj.j603

World Health Organization. (2012). *Night noise guidelines for Europe*. Retrieved from http://www.euro.who.int/data/assets/pdf_file/0017/43316.E92845.pdf

Xu, Q., & Lang, C. P. (2018). Revisiting the alerting effect of light: A systematic review. *Sleep Medicine Reviews, 41*, 39–49. Retrieved from http://www.sciencedirect.com/science/article/pii/S1087079217300989. https://doi.org/10.1016/j.smrv.2017.12.001

Xyrichis, A., Wynne, J., Mackrill, J., Rafferty, A. M., & Carlyle, A. (2018). Noise pollution in hospitals. *BMJ, 363*, k4808. Retrieved from http://www.bmj.com/content/363/bmj.k4808.abstract. https://doi.org/10.1136/bmj.k4808

Ye, L., & Richards, K. C. (2018). Sleep and long-term care. *Sleep Medicine Clinics, 13*(1), 117–125. Retrieved from https://www.ncbi.nlm.nih.gov/pubmed/29412978. Retrieved from https://www.ncbi.nlm.nih.gov/pmc/PMC5957502/. https://doi.org/10.1016/j.jsmc.2017.09.011

Yeung, V., Sharpe, L., Glozier, N., Hackett, M. L., & Colagiuri, B. (2018). A systematic review and meta-analysis of placebo versus no treatment for insomnia symptoms. *Sleep Medicine Reviews, 38*, 17–27. Retrieved from http://www.sciencedirect.com/science/article/pii/S1087079217300795. https://doi.org/10.1016/j.smrv.2017.03.006

Yeung, W.-F., Chung, K.-F., Yung, K.-P., Ho, F. Y.-Y., Ho, L.-M., Yu, Y.-M., & Kwok, C.-W. (2014). The use of conventional and complementary therapies for insomnia among Hong Kong Chinese: A telephone survey. *Complementary Therapies in Medicine, 22*(5), 894–902. Retrieved from http://www.sciencedirect.com/science/article/pii/S0965229914001162. https://doi.org/10.1016/j.ctim.2014.08.001

Zhou, E. S., Gardiner, P., & Bertisch, S. M. (2017). Integrative medicine for insomnia. *Medical Clinics of North America, 101*(5), 865. Retrieved from Go to ISI: WOS:000410717100004. https://doi.org/10.1016/j.mcna.2017.04.005

Treating Noncompliance in Persons with Dual Diagnosis

37

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Introduction

Noncompliance is a common behavior problem among individuals with intellectual and developmental disabilities (Coe, Matson, Russel, & Stallings, 1999; Mace et al., 1988). This behavior can cause disruptions in school, community, and home settings for the individual and those surrounding them. In schools, noncompliance immediately interferes with academic engagement and skill acquisition, and an individual may face social consequences (e.g., isolation) depending on the frequency and severity of the noncompliant behavior (Wadsworth, Hansen, & Wills, 2015). Additionally, noncompliance to community rules and expectations may prohibit an individual from participating in social outings with family or with peers. In emergency situations, noncompliance to demands from caregivers or emergency personnel can even be life-threatening. Thus, addressing noncompliance can have lasting impact across many domains of an individual's daily functioning.

This chapter reviews decades of research informing compliance training for individuals with intellectual and developmental disabilities. First, the chapter provides definitions of noncompliance, followed by individual and environmental factors related to noncompliance. A brief overview of functional behavior assessments as they relate to noncompliance is then provided. Several behavioral intervention strategies to address noncompliance are discussed. It is important to note that many of the intervention procedures described in this chapter are often implemented simultaneously, as the most comprehensive approach to noncompliant behavior is likely to be most effective.

Defining Noncompliance

Noncompliance refers to various behaviors that occur following the delivery of an instruction, which may be categorized as behavioral excesses (i.e., behaviors the individual engages in instead of the instructed task) or behavioral deficits (i.e., the instructed task is not initiated at all or is not initiated in a timely manner). Because the term "noncompliance" can have many forms, or topographies, a behavioral definition must be formulated before functional assessment.

Hawkins and Dobes (1977) state that a behavioral definition must be complete, specific, and objective, such that independent observers of the behavior could read said definition and identify occurrences of the same behavior reliably. Using this definition, noncompliance in the form of a behavioral excess (i.e., behaviors the individual engages in instead of the instructed task) may be

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observed as verbal noncompliance (e.g., saying "No," cursing), elopement (e.g., walking away) or the attempt at elopement from the instructional setting, or the occurrence of challenging behavior upon the presentation of the stimulus (i.e., the instruction). Noncompliance in the form of behavioral deficits may be observed as a lack of response or engagement in the given instruction, or as the engagement in the instruction following too lengthy a time period after the presentation of the stimulus.

Once a behavioral definition is formulated, the clinician must specify procedures for measuring the behavior in order to inform the functional assessment. With noncompliance, this is typically measured by the individual's latency to respond to the given instruction, which may range from 5 to 30 seconds (Shriver & Allen, 1997). It is also critical to document the frequency and duration of noncompliance, as this may inform intervention selection as well (Miltenberger & Weil, 2013). Many typically developing children engage in noncompliant behaviors on occasion, especially in a school setting (Bryce & Jahromi, 2013). However, Bryce and Jahromi (2013) also found that students with autism spectrum disorder (ASD) engaged in noncompliant behaviors at higher rates than their typically developing peers. Chronic noncompliance would thus require more intensive and individual intervention strategies.

Individual and Environmental Factors

There are several individual and environmental factors to consider in order to provide individualized and effective treatment to increase compliance. These include comorbidities, the presence of challenging behavior, intellectual ability, the setting in which noncompliance typically occurs, and cultural issues. Comprehensive assessment and understanding of these factors is critical for clinicians planning and implementing intervention procedures.

Comorbid Psychopathology

Individuals with intellectual disabilities evince a range of comorbid psychopathologies and conditions, including anxiety, depression, schizophrenia, and stereotypies; the condition with the greatest overlap with intellectual disability is autism spectrum disorder (Matson & Shoemaker, 2009). Researchers have found that comorbid psychopathologies increase the likelihood of the presence of challenging behaviors (Matson & Shoemaker, 2009; Shattuck et al., 2006). As such, comorbidities should be considered when developing intervention strategies to address noncompliance in individuals with intellectual disability.

Presence of Challenging Behavior

The relationship between noncompliance and challenging behavior is highly prevalent among individuals with intellectual disability, such that noncompliance is often considered a subtype of challenging behavior. In fact, Emerson and colleagues (2001) found that 80% of individuals who display serious challenging behavior also engaged in chronic noncompliance. Several studies have found that individuals with comorbid intellectual disability and autism spectrum disorder engage in higher rates of challenging behavior, such as aggression (McClintock, Hall, & Oliver, 2003). In a sample of toddlers referred to early intervention services, Fox, Keller, Grede, and Bartosz (2007) found that 24% of toddlers displayed aggressive behavior and 41% exhibited tantrum behaviors. These behavior problems put individuals with intellectual disabilities at risk for limited community involvement and long-term inpatient care, and put the individual and caregivers at risk for harm. Additionally, in severe cases of challenging behavior, restrictive restraint procedures or medication may be warranted in addition to behavioral intervention strategies. Murphy and colleagues (2005) report that these problems are likely to persist unless addressed, and so clinicians should be prepared to develop additional programming to address these

problems when considering compliance training for individuals with intellectual disability.

Intellectual Abilities

An individual's intellectual abilities impact their ability to communicate effectively and their acquisition of skills necessary to comply with many instructions (Emerson et al., 2001; Matson & Shoemaker, 2009). Thus, these abilities must be considered while treatment planning, as an individual's behavior repertoire may need to be developed further in order to increase compliance. Individuals with intellectual disabilities may not comply with instructions if they lack the skill to perform the behavior; for example, telling a child to "Put your utensils away" before developing their vocabulary to include "utensils" may result in noncompliance with the demand initially. Additionally, expanding an individual's behavioral repertoire may actually promote compliance, as the individual may encounter natural reinforcement as their compliance with behaviors within their repertoire increases. This in turn can increase compliance to more complex instructions.

Setting

When clinicians are planning interventions to increase compliance, it is important to account for the setting in which training takes place. Most intensive behavioral interventions take place in clinic settings, often with individuals working one-on-one with a trained clinician (Linstead et al., 2017). Clinicians should be sure to provide other relevant caretakers (e.g., parents, teachers, family members) with resources to generalize compliance with demands across multiple settings (Cooper, Heron, & Heward, 2007). These types of interventions are discussed later in this chapter; however, it is important for clinicians to consider how consistently their procedures are implemented across settings in order to provide for generalization and maintenance of skills.

Cultural Considerations

Researchers have explored how cultural influences may impact treatment decision-making for parents and have found that cultural beliefs impact not only parents' interpretations of symptoms, but also their beliefs regarding the course of a disorder (Mandell & Novak, 2005). This then impacts which treatments parents pursue in order to best help their children. Though not studied extensively in the intellectual disabilities population, research from other medical fields also shows that patients' perceptions of a clinician's cultural competency influences their compliance with recommended treatments. In the context of treatment of noncompliance, it is important for clinicians to provide parents with resources to understand behavioral approaches to treatment. Though behavioral interventions are the most evidence-based approaches to treatment of any problem behavior, parents may be hesitant to pursue what may appear to be a costly and time-extensive procedure.

Functions of Noncompliance

Noncompliance occurs following the presentation of a demand or instruction, and as such the function of escape from demand is often relevant. If an individual responds noncompliance after several demands have been given or if a history of noncompliance is known following the presentation of certain requests, caregivers, teachers, and others who work with individuals with intellectual disabilities may decrease the frequency in which they make these demands. This strengthens the individual's noncompliant behavior through negative reinforcement. Since the term "noncompliance" is generally associated with task avoidance, this could indicate that noncompliance is consistently maintained by escape from demands (i.e., negative reinforcement). In fact, a review of functional assessment studies (Matson et al., 2011) found that escape was the most commonly reported function of several challenging behaviors. However, Matson and Nebel-Schwalm (2007) discuss how challenging

behaviors may appear straightforward, but the complex patterns of behavior maintaining them may not be fully understood. For example, a child may engage in repetitive motor movements (e.g., hand flapping, spinning) following a demand. This behavior may serve as automatic reinforcement, since the behavior itself is pleasurable to the individual, rather than escape from the demand placed (Rapp & Vollmer, 2005). Researchers have also found that noncompliance may also be maintained by attention (Rodriguez, Thompson, Schlichenmeyer, & Stocco, 2012). Clinicians should be sure to assess for multiple functions of noncompliant behavior in order to individualize the appropriate intervention strategy.

Functional Assessment

The initial procedure in determining an intervention strategy to increase compliance with demands should be investigation into the function of the noncompliant behavior. As discussed previously, a clear, objective definition of the form of noncompliant behavior is necessary, as the behavior may serve an underlying function that is not readily apparent. Research suggests that a functional behavioral assessment (FBA) is an effective method of identifying these functions as well as the reinforcers maintaining noncompliant behavior (Didden, Duker, & Korzilius, 1997; Iwata, Dorsey, Slifer, Bauman, & Richman, 1994; Roane, Fisher, & Carr, 2016). FBA refers to the process of gathering and interpreting data related to the function of a problem behavior (O'Neill, Albin, Storey, Horner, & Sprague, 2014). This can be achieved through informant methods, such as structured interviews or clinical rating scales of behavior (Kozlowski & Matson, 2012). These methods are simple to administer and time-efficient, and in many cases of noncompliant behavior may be sufficient to inform intervention planning. However, a clinician should employ more comprehensive methods when intervention methods do not appear effective or when multiple functions are suspected.

One method of assessment is systematic, direct observation of the behavior and environmental variables related to the behavior through analysis of the antecedents and consequences, known as the "ABC" method of observation (O'Neill et al., 2014); the use of this method is very common in a variety of clinical settings as well as schools (Kozlowski & Matson, 2012). Data is collected regarding the antecedent event ("A"), the behavior ("B"), and the immediate consequence ("C") of the behavior, which assists the clinician in formulating their hypothesis regarding the behavior's function. For example, in a classroom setting, a student with intellectual disability repeatedly yells, "No!" when the teacher prompts him to share certain preferred toys with peers; this occasionally escalates into aggressive behavior towards the teacher. The teacher responds to the student's noncompliance by placing him in "time-out" in which the student is isolated from peers to play in the back corner of the room. A clinician conducting an FBA using the ABC method of observation may find that the antecedent ("A") of this behavior is typically the teacher's verbal request to share preferred items. The student's behavior ("B") typically results in the consequence ("C") of time-out. With the available information, a clinician might hypothesize that the noncompliant behavior is maintained by escape, as the student effectively removes himself from situations where he is prompted to share these items.

Another method for identifying the function of behavior is an *experimental functional analysis*(EFA). EFA is a method that involves the systematic manipulation of antecedents and consequences associated with the target behavior during four assessment conditions (i.e., attention, demand, play, and alone) (Iwata et al., 1994). The target behavior is measured and compared across these alternating conditions, and higher rates of behavior in a condition would indicate a stronger functional relationship. Additionally, the EFA method easily allows the clinician to identify whether the behavior serves multiple functions, which may be valuable in assessing noncompliant behaviors. Unfortunately, though this approach gives the best insight into the function of a behav-

ior, EFA procedures are often time consuming and require a large number of resources; this may limit their appropriateness for use in certain settings (Matson & Minshawi, 2007).

Interventions to Address Noncompliance

High and Medium Probability Command Sequences

High-probability command sequences (HPCS) and medium-probability command sequences (MPCS) are commonly used as a means to increase compliance with demands among people with intellectual disabilities (Lipschultz & Wilder, 2017). These procedures can be used across a variety of settings, including clinical, academic, and home environments (Belfiore, Basile, & Lee, 2008; Lee, Belfiore, Scheeler, Hua, & Smith, 2004). Behavioral momentum refers to the concept that behaviors reinforced at higher rates (e.g., high probability instructions) will have greater resistance to change (Mace et al., 1988). As such, the low probability instruction that follows has a higher likelihood of compliance following the series of high probability instructions preceding it. A high probability instruction is typically defined as instructions to which an individual complies appropriately at least 80% of the time. For medium-probability instructions, compliance should be observed 50–70% of the time. In clinical practice, the addition of medium-probability instructions to the command sequences provides opportunities for responses from individuals with lower ability levels, who may not achieve 80% compliance with instructions.

Both HPCS and MPCS procedures are implemented similarly. In order to implement these procedures, clinicians must first develop a hierarchy of relevant instructions for the individual. Baseline data should be collected on the probability of correct responses to these instructions, in order to develop the hierarchy of high to medium- to low-probability instructions. The procedures are then implemented by in a series of instructions, with high- or medium-probability instructions preceding the target low-probability instruction. Reinforcement is provided following each instance of compliance. Noncompliant behaviors should not receive reinforcement in this procedure.

HPCS and MPCS procedures are highly supported in the literature for increasing compliance among individuals with intellectual disabilities (Radley & Dart, 2016). Additionally, Lipschultz and Wilder's (2017) review of HPCS procedures recommended additional procedures for effective HPCS procedures; they recommend that at least three high-probability instructions precede a low-probability instruction and that no more than 5 seconds follow each instruction. These recommendations are easily applied to clinical practice and improve the effectiveness of these procedures. Finally, Romano and Roll (2000) compared the effectiveness of HPCS and MPCS procedures. In their study, both high- and medium-probability requests increased compliance to low-probability requests and thus support the effectiveness of both procedures in increasing compliance to instructions (Romano & Roll, 2000).

In both HPCS and MPCS procedures, as well as the procedures discussed in the remainder of the chapter, clinicians and parents or caregivers must ensure they are delivering effective instructions. That is, instructions that result in increased compliance among individuals with intellectual disabilities are typically affirmative, in that they present the child with the behavior that should be engaged in. In contrast, a negative instruction informs the child about not to do. Researchers have found that presenting affirmative instructions to individuals with intellectual disabilities results in higher levels of compliance (Ducharme & Worling, 1994). Presenting instructions in an affirmative rather than a negative format can result in greater compliance for PWIDD (Ducharme & Worling, 1994). However, clinicians should ensure that they program for generalization to other instructive formats, as individuals with intellectual disabilities may still encounter natural situations where a negative instruction is given, particularly in environments outside of the intervention setting (Neef, Shafer, Egel, Cataldo, & Parrish, 1983).

Guided Compliance

The HPCS and MPCS procedures described previously are typically combined in clinical practice with other procedures, as they do not provide methods for the occurrence of noncompliance or challenging behavior beyond reducing reinforcement for those behaviors. The procedure of guided compliance may be used in conjunction with those procedures or other interventions described in this chapter, and it is particularly useful when an individual exhibits noncompliance due to a behavioral deficit (e.g., a lack of skills necessary to complete the instruction). As with other interventions, an understanding of the functions maintaining the behavior is critical, as the procedure may reinforce certain noncompliant behaviors (i.e., those maintained by social attention) (Kern, Delaney, Hilt, Bailin, & Elliot, 2002). Guided compliance is typically implanted via the least-to-most prompting procedure originally described by Horner and Keilitz (1975). In this procedure, when noncompliance occurs, the clinician initiates a verbal prompt, followed by a gestural prompt, and finally, partial-physical or full-physical redirection to the task. Researchers have evaluated this method's efficacy for use, though little research has looked specifically at noncompliance (Smith & Lerman, 1999). However, Smith and Lerman (1999) did find that compliance increased to upwards of 70% when the guided compliance procedure was implemented. Guided compliance must still be used cautiously, as researchers have found that these procedures (i.e., physical redirection) may be aversive to individuals and actually increase challenging behaviors (Wilder, Saulnier, Beavers, & Zonneveld, 2008).

Errorless Compliance Training

Another procedure similar to the HPCS and MPCS procedures is that of errorless compliance training (ECT), which was originally developed by Ducharme and Popynick (1993). In this procedure, instructions with variable probabilities (i.e., not only high-probability or medium-probability instructions) are presented to the individual in order to increase compliance with low-probability requests. As Radley and Dart (2016) discuss in their review, there is substantial support for the use of this procedure, largely due to the fact that it draws from methods outlined in HPCS procedures. In ECT, four distinct levels are developed based on the individual's probability of responses. The clinician decides what percentage of compliance categorizes each level. The procedure differs from both HPCS and MPCS procedures in that there is no predetermined percentage cut-off for each level, which makes the procedure highly flexible to meet the individual needs of the client. Additionally, this would allow clinicians to develop compliance training procedures for individuals with minimal amounts of high- and medium-probability responses, instead organizing the instructions by the actual levels of compliance the individual evinces. This procedure can be used across settings and has been tested in individuals with intellectual disabilities and autism spectrum disorder.

Differential Reinforcement

There are four types of differential reinforcement procedures: differential reinforcement of incompatible behavior (DRI), differential reinforcement of alternative behavior (DRA), differential reinforcement of other behavior (DRO), and differential reinforcement of low rates of behavior (DRL). Differential reinforcement is an effective procedure which consists of providing reinforcement based on engagement in an acceptable behavior or a decreased in the undesired behavior (Cooper et al., 2007; Fischetti et al., 2012). In addition, reinforcement for the target behavior (i.e., noncompliance) is reduced or removed completely.

Differential reinforcement of incompatible behavior (DRI) and differential reinforcement of alternative behavior (DRA) procedures provide reinforcement for alternative and appropriate behaviors (Cooper et al., 2007). In terms of noncompliance, the acceptable behaviors would be compliance with the given demand. These proce-

dures are typically more applicable when noncompliance involves behavioral excess, such as challenging behavior. For example, if a child yells, "No!" in response to a teacher's instruction, DRA procedures could be implemented to teach the child to request a break. Additionally, in the case of a skill deficit regarding the instruction, an acceptable alternative would be to have the individual ask for help with the instruction rather than engage in noncompliance.

More applicable to addressing noncompliant are differential reinforcement of other behavior (DRO) and differential reinforcement of low rates (DRL) procedures (Fischetti et al., 2012). In DRO procedures, reinforcement is provided contingent on the absence of noncompliance at a specified time or during a specified time interval (Cooper et al., 2007). For noncompliance maintained by escape, a DRO procedure could be implemented by providing the individual with breaks set at specific time intervals contingent on compliance throughout the specified time. Should the individual engage in noncompliance during the interval, reinforcement would not be provided and the time interval would restart. Time intervals are easily increased and decreased based on the needs of the individual.

Finally, DRL procedures provide reinforcement only if noncompliance occurs at a predetermined low rate during a specified time period (Cooper et al., 2007). For instance, noncompliance may be permitted to no more than five demands in a 1-hour period, after which reinforcement would be provided. The occurrences of noncompliant behavior or time period can be increased or decreased depending on the individual's behavior.

Noncontingent Reinforcement

Though not specifically investigated with regard to noncompliance, noncontingent reinforcement (NCR) procedures can be applied to noncompliance that is attention-maintained. In this procedure, reinforcement is delivered according to a variable or fixed time schedule, regardless of the presence of noncompliance or other challenging behavior (Carr et al., 2000). This procedure can also be applied to noncompliance maintained by escape from demands, by providing intermittent breaks according to predetermined schedule (Kodak, Miltenberger, & Romaniuk, 2003). These procedures are believed to decrease the individual's motivation to engage in noncompliant behaviors. However, it should be noted that this procedure does not provide the individual with the appropriate response and as such should be paired with other intervention strategies to increase skill acquisition. Research supports the use of NCR methods with other challenging behaviors, which indicates NCR's potential for addressing noncompliance specifically (Carr et al., 2000; Ingvarsson, Kahng, & Hausman, 2008). This procedure has also been taught in 1-hour training sessions for parents with little to no previous experience with these methods, making this procedure a valuable tool for parent training modules (Noel & Getch, 2016).

Extinction

Extinction procedures are commonly utilized when an individual engages in noncompliance in the form of behavioral excesses (e.g., aggression, tantrums) in order to access reinforcement. In an extinction procedure, the relationship between the environmental variable that was maintaining this behavior through reinforcement and the noncompliant behavior itself is diminished through the removal of reinforcement to the extent possible. A simple example of this could be if a child's noncompliance was maintained by the attention from a teacher in the classroom. In this case, attention could be in the form of a reprimand from the teacher. An extinction procedure would involve a plan to no longer provide this attention to the child when noncompliance occurs. Removing reinforcement for the noncompliance then decreases the likelihood that the behavior will occur again.

However, there are many situations where extinction would not be an appropriate procedure to implement. If the challenging behavior the child engages in rather than complying with the

instruction is too severe, extinction procedures may not be appropriate. For example, children may engage in self-injurious behavior when given an instruction. Extinction procedures alone may lead the child to escalate this behavior in order to access some form of reinforcement, which can put the child's safety at risk. Also, there is the possibility of extinction bursts with these severe behaviors, which is an increase in the behavior immediately following the extinction procedures (Cooper et al., 2007). This burst in challenging behavior can have obvious and severe consequences for all involved. Additionally, researchers have found that extinction procedures may actually lead to increases in aggression (Lerman, Iwata, & Wallace, 1999). It is important that this procedure be used in conjunction with other behavioral methods, such as the ones previously described. Matson, Dixon, and Matson (2005) also found that successful interventions often utilized antecedent alterations, reinforcement-based strategies, and consequence manipulations.

Functional Communication Training

Functional communication training (FCT), as first described by Carr and Durand (1985), is a popular intervention for individuals with intellectual disabilities because it helps acquire known skill deficits by increasing verbal communication skills (Chezan, Drasgow, & Martin, 2014; Luiselli, 2009). In FCT, a child's communicative behavior is reinforced rather than the noncompliant behavior. Depending on the function of the noncompliant behavior, this communicative behavior can be a verbalization (e.g., saying, "I want a break"), a written response, signed request (e.g., using the sign for "Done"), or through the use of an assistive communication device. Should the function of the behavior be attention, a clinician may implement FCT to teach the individual to appropriately request social attention. Reinforcing appropriate requests decreases the likelihood that the individual engages in noncompliance. In this procedure, noncompliant behavior should not be reinforced by allowing the individual to escape from the demand or by providing attention to the behavior (Zangrillo, Fisher, Greer, Owen, & De Souza, 2016).

Self-Monitoring

Research has demonstrated some evidence for the use of self-monitoring as an intervention strategy to increase compliance for individuals with intellectual disabilities. Self-monitoring requires that the individual record data on their own behavior and, thus, would require the individual to have an advanced skill repertoire (Bialas & Boon, 2010). This approach may be suitable for individuals with mild or moderate intellectual disability who have gained the appropriate skills to implement this strategy. Clinicians should ensure that self-monitoring strategies are used to increase compliance with instructions that are within the individual's behavioral repertoire as well as ensure adequate reinforcement is contingent upon compliance.

Wadsworth and colleagues (2015) implemented self-monitoring procedures modified by teacher prompts with two children in a special education classroom. A token board with the reinforcer noted on the board was given to each student. Students earned tokens for each instruction in which they demonstrated compliance with 5 seconds of the request. Initially, teachers provided token reinforcers for compliance, and after 80% compliance was achieved, students were prompted to place tokens themselves. Compliance was maintained as teacher prompts to utilize the token board were faded. These data provide support for the use of this strategy to increase compliance in a classroom setting. Bialas and Boon (2010) evaluated self-monitoring with three kindergarten students in an inclusive classroom setting by the use of a checklist and visual prompts to increase compliance. In this study, teachers provided no more than two verbal prompts of the target instruction. Effects were almost immediate. Compliance with the procedure was achieved for all three students and was maintained at follow-up. Some notable limitations of this procedure include the lack of participants with a diagnosis of intellectual disability.

Students were in an inclusive kindergarten classroom. Advanced skills may not be required or even expected at this age, limiting the generalizability of the results to increase compliance in other settings or with individuals of different ages and functioning levels.

Further investigation into the generalizability of the effects of self-monitoring is necessary. Additionally, the two studies discussed tested this procedure for children with more advanced skills. Therefore, researchers must investigate the effectiveness of self-monitoring in adolescents and adults as well as individuals with more functional impairments. At present, the strategy has some evidence promoting its use in a classroom setting for young children with intellectual disability (Bialas & Boon, 2010; Wadsworth et al., 2015). Clear benefits relate to promoting the individual's independence in the classroom as well as the feasibility of implementation for teachers were noted. Teachers may be more likely to implement a strategy that requires minimal effort on their own part, allowing them to focus on lessons and other instructional activities.

Conclusion

Decades of research on behavioral intervention strategies have guided clinicians' approaches to decreasing noncompliance in individuals with intellectual disabilities. As noncompliance is a common referral concern for behavioral treatment, clinicians must be prepared to implement the most appropriate strategies which are individualized to the needs and abilities of the person. A clear behavioral definition of the noncompliant behavior is a necessity, as the term "noncompliance" may refer to a number of behavioral deficits and excesses. Functional behavioral assessments must inform all intervention procedures so as not to inadvertently maintain and reinforce noncompliant behavior. A variety of interventions were discussed in this chapter. In general, multiple strategies may be implemented which focus on reducing reinforcement provided to noncompliance while reinforcing acceptable alternative behaviors.

References

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

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.

Bryce, C. I., & Jahromi, L. B. (2013). Brief report: Compliance and noncompliance to parental control strategies in children with high-functioning autism and their typical peers. *Journal of Autism and Developmental Disorders, 43*(1), 236–243.

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., Coriaty, S., Wilder, D. A., Gaunt, B. T., Dozier, C. L., Britton, L. N., … Reed, C. L. (2000). A review of "noncontingent" reinforcement as treatment for the aberrant behavior of individuals with developmental disabilities. *Research in Developmental Disabilities, 21*(5), 377–391.

Chezan, L. C., Drasgow, E., & Martin, C. A. (2014). Discrete-trial functional analysis and functional communication training with three adults with intellectual disabilities and problem behavior. *Journal of Behavioral Education, 23*(2), 221–246.

Coe, D. A., Matson, J. L., Russel, D., & Stallings, S. (1999). Behavior problems of children with Down syndrome and life events. *Journal of Autism and Developmental Disorders, 29*, 149–156.

Cooper, J. O., Heron, T. E., & Heward, W. L. (2007). *Applied behavior analysis* (2nd ed.). Upper Saddle River, NJ: Pearson.

Didden, R., Duker, P. C., & Korzilius, H. (1997). Meta-analytic study on treatment effectiveness for problem behaviors with individuals who have mental retardation. *American Journal of Mental Retardation, 101*, 387–399.

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

Fischetti, A. T., Wilder, D. A., Myers, K., Leon-Enriquez, Y., Sinn, S., & Rodriguez, R. (2012). An evaluation of evidence-based interventions to increase compli-

ance among children with autism. *Journal of Applied Behavior Analysis, 45*(4), 859–863.

Fox, R. A., Keller, K. M., Grede, P. L., & Bartosz, A. M. (2007). A mental health clinic for toddlers with developmental delays and behavior problems. *Reserach in Developmental Disabilities, 28*(2), 119–129.

Hawkins, R. P., & Dobes, R. W. (1977). Behavioral definitions in applied behavior analysis: Explicit or implicit. In B. C. Etzel, J. M. LeBlanc, & D. M. Baer (Eds.), New developments in behavioral research: Theory, method, and application (pp. 167–188). Lawrence Erlbaum Associates.

Horner, R. D., & Keilitz, I. (1975). Training mentally retarded adolescents to brush their teeth1. *Journal of Applied Behavior Analysis, 8*(3), 301–309. https://doi.org/10.1901/jaba.1975.8-301

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

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*(2), 197–209.

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

Kozlowski, A., & Matson, J. L. (2012). Interview and observation methods. In J. L. Matson (Ed.), *Functional assessment for challenging behaviors* (pp. 105–124). New York, NY: Springer Science & Business Media.

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

Lerman, D.C., Iwata, B.A., & Wallace, M.D. (1999). Side effects of extinction: Prevalence of bursting and aggression during the treatment of self-injurious behavior. *Journal of Applied Behavior Analysis, 32*(1), 1–8.

Linstead, E., Dixon, D. R., French, R., Granpeesheh, D., Adams, H., German, R., … Kornack, J. (2017). Intensity and learning outcomes in the treatment of children with autism spectrum disorder. *Behavior Modification, 41*(2), 229–252. https://doi.org/10.1177/0145445516667059

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

Luiselli, J. K. (2009). Aggression and noncompliance. In *Applied behavior analysis for children with autism spectrum disorders*. New York, NY: Springer.

Mace, F. C., Hock, M. L., Lalli, J. S., West, B. J., Belfiore, P. J., Pinter, E., & Brown, D. K. (1988). Behavioral momentum in the treatment of noncompliance. *Journal of Applied Behavior Analysis, 21*, 123–141.

Mandell, D. S., & Novak, M. (2005). The role of culture in families' treatment decisions for children with autism spectrum disorders. *Mental Retardation and Developmental Disabilities Research Reviews, 11*(2), 110–115. https://doi.org/10.1002/mrdd.20061

Matson, J. L., & Minshawi, N. F. (2007). Functional assessment of challenging behavior: Toward a strategy for applied settings. *Research in Developmental Disabilities, 28*(4), 353–361. https://doi.org/10.1016/j.ridd.2006.01.005

Matson, J.L., Dixon, D.R., & Matson, M.L. (2005). Assessing and treating aggression in children and adolescents with developmental disabilities: A 20-year overview. *Educational Psychology, 25*(2), 151–181.

Matson, J. L., & Nebel-Schwalm, M. (2007). Assessing challenging behaviors in children with autism spectrum disorders: A review. *Research in Developmental Disabilities, 28*(6), 567–579. https://doi.org/10.1016/j.ridd.2006.08.001

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., 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

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. https://doi.org/10.1046/j.1365-2788.2003.00517.x

Miltenberger, R. G., & Weil, T. M. (2013). Observation and measurement in behavior analysis. In G. J. Madden (Ed.), *APA handbook of behavior analysis: Vol. 1 Methods and principles*. Washington, DC: APA.

Murphy, G. H., Beadle-Brown, J., Wing, L., Gould, J., Shah, A., & Holmes, N. (2005). Chronicity of challenging behaviours in people with severe intellectual disabilities and/or autism: A total population sample. *Journal of Autism and Developmental Disorders, 35*(4), 405–418.

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

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

O'Neill, R. E., Albin, R. W., Storey, K., Horner, R. H., & Sprague, J. R. (2014). *Functional assessment and program development for problem behavior: A practical handbook* (3rd ed.). Stamford, CT: Cengage Learning.

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

Rapp, J. T., & Vollmer, T. R. (2005). Stereotypy I: A review of behavioral assessment and treatment. *Research in Developmental Disabilities, 26*(6), 527–547. https://doi.org/10.1016/j.ridd.2004.11.005

Roane, H. S., Fisher, W. W., & Carr, J. E. (2016). Applied behavior analysis as treatment for autism spectrum disorder. *The Journal of Pediatrics, 175*, 27–32.

Rodriguez, N. M., Thompson, R. H., Schlichenmeyer, K., & Stocco, C. S. (2012). Functional analysis and treatment of arranging and ordering by individuals with an autism spectrum disorder. *Journal of Applied Behavior Analysis, 45*(1), 1–22. https://doi.org/10.1901/jaba.2012.45-1

Romano, J. P., & Roll, D. (2000). Expanding the utility of behavioral momentum for youth with developmental disabilities. *Behavioral Interventions, 15*(2), 99–111.

Shattuck, P. T., Seltzer, M. M., Greenberg, J. S., Orsmond, G. I., Bolt, D., Kring, S., … Lord, C. (2006). 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

Shriver, M. D., & Allen, K. D. (1997). Defining child noncompliance: An examination of temporal parameters. *Journal of Applied Behavior Analysis, 30*(1), 173–176.

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

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.

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.

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

Treatment of Social Skills in Dual Disorders

38

Jeff Sigafoos, Vanessa A. Green, Mark F. O'Reilly, and Giulio E. Lancioni

Introduction

The development of social skills is an important treatment priority for many individuals with intellectual disability (Matson, Terlonge, & Minshawi, 2008). This priority reflects the fact that social skills deficits—as well as excessive and inappropriate social behaviors—are prevalent among individuals with intellectual disability (American Association on Intellectual and Developmental Disabilities, 2010; American Psychiatric Association, 2013; Buntinx, 2016; Matson et al., 2008). Indeed, failure to engage in age-appropriate, contextually appropriate, and culturally appropriate social behavior is a commonly noted characteristic of people with intellectual disability (American Psychiatric Association, 2013).

Common types of social skills deficits that have been reported among people with intellectual disability include (a) failing to greet others or make eye contact during conversations, (b) having difficulties with respect to perceiving social cues, (c) using immature language forms, and (d) experiencing difficulties in regulating emotions (American Psychiatric Association, 2013, p. 34). Buntinx (2016) delineated a number of additional social skills deficits that appear prevalent among individuals with intellectual disabilities. These specific deficits include difficulties with respect to (a) engaging in conversation, (b) expressing feelings, (c) demonstrating self-control (e.g., anger management), (d) accurately estimating own abilities and limitations, (e) politeness and manners, (f) maintaining positive peer relations, (g) social problem solving, and (h) resisting negative peer pressure.

Excessive social behaviors are also prevalent among individuals with intellectual disability (Singh, 2016; Sturmey & Didden, 2014). Excessive social behaviors include a general tendency to act inappropriately within different contexts, such as talking too loudly in a café or failing to remain seated during a classroom lecture. A person might also engage in acceptable forms of social behaviors, but do so at a rate that is considered excessive, such as frequently interrupting others, asking the same question over and over, or hugging peers too often and/or in situations where such displays of affection would be seen as inappropriate.

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Given the prevalence of social skills deficits and excesses, it is not surprising that a considerable amount of research has focused on developing and evaluating different intervention approaches for improving the social functioning and coping skills of persons with intellectual disability. Specific treatment aims might—depending on the individual's unique needs and characteristics—include (a) teaching new social skills, (b) increasing or refining existing social skills, and/or (c) replacing or reducing inappropriate/excessive social behavior. Addressing these types of treatment aims would ideally lead to an overall improved quality of life. Social skills interventions should also aim to (a) promote inclusion, (b) increase participation, (c) establish positive peer relations, and (d) reduce the social stigma associated with limited social skills. Social skills intervention is also a high priority in light of evidence suggesting associations between social skills deficits and mental health issues and challenging behavior among individuals with intellectual disability.

Social Skills and Mental Health

A wide range of mental health problems have been reported among individuals with intellectual disabilities, including anxiety, phobia, affective/mood disorders, psychosis, and schizophrenia (Matson & Shoemaker, 2009). Indeed, the risk of mental health problems for people with intellectual disabilities appears to be rather considerable (Hove & Havik, 2010). Totsika, Felce, Kerr, and Hastings (2010), for example, found evidence of psychiatric disorders in 23.3% of a sample of older adults with intellectual disability.

Underwood, McCarthy, and Tsakanikos (2010) reviewed literature on factors affecting the mental health of individuals with intellectual disability. Their review indicated that severity of intellectual disability and the extent of adaptive behavior deficits, including social skills deficits, were "associated with poorer mental health" (p. 421). With respect to the influence of social skills on mental health, Matson, Dempsey, and Rivet (2009) sought to determine if there was any association between social skills and psychopathology among individuals with intellectual disability. In this study, they assessed 302 adults with severe to profound intellectual disability using standardized measures of psychopathology and social skills. Analyses of the assessment data from these two measures revealed significant associations between the presence of negative social behavior and increased severity of anxiety, impulsivity, and manic symptoms. Similar results were reported by Ratcliffe, Wong, Dossetor, and Hayes (2015) with a sample of 292 children with ASD. About a quarter of these children (n = 76) were also reported to have mild intellectual disability. Specifically, children with lower social skill ratings were also rated as having greater mental health difficulties and more problematic behavior.

The findings reported by Matson, Dempsey, and Rivet (2009) and Ratcliffe et al. (2015) suggest that interventions to develop appropriate social skills and reduce negative social behaviors might also help to prevent or reduce mental health problems among individuals with intellectual disability. In light of the association between social skills and psychopathology, it is possible that interventions to address social skills deficits and excesses might also have some positive collateral effects on mental health. Whether such collateral effects have been examined would therefore seem a pertinent point to consider when evaluating different intervention approaches.

In addition to serious mental health problems, social skills deficits and excesses might also negatively impact on a person's ability to engage in social interactions, participate in social activity, and develop and maintain friendships. Indeed, evidence suggests that individuals with intellectual disabilities generally spend more time alone, report greater degrees of loneliness, participate in fewer social interactions, and have fewer friends than their same-aged peers without disabilities (Bigby & Knox, 2009; Emerson & Hatton, 2008; Lippold & Burns, 2009; Solish, Perry, & Minnes, 2010). Gilmore and Cuskelly (2014) suggested that social skills deficits are one factor that may be contributing to the social isolation of persons with intellectual disability. They hypothesized

that the lack of social skills might contribute to reduced opportunities for social interaction, which could in turn limit opportunities to develop appropriate social skills through modeling. One implication of this hypothesis is that intervention needs to focus not just on addressing social skills deficits, but also on creating increased opportunities for social engagement.

Social Skills and Challenging Behavior

Challenging behavior (e.g., aggression, self-injury, stereotyped mannerisms, and extreme tantrums) are prevalent among people with intellectual disability (Dworschak, Ratz, & Wagner, 2016; Felce & Kerr, 2013). Results from several correlational studies suggest an inverse relation between the frequency and severity of challenging behavior and social functioning among people with intellectual disability. Felce and Kerr (2013), for example, found an inverse relation between challenging behavior and adaptive behavior functioning in a sample of 818 adults with intellectual disability. Specifically, individuals with lower overall adaptive behavior scores, including lower social functioning, showed more frequent and severe challenging behavior. Matson, Fodstad, and Rivet (2009) found a similar relation between the type and degree of social impairment and challenging behavior in a sample of 114 adults with severe to profound intellectual disability. Specifically, the presence of negative social behaviors—such as crying at inappropriate times and excessive attention seeking—was associated with increased levels of aggression, destructive acts, and disruption. In another relevant study, Kearney and Healy (2011) assessed challenging behavior in relation to social skills deficits in a sample of 39 adults with moderate to severe intellectual disability. They found that individuals with severe challenging behavior ". . . scored significantly lower on social skills measures . . ." (p. 1556).

In addition to the relations between challenging behavior and social skills deficits identified in the above cited correlational studies, results from a number of experimental studies suggest that challenging behaviors are often socially motivated, that is, challenging behavior often appears to serve a social function or purpose for the individual (Hanley, Iwata, & McCord, 2003; Iwata et al., 1994). For example, a child with intellectual disability might learn to engage in challenging behavior as a means of (a) recruiting attention from an adult, (b) escaping from or avoiding non-preferred activities, (c) soliciting assistance with difficult tasks, and/or (d) requesting access to preferred objects and activities (Reichle & Wacker, 2017). Such socially motivated challenging behavior is perhaps more likely to emerge in cases where appropriate social skills have failed to develop or where social skills development has been significantly delayed (Sigafoos, Arthur, & O'Reilly, 2003).

In line with evidence from correlational and experimental studies, it is possible that interventions to address social skills deficits and excesses might also have some positive collateral effects on challenging behavior. It is also possible that early introduction of social skills intervention could help to prevent the emergence of frequent and severe challenging behavior. Whether such collateral effects have been examined would therefore seem a pertinent point to consider when evaluating different intervention approaches.

Aim of This Chapter

Given that people with intellectual disability often have major social skills deficits and that these deficits have been linked to mental health problems and co-occurring challenging behavior, it is not surprising that a significant amount of research has focused on the treatment of social skills deficits and excesses of individuals with intellectual disability. This research has led to a number of intervention approaches. The aim of this chapter is to provide an overview of several promising and evidence-based approaches for supporting social skills development in people with intellectual disability.

Overview of Social Skills Intervention Approaches

Individuals with intellectual disability often have considerable difficulty in learning social norms and developing social competence and effective coping skills (Brooks, Floyd, Robins, & Chan, 2015). The learning difficulties associated with intellectual disability might help to account for at least some of the pervasive deficits in social functioning that are characteristic of people with this diagnosis. For example, unlike their typically developing peers, children with intellectual disability may have more difficulty with acquisition of skills through incidental learning processes (Hardman & Drew, 1975), that is, as a by-product of everyday social interactions (Boers, 2018). Given that many social skills appear to be acquired incidentally, a diminished capacity to learn incidentally would be expected to negatively influence social development. Consequently, people with intellectual disability would often seem to require a more intentional approach to social skills development. Intentional learning approaches involve deliberately creating teaching episodes for the purpose of directly addressing a specific issue, such as teaching a specific social skill. Such approaches are generally more structured and systematic than incidental learning approaches. They are also often more intensive, involving frequent intervention opportunities or sessions over an extended period of time (10–12 weeks). A number of structured approaches for addressing social skills deficits and excesses in persons with intellectual disability have been developed and evaluated in applied intervention research. Promising approaches for supporting social skills development in persons with intellectual disability—as well as addressing mental health issues and co-occurring challenging behavior—include (a) cognitive behavior therapy, (b) applied behavior analysis, (c) bibliotherapy, and (d) school curriculum approaches. The following sections provide an overview and summary of research related to each of the aforementioned approaches.

Cognitive Behavior Therapy

Cognitive behavior therapy (CBT) has been widely used to address a range of social adjustment and mental health problems, including anxiety, mood disorders, obsessive-compulsive disorder, aggression, anger management, and disruptive behavior (McQueen et al., 2018; Sturmey & Hersen, 2012; Willner et al., 2013). CBT has also been used to enhance social skills (Laugeson & Park, 2014) and promote greater emotional awareness and regulation (Lindsey, Neilson, & Lawrenson, 1997). CBT has been successfully applied across a wide range of age groups, cultures, diagnostic categories, and settings, including children, adolescents, and adults with intellectual disability (Dagnan & Lindsey, 2004; Dodd et al., 2013; Vereenooghe & Langdon, 2013). Overall, CBT can be classified as a well-established, evidence-based treatment for a range of mental health concerns and a range of populations (David, Cristea, & Hofmann, 2018).

CBT is based on the hypothesis that various mental health issues (e.g., anxiety, depression, and anger control problems) stem from dysfunctional thoughts, beliefs, self-perceptions, and feelings (Beck, 1976). In line with this hypothesis, CBT focuses on teaching individuals to identify and change dysfunctional thought patterns and adopt alternative and more productive courses of action (Percy, Fung, Brown, & Hassiotis, 2017). CBT typically involves implementation of a treatment package that could include efforts to assist the person in (a) identifying and describing specific circumstances that trigger dysfunctional thoughts, (b) determining and practicing appropriate thought patterns, (c) identifying and adopting more socially appropriate responses to triggers, (d) learning how to read social cues, and (d) adjusting performance in light of feedback (Harchik, Sherman, & Sheldon, 1992; Laugeson & Park, 2014; Willner et al., 2013).

There is growing interest and research into the use of CBT as a treatment for people with intellectual disability (Cooney, Tunney, & O'Reilly,

2018; Dodd et al., 2013), but the evidence base on this front is still relatively meager. Vereenooghe and Langdon (2013), for example, searched for studies in this area and identified 13 experimental evaluations into the use of CBT as a treatment for people with intellectual disability. Of these 13 studies, 7 delivered CBT in a small-group format (e.g., 4–8 participants per group), whereas the remaining 6 conducted individual CBT sessions. Most of the 13 CBT studies ($n = 9$) targeted anger management. Other studies targeted depression or depression and anxiety. The amount of CBT delivered varied across the studies, but most studies provided for weekly (1–2 hour) sessions over 12 to 18 weeks. From this meta-analytic review, Vereenooghe and Langdon (2013) concluded that CBT was "efficacious for both anger management and depression" (p. 4085).

The ability to manage anger appropriately could be seen as an important skill for harmonious social interaction and it will therefore be instructive to examine a prototypic study into the use of CBT for anger management in some detail. To this end, Willner et al. (2013) evaluated the effects of a group-based CBT program for improving anger management skills in a sample of adults with mild to moderate intellectual disability. The 179 participants attended 31 different day programs. The day programs were randomly assigned to a CBT or treatment-as-usual condition. Participants in the two conditions were comparable in terms of mean age (37–38 years), IQ (55–59), and adaptive behavior. In addition to anger management issues, clinically significant levels of anxiety and depression were reported among 73% and 34% of the total participants, respectively, whereas 26% were identified as having severe challenging behavior. Outcome measures—including felt responses to anger-provoking situations, coping skills, psychopathology, and challenging behavior—were collected at the time of randomization and then again at 16 weeks and 10 months post-randomization. The CBT intervention consisted of 12 weekly sessions that were supplemented with homework tasks. Group sizes ranged from four to eight adults. During sessions, participants in the CBT groups received instruction from staff, who had received brief training on the use of CBT. These therapists were trained on how to assist the participants with identifying anger-provoking triggers, learning relaxation techniques, and coping with and reducing feelings of anger. Participants also learned to use appropriate assertiveness skills as alternatives to expressing anger. Specific instructional tactics included asking questions (e.g., *What makes you angry?*), role-playing, and describing and analyzing situations that had provoked anger over the previous week, and exploring better ways of coping with those situations. A major strength of this study was the fact that with relatively little training (i.e., three sessions over a single day followed by fortnightly supervision), the regular day program staff were able to implement the CBT program with good fidelity (68.6% overall). While implementation by regular staff is a strength, the overall results of the intervention were somewhat mixed. Specially, there were no significant differences across groups on the primary outcome measure (i.e., participants' self-reported anger in response to hypothetical anger-provoking situations). However, the authors did find significant changes (favoring the CBT group) on anger coping skills and participants' challenging behavior.

In line with the overall conclusions by Vereenooghe and Langdon (2013), these latter findings by Willner et al. (2013) suggest that their group-based CBT intervention was successful for improving anger management and reducing challenging behavior in adults with mild to moderate intellectual disability. This is an important demonstration as it suggests that CBT may be practical for application within the living environments and day programs that many adults with intellectual disabilities are likely to experience (Braddock, Emerson, Felce, & Stancliffe, 2001).

While a number of studies support the use CBT for addressing anger management and depression in adults with mild to moderate intellectual disability, its efficacy across other areas of social functioning and with younger and more severely impaired participants has yet to receive sufficient research attention. One factor

hindering such research could be that CBT is often seen as being beyond the language and cognitive capabilities of younger participants and those with more severe intellectual disability (McQueen et al., 2018). Hronis, Roberts, and Kneebone (2017) noted that the significant attentional, learning, memory, and executive functioning deficits associated with intellectual disability would seem to contraindicate the use of CBT, at least as it is typically applied in clinical psychology practice. However, Hronis et al. (2017) went on to argue that CBT might be effectively modified to account for the various cognitive deficits experienced by people with intellectual disabilities. Proposed modifications include (a) use of visual aids, (b) keeping instructions short and simple, and (c) pre-training on necessary prerequisite skills, such as teaching children to correctly identify emotions (Dagnan, Johoda, & Stenfert Kroese, 2007; Hronis et al., 2017). With respect to the possibility of successfully teaching prerequisite skills, Stauch, Plavnick, Sankar, and Gallagher (2018) demonstrated that a video-based intervention was successful in teaching social perception skills (e.g., correctly identifying, interpreting, and responding to the affective behavior of another person) to five adolescents with autism and/or intellectual disability. Social perception skills are important in their own right and they might also facilitate the CBT process.

In summary, while CBT appears promising for people with intellectual disabilities, there is a clear need for additional research. Most research to date has involved adults with mild to moderate intellectual disability and anger management issues. Future research could therefore focus on children and adolescents and on people with more severe intellectual disability. Future research should also target the wider range of social skills deficits and excesses that are prevalent among individuals with intellectual disability. Research with these foci is needed to establish the generality and limits of CBT as a treatment for people with intellectual disability. Another area for future research would be to evaluate treatment packages that combine aspects of CBT (e.g., self-monitoring, emotional regulation, relaxation training) with other evidence-based approaches for promoting social skills development among individuals with intellectual disability.

Applied Behavior Analysis

Applied behavior analysis (ABA) has had, and continues to have, a major influence in intellectual disability services (Condillac & Baker, 2017; Remington, 1998). One of its main influences has been in the area of intervention, that is the design, development, and delivery of habilitation and educational programs (Ardila, 2001; Reid, 1987). Indeed, since the 1960s, educational and habilitation programs serving children, adolescents, and adults with intellectual disability have increasingly adopted ABA-based approaches (Neidert, Dozier, Iwata, & Hafen, 2010). For such individuals, ABA-based interventions are frequently used for the development of adaptive skills, including social skills, and in the treatment of challenging behavior (Singh, 2016; Sturmey & Didden, 2014).

Behavior analytic interventions are largely derived from principles of operant conditioning (Ardila, 2001; Sukhodolsky & Butter, 2007). The basic operant conditioning principles underpinning ABA-based interventions include (a) reinforcement, (b) extinction, (c) punishment, (d) stimulus control, and (e) shaping, chaining, response prompting, and fading (Duker, Didden, & Sigafoos, 2004; Skinner, 1953; Sundel & Sundel, 2018). However, as noted by Sukhodolsky and Butter (2007) behaviorally based social skills interventions also often make use of procedures (e.g., modeling, self-reinforcement, vicarious reinforcement) derived from social learning theory (Bandura, 1977). In addition, ABA-based interventions have incorporated additional elements (e.g., self-monitoring) associated with CBT (Foxx, McMorrow, & Schloss, 1983).

Matson, Kazdin, and Esveldt-Dawson (1980) conducted an important early study that demonstrated positive outcomes from a social skills training program that was based on operant conditioning principles and social learning theory.

The 14-week intervention involved daily sessions in which the participants (two, 11- to 12-year-old boys with moderate intellectual disability) received verbal instruction, social reinforcement, and performance feedback. Teaching also made use of modeling and role-playing of appropriate social behavior. With intervention, the children showed gains in their conversational skills (e.g., content, intonation). They also learned a number of subtle aspects of social interaction, such as making appropriate facial expressions and eye contact, correct social proximity, and use of appropriate gestures.

Since Matson et al.'s (1980) pioneering work, a considerable amount of research evidence has accumulated to support the use of similar ABA-based interventions for teaching social skills to individuals with intellectual and other developmental disabilities (Carter & Hughes, 2005; Gresham, 2016; Höher Camargo et al., 2016; Reichow & Volkmar, 2009; Schneider, Goldstein, & Parker, 2008; Sturmey, 2014; Wang, Parilla, & Cui, 2013; Watkins et al., 2016). A common thread in these studies is the use of a flexible ABA-based approach in which a rather generic set of operant procedures are combined with additional procedures associated with CBT and social learning theory. Commonly used procedures in these social skills training programs are (a) verbal instruction, (b) modeling, (c) role-playing, (d) reinforcement, and (e) error correction (Matson & Ollendick, 1988; Sukhodolsky & Butter, 2007).

ABA-based social skills interventions generally progress through a number of sequential steps, such as: (a) defining target behaviors in observable and measurable terms; (b) creating multiple, discrete, and structured learning opportunities; (c) presenting clear and precise instructional cues or prompts to ensure that the target behavior occurs in response to each learning opportunity; (d) preventing/correcting errors as necessary; (e) systematically fading prompts to promote independent use of targeted skills; (f) scheduling performance contingent reinforcement to promote acquisition and maintenance of targeted skills; and (g) programming for generalization to ensure newly acquired skills occur in response to a wide range of relevant environmental conditions (Panyan, 1998; Stokes & Baer, 1977).

In addition to the systematic, structured application of a generic set of instructional procedures, a number of innovations in ABA-based social skills training programs have been developed. O'Reilly et al. (2004), for example, added a problem-solving component to a generic ABA-based social skills training program in a study involving five adults with mild intellectual disability. Participants were taught to decode and evaluate conflict situations and then decide on an appropriate course of action, which in turn enabled them to better manage conflict situations and respond appropriately to corrective feedback. Results from several additional studies support the use of similar problem-solving interventions for enhancing social skills and reducing challenging behavior in persons with intellectual disabilities (Anderson & Kazantzis, 2008; Loumidis & Hill, 1997; O'Connor, 1996).

Representing another innovation, Olçay Gül (2016) used video modeling to teach social skills to three young adults with mild to moderate intellectual disability (IQ scores between 35 and 55). Specifically, participants watched a scripted video showing a model performing the target skills, which consisted of making eye contact with a person who was not feeling well and making an empathetic comment (e.g., *Get better soon*.) The results showed that after only two to three video viewings, participants were consistently performing the targeted social skills. These results are consistent with the generally positive outcomes that have been reported in numerous other video modeling studies (Walton & Ingersoll, 2013; Weiss, 2013).

Foxx and colleagues reported success with another innovative approach to social skills training (Foxx et al., 1983; Foxx & Faw, 1992; Foxx, McMorrow, Bittle, & Ness, 1986; Foxx, McMorrow, & Mennemeier, 1984). In these studies, a range of social skills (e.g., giving compliments, politeness, and appropriate responses to social confrontation) improved as a function of playing a board game. As part of the game, participants responded to a number of

question cards and received feedback and reinforcement on their answers. Participants were also taught self-monitoring skills in relation to individualized performance goals. The gains made with intervention also showed good generalization and maintenance. This board game approach represents a practical and cost-effective means of teaching social skills. The approach would also seem relatively easy for teachers and front-line staff to implement because it makes use of standardized (generic) teaching procedures and is intended for use in a group-training format. By simply incorporating different question cards/scenarios into the game, the intervention can be individualized to target a range of social skills.

In summary, operant conditioning principles and associated ABA instructional procedures have been packaged and applied in various combinations and configurations to achieve a range of important learning and developmental outcomes for persons with intellectual disability. This range includes teaching appropriate social skills and treatment of excessive/problematic social behavior and other forms of challenging behavior (Condillac & Baker, 2017; Singh, 2016; Sturmey & Didden, 2014; Watkins et al., 2016). ABA-based social skills interventions commonly employ a generic set of operant conditioning procedures and often also incorporate procedures associated with CBT and social learning theory. A strong research base supports the use of ABA-based interventions for addressing the social skills deficits and excesses associated with intellectual disability.

Bibliotherapy

Bibliotherapy refers to the therapeutic use of books and other literary sources (Sullivan & Strang, 2002/2003). The following example illustrates the process. A group of school children read a short story about a girl who is being teased and bullied and who—in the course of the story—learns to respond successfully to her tormenters. After reading the story, the teacher discusses it with the children, checking for their level of comprehension, clarifying any misunderstandings, and highlighting the story's main lessons. With this guidance from the teacher, the children learn to identify and define instances of teasing and bullying. They also talk about the character's emotional state. Furthermore, the teacher makes a special point of prompting the children to identify specific coping skills used by the character. The teacher might even model these coping skills and have the children practice using those skills in role-play scenarios (Lamb, Bigler, Liben, & Green, 2009). The hoped for outcome from all of these efforts is that the children will now be better equipped to respond successfully to teasing and bullying.

An underlying premise of bibliotherapy is that reading pertinent literature about a particular issue, concern, or problem—followed by guided discussion of that literature—can promote positive changes in the reader's knowledge, understanding, attitudes, and behavior (Jack & Ronan, 2008). Improved self-esteem and the ability to adjust to, and cope with, significant concerns or problems are also some of the anticipated outcomes from bibliotherapy (Iaquinta & Hipsky, 2006; Jack & Ronan, 2008). For example, reading positive stories about different disability conditions (e.g., attention-deficit hyperactivity disorder, intellectual disability, and autism spectrum disorders) might improve readers' understanding of those different conditions and improve their attitudes towards people with such conditions (Iaquinta & Hipsky, 2006; Provost, 2017).

The practice of bibliotherapy involves more than just reading relevant books and/or other literary sources. While reading the source material is a critical step in the process, researchers have emphasized the critical role played by subsequent guided discussion of the content (Jack & Ronan, 2008). Forgan (2002), for example, suggested a four-step process consisting of: (a) pre-reading, (b) guided/supported reading, (c) post-reading discussion, and (d) practice with feedback. Cartledge and Kiarie (2001) presented a refinement of Forgan's (2002) model for teaching social skills to children and adolescents with disabilities. Their refinements consisted of (a)

identifying one or more specific intervention targets (e.g., resisting peer pressure) and reading literature that explicitly exemplifies corresponding social skills (e.g., speaking assertively), (b) ensuring students understand the story with the teacher clarifying as necessary, (c) ensuring students learn the specific social skills exemplified in the story, (d) creating role-play scenarios for children to practice these new social skills, and (e) promoting maintenance by additional reading and practice. Additional structure, such as outlined above, might be especially indicated when providing bibliotherapy to younger children or persons with intellectual and other developmental disabilities. In addition, when working with children with intellectual disability, Cartledge and Kiarie (2001) recommended that the stories be kept simple, short, and easy to comprehend. Reading material should also be developmentally and culturally appropriate. For children with limited reading ability, teachers can select or create picture books with minimal text. Non-readers will also, of course, need to have the story read to them or access an audio recording of the story.

Historically, bibliotherapy has most often been conceptualized as a classic psychodynamic process involving three underlying mechanisms: (a) identification, (b) catharsis, and (c) insight (Iaquinta & Hipsky, 2006; Sullivan & Strang, 2002/2003). The process will presumably be more relevant and effective when readers share some affinity and thus identify with the storylines and characters. Catharsis is said to be achieved when the story is inspirational and motivates positive behavior change. Finally, insight—in the sense of gaining new understandings and effective coping mechanisms—is presumed to arise as a result of reading and discussing selected literature.

While identification, catharsis, and insight are considered to be the main underlying principles, Mehdizadeh and Khosravi (2018) suggested that bibliotherapy involves processes similar to those that underpin CBT. Several descriptions of bibliotherapy have also made reference to the use of various techniques (e.g., role-playing, modeling, and reinforcement) associated with CBT, social learning, and applied behavior analysis (Cartledge & Kiarie, 2001; Forgan, 2002; Iaquinta & Hipsky, 2006; Mehdizadeh & Khosravi, 2018). Brinton and Fujiki (2017), for example, recommended the use of prompting (e.g., asking questions to highlight critical story elements), role-playing (e.g., acting out story elements), and rehearsal (i.e., practicing specific behaviors that were illustrated in the story) when using bibliotherapy with children with limited language ability, which is often true of children with intellectual disability.

Although various forms of bibliotherapy have been around for hundreds of years (see Jack & Ronan, 2008 for a review of its history), there are surprisingly few well-controlled studies into its effectiveness. Jack and Ronan (2008) noted that the literature base is largely characterized by uncontrolled studies and anecdotal case reports. Additionally, the overall results have been fairly mixed. Studies more specifically targeting social skills and involving persons with intellectual or other developmental disabilities have also yielded mixed results. Taft, Hotchkiss, and Lee (2016), for example, reviewed five bibliotherapy studies involving students with emotional and behavioral disorders. They found positive outcomes on measures of self-esteem and challenging behavior. In contrast, Reynhout and Carter (2008) evaluated the effects of a streamlined (Social Stories™; Gray & Garand, 1993) version of bibliotherapy on the social participation (attending to the teacher) of an 8-year-old girl (Debbie) with moderate intellectual disability and autism spectrum disorder. Debbie's social story had four pages with one photograph (e.g., Debbie attending to a small-group book reading activity directed by the teacher) and one sentence (e.g., "I will enjoy the story if I look at the book.") on each page. The intervention procedure involved reading the social story and checking on Debbie's comprehension prior to her entering the small-group activity. Despite 28 daily reading sessions, Debbie's level of attending showed no improvement and she also failed to show any correct responses to the comprehension questions. In a second case study, Reynhout and Carter (2007) reported modest reductions in

an excess social behavior (i.e., making noise by disruptive tapping of hands) when a Social Stories™ intervention was implemented in a classroom setting. The participant in this study was an 8-year-old boy named Adam with mild-to-moderate intellectual disability. It is important to note that unlike Debbie, Adam showed a high level (80–100% correct) of story comprehension, which might account for the differential impact that this intervention had on these two children. In fact, differences in participant's comprehension abilities might account for the mixed results that plague the research literature on bibliotherapy.

Intuitively, it makes sense that comprehension of the literature being read would be a prerequisite for successful use of bibliotherapy and related interventions, such as Social Stories™. Consequently, bibliotherapy would seem best suited to individuals with mild to moderate intellectual disability and relatively good receptive language ability. For such individuals, greater levels of comprehension might be obtained by using short, simple stories and visual supports (e.g., photographs or video) to illustrate the story's main concepts.

In summary, the evidence base supporting bibliotherapy is not very strong. In particular, its value as a social skills intervention for persons with people with intellectual disability has yet to be established as the field lacks a sufficient number of high quality studies. Still, bibliotherapy might be of some benefit when the selected literature is "not too intellectually demanding" (Jack & Ronan, 2008, p. 165) and suited to the person's level of comprehension. Even though its current evidence base is relatively limited, bibliotherapy would seem to represent a fairly benign intervention approach that is perhaps unlikely to cause any harm. On the other hand, Visser Knoth (2006) cautioned that some people might become disheartened if their own problems are not as easily resolved as they were in the story. Clinicians therefore need to communicate realistic expectations during their guided discussions of the literature.

For addressing social skills deficits and excesses of persons with intellectual disability, bibliotherapy might be a useful component of a broader intervention approach that also includes well-established strategies based on CBT and applied behavior analytic approaches. Bibliotherapy could be seen as particularly well suited to classroom use as noted by Cartledge and Kiarie (2001). Indeed, its two main components (i.e., reading and guided discussion) represent standard educational practices that can be incorporated into a range of curriculum areas. In fact, selecting, reading, and discussing developmentally appropriate literature for a wide range of curriculum areas is one of a teacher's primary roles (Catalano, 2008).

School Curriculum Approaches

A number of school curricula and school-based intervention programs have been developed for teaching social skills. Some of the more widely used and well-researched programs include (a) *Peacemakers* (Johnson & Johnson, 2004), (b) The *PEERS® Method* (Laugeson, 2014), and (c) *I Can Problem Solve* (Shure, 2001a). These programs generally include scripted lesson plans, accompanying teaching procedures, and detailed instructions to guide teachers' implementation. Many of these programs appear promising for enhancing the social competence of students with intellectual disability.

The *I Can Problem Solve* (ICPS; Shure, 2001a) program, for example, consists of 59 structured lessons that are intended to enhance children's functioning across a range of skill areas (e.g., communication, problem solving, and prosocial behavior). The lessons also address a variety of learning and behavior problems, such as impulsivity, aggression, and social withdrawal. The scripted didactic lessons aim to familiarize children with key problem-solving strategies (e.g., anticipating the consequences of one's actions and identifying alternative solutions to problems). Lessons are supplemented with classroom games and interactions for practicing problem-solving strategies. The program is intended to be integrated into a range of curriculum areas, such as math, reading, and social

studies. Three versions of the program have been developed, one for each of three different age groups (i.e., kindergarten/preschool, early primary, and intermediate primary up Grade 6). Several evaluations of the ICPS curriculum (Boyle & Hassett-Walker, 2008; Dincer & Guneysu, 1997; Dos Santo Elias, Marturano, De Almeida Motta, & Giurlani, 2003; Feis & Simons, 1985; Shure & Spivack, 1982) have reported improvements in children's overall adjustment and prosocial behavior, as well as reductions in impulsivity and aggression. Although the ICPS program was not specifically designed for children with intellectual disability, it has been successfully used with young, disadvantaged students (Feis & Simons, 1985; Shure, 2001b). It might therefore be suitable for students with intellectual disability who are functioning at the preschool or primary school level.

Margalit (1995) reported on a computer-based curriculum that was specifically designed for teaching social skills to students with mild intellectual disability. The computer program, *I Found a Solution* (Margalit, 1990), presents a series of 24 conflict scenarios (e.g., a student is feeling restless and bored at school) and asks students to choose from among a number of possible solutions. Some of the solutions are deemed socially appropriate (e.g., the student should ask the teacher for help), whereas others are viewed as socially inappropriate (e.g., the student should hit and swear). In addition to completing the computer-presented scenarios, teachers also engage the students in relevant small-group discussions and provide homework tasks and role-play activities related to the conflict situations. An evaluation was conducted that involved 38 students in an experimental group and 35 students in a control group. The students were 11- to 15-year-olds and had mild intellectual disability. Results showed significant gains for the experimental students based on teacher-, peer-, and self-reports. Gains were reported with respect to cooperation, self-control, assertion, peer acceptance, and reductions in behavioral difficulties. Interestingly, both groups had relatively high ratings on loneliness both prior to and after the intervention. This latter finding suggests the need for interventions that specifically address loneliness.

Along these lines, the *PEERS® Method* (Laugeson, 2014) aims to teach social skills related to making and keeping friends. This group-based treatment program involves the use of a range of strategies, such as (a) didactic instruction, (b) comprehension assessment via question asking, (c) modeling, (d) role-playing, (e) practicing new skills, and (f) performance feedback. These strategies are used to teach a range of skills related to both making friends (e.g., entering into a conversation by making relevant comments) and maintaining friendships (e.g., giving compliments). Laugeson and Park (2014) listed a number of studies that have reported successful use of the *PEERS® Method* for improving the friendship-related skills of adolescents and adults with intellectual and other developmental disabilities.

O'Handley, Ford, Radley, Helbig, and Wimberly (2016) reported on the use of another school-based intervention program for teaching social skills. Four adolescents with mild to moderate intellectual disability completed the *Superheroes Social Skills* program (Jenson et al., 2011). This manualized program includes a range of materials (e.g., lesson plans, cartoon videos, posters of classroom rules) and associated instructional strategies (e.g., didactic lessons, videos modeling of target skills, live modeling of target skills by instructors, and role-playing with feedback). The program involved a total of six, 90-min sessions. In a multiple-baseline across behaviors design, O'Handley et al. (2016) demonstrated gains in social conversation, turn taking, and expressing wants and needs for all four participants following a 3-week period of intervention. The newly acquired skills were also maintained during follow-up checks conducted at 3- and 5-weeks post-intervention. These results, while limited to a small number of participants, suggest that the *Superheroes Social Skills* program may be an effective option for teaching social communication skills to adolescents with mild to moderate intellectual disability. The program also appears to be a practical option for use in high school classrooms.

In another relevant study, Adeniyi and Omigbodun (2016) evaluated an adapted version of the *Explore Social Skills Curriculum* (Kinney, 2012) for use in a Nigerian residential school for students with intellectual disability. The curriculum was adapted to suit the Nigerian cultural context by having teachers review and change aspects of the content to ensure cultural relevance of the skills being taught. Teachers then implemented the adapted (15-lesson) curriculum over an 8-week period. Lessons occurred three times per week, and each lesson lasted 45 min. The curriculum targeted a range of social skills (e.g., responding to teasing, joining a peer group, respecting friends). Teachers first gave a narrative overview of the lesson topic and then involved the students in role-play activities (e.g., role-playing the social skills for meeting a new person). The effects of this curriculum were evaluated using the Matson Evaluation of Social Skills for Individuals with Severe Retardation (MESSIER; Matson, 1995). Thirty (12- to 19-year-old) students with moderate intellectual disability (mean IQ = 44) participated in the evaluation. Analysis of the pre-post MESSIER scores revealed significant improvement in the participants' social skills. These findings are encouraging in that they suggest the potential value of a relatively low-intensity, teacher-delivered social skills intervention for students with moderate intellectual disability. The study is unique in illustrating a process for cultural adaptation of curriculum content. However, the lack of a control group reduces the certainty of evidence generated by this study.

In summary, education professionals can draw upon a number of research-based curricula and school-based intervention programs to address the social skill deficits and excesses of students with intellectual disability. Programs of this type could be quite appealing to educators because of the accompanying structure, lesson plans, materials, standardized teaching procedures, and implementation guidelines. School curricula could be viewed as a universal-level intervention approach. Universal interventions typically involve providing the same teaching program and instructional supports to all students in the school (Greenberg & Abenavoli, 2017). CBT and applied behavior analytic approaches, in contrast, are generally used in a more targeted and individualized manner. There are obvious practical and cost advantages to universal interventions. Such approaches have been successfully used with students with mild to moderate intellectual disability. However, there appears to be a relative dearth of universal interventions/school curricula for addressing social skill deficits and excesses of students with more severe intellectual disability. This could reflect the fact that students with severe to profound intellectual disability generally have more extreme learning difficulties and social skills deficits. They may therefore require more targeted, specialized, intensive, and individualized interventions, perhaps based primarily on operant conditioning principles.

Summary and Conclusion

This chapter explored the link between social skills and mental health disorders and challenging behavior. A number of studies point to a close link between the degree of social skills impairment and the presence of co-occurring mental health problems. Social skills deficits are also associated with an increased risk of challenging behavior. Conceptually, some behaviors are challenging because they occur at rates that are considered excessive or occur at times and/or in settings that make the behaviors contextually and socially inappropriate. The link between social skills deficits and mental health problems and challenging behaviors highlight the importance of providing effective intervention to promote the social development of people with intellectual disability.

Promising intervention approaches for addressing social skills deficits and excesses associated with intellectual disability include (a) cognitive behavior therapy (CBT), (b) applied behavior analysis, (c) bibliotherapy, and (d) school curriculum/school-based intervention programs. Distinguishing features of these differing approaches were described, and key studies illustrating applications and evaluations of each

approach were provided. The specific applications of these different intervention approaches were often found to share a number of well-established instructional tactics, such as the use of verbal and written instructions, modeling, role-playing, practice, and feedback/reinforcement. Consequently, these four approaches are not necessarily mutually exclusive. Rather they would appear to be quite compatible and perhaps even complementary. Aspects of these different approaches might therefore be combined into flexible treatment packages that can be tailored to suite an individual's unique needs and circumstances.

CBT, applied behavior analysis, bibliotherapy, and school curriculum programs are each promising approaches for promoting the social development of persons with intellectual disability. Future research should therefore aim towards supporting the uptake of these approaches by key stakeholders (e.g., parents, teachers, and front-line staff) under real-world conditions. More research is also needed into the use of social skills interventions that aim to prevent or reduce mental health problems and/or challenging behavior. Promoting the social development and addressing the mental health concerns and challenging behavior of persons with intellectual disability may depend, to some extent, on the amount and quality of social skills intervention that they receive.

References

Adeniyi, Y. C., & Omigbodun, O. O. (2016). Effects of a classroom-based intervention on the social skills of pupils with intellectual disability in Southwest Nigeria. *Child and Adolescent Psychiatry and Mental Health, 10*, 1–12. https://doi.org/10.1186/s13034-016-0118-3

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

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

Anderson, G., & Kazantzis, N. (2008). Social problem-solving skills training for adults with mild intellectual disability: A multiple case study. *Behaviour Change, 25*, 97–108. https://doi.org/10.1375/bech.25.2.97

Ardila, R. (2001). Behavior analysis, applied. In N. J. Smelser & P. B. Baltes (Eds.), *International encyclopedia of the social and behavioral sciences* (pp. 1064–1069). Amsterdam, The Netherlands: Elsevier/Pergamon. Retrieved from: https://doi.org/10.1016/B0-08-043076-7/01301-2

Bandura, A. (1977). *Social learning theory*. Engelwood Cliffs, NJ: Prentice-Hall.

Beck, A. (1976). *Cognitive therapy and the emotional disorders*. New York, NY: International Universities Press.

Bigby, C., & Knox, M. (2009). "I want to see the queen": Experiences of service use by ageing people with an intellectual disability. *Australian Social Work, 62*, 216–231. https://doi.org/10.1080/03124070902748910

Boers, F. (2018). Intentional versus incidental learning. In J. I. Liontas (Ed.), *The TESOL encyclopedia of English language teaching* (pp. 1–6). New York, NY: John Wiley & Sons. https://doi.org/10.1002/9781118784235.eelt0074

Boyle, D., & Hassett-Walker, C. (2008). Reducing overt and relational aggression among young children: The results of a two-year outcome evaluation. *Journal of School Violence, 7*, 27–42. https://doi.org/10.1300/J202v07n01_03

Braddock, D., Emerson, E., Felce, D., & Stancliffe, R. J. (2001). Living circumstances of children and adults with mental retardation or developmental disabilities in the United States, Canada, England and Wales, and Australia. *Mental Retardation and Developmental Disabilities Research Reviews, 7*, 115–121. https://doi.org/10.1002/mrdd.1016

Brinton, B., & Fujiki, M. (2017). The power of stories: Facilitating social communication in children with limited language abilities. *School Psychology International, 38*, 523–540. https://doi.org/10.1177/0143034317713348

Brooks, B. A., Floyd, F., Robins, D. L., & Chan, W. Y. (2015). Extracurricular activities and the development of social skills in children with intellectual disabilities and specific learning disabilities. *Journal of Intellectual Disability Research, 59*, 678–687. https://doi.org/10.1111/jir.12171

Buntinx, W. (2016). Adaptive behaviour and support needs. In A. Carr, C. Linehan, G. O'Reilly, P. Noonan Walsh, & J. McEvoy (Eds.), *The handbook of intellectual disability and clinical psychology practice* (pp. 107–135). London, UK: Routledge.

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*, 179–193. https://doi.org/10.2511/rpsd.30.4.179

Cartledge, G., & Kiarie, M. W. (2001). Learning social skills through literature for children and adolescents. *Teaching Exceptional Children, 34*(2), 40–47. https://doi.org/10.1177/004005990103400206

Catalano, A. (2008). Making a place for bibliotherapy on the shelves of a curriculum materials center: The

case for helping pre-service teachers use developmental bibliotherapy in the classroom. *Education Libraries: Children's Resources, 31*, 17–22. https://doi.org/10.26443/el.v31i3.258

Condillac, R. A., & Baker, D. (2017). Behavioral intervention. In M. L. Wehmeyer, I. Brown, M. Percy, K. A. Shogren, & W. L. A. Fung (Eds.), *A comprehensive guide to intellectual and developmental disabilities* (pp. 401–411). Baltimore, MD: Paul H. Brookes Publishing.

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 intellectual disability. *Journal of Applied Research in Intellectual Disabilities, 31*, 23–42. https://doi.org/10.1111/jar.12365

Dagnan, D., Johoda, A., & Stenfert Kroese, B. (2007). Cognitive behaviour therapy. In A. Carr, G. O'Reilly, P. Noonan Walsh, & J. McEvoy (Eds.), *The handbook of intellectual disability and clinical psychology practice* (pp. 281–299). London, UK: Routledge.

Dagnan, D., & Lindsey, W. (2004). Cognitive therapy with people with learning disabilities. In E. Emerson, C. Hatton, T. Thompson, & T. Parmenter (Eds.), *International handbook of applied research in intellectual disabilities* (pp. 517–530). Chichester, UK: Wiley.

David, D., Cristea, I., & Hofmann, S. G. (2018). Why cognitive behavioral therapy is the current gold standard of psychotherapy. *Frontiers in Psychiatry, 9*, 4. https://doi.org/10.3389/fpsyt.2018.00004

Dincer, C., & Guneysu, S. (1997). Examining the effects of problem solving training on the acquisition of interpersonal problem-solving skills by 5-year-old children in Turkey. *International Journal of Early Years Education, 5*, 37–46. https://doi.org/10.1080/0966976970050104

Dodd, K., Austin, K., Baxter, L., Jennison, J., Kenny, M., Lippold, T., … Wilcox, E. (2013). Effectiveness of brief training in cognitive-behaviour therapy techniques for staff working with people with intellectual disabilities. *Advances in Mental Health and Intellectual Disabilities, 7*, 300–311. https://doi.org/10.1106/AMHID-06-2013-0037

Dos Santo Elias, L. C., Marturano, E. M., De Almeida Motta, A. M., & Giurlani, A. G. (2003). Treating two boys with low school achievement and behavior problems: Comparison of two kinds of intervention. *Psychological Reports, 92*, 105–116. https://doi.org/10.2466/pr0.2003.92.1.105

Duker, P., Didden, R., & Sigafoos, J. (2004). *One-to-one training: Instructional procedures for learners with developmental disabilities*. Austin, TX: Pro-Ed.

Dworschak, W., Ratz, C., & Wagner, M. (2016). Prevalence and putative risk markers of challenging behavior in students with intellectual disabilities. *Research in Developmental Disabilities, 58*, 94–103. https://doi.org/10.1016/j.ridd.2016.08.006

Emerson, E., & Hatton, C. (2008). *People with learning disabilities in England*. Lancaster, UK: Centre for Disability Research.

Feis, C. L., & Simons, C. (1985). Training preschool children in interpersonal cognitive problem solving skills. A replication. *Prevention in Human Services, 3*, 59–70. https://doi.org/10.1300/J29v03n04_07

Felce, D., & Kerr, M. (2013). Investigating low adaptive behaviour and presence of the triad of impairments characteristic of autistic spectrum disorder as indicators of risk for challenging behaviour among adults with intellectual disabilities. *Journal of Intellectual Disability Research, 57*, 128–138. https://doi.org/10.1111/j.1365-2788.2011.01524.x

Forgan, J. W. (2002). Using bibliotherapy to teach problem solving. *Intervention in School and Clinic, 38*, 75–82. https://doi.org/10.1177/10534512020380020201

Foxx, R. M., & Faw, G. D. (1992). An eight-year follow-up of three social skills training studies. *Mental Retardation, 50*, 63–66.

Foxx, R. M., McMorrow, M. J., Bittle, R. G., & Ness, J. (1986). An analysis of social skills generalization in two natural settings. *Journal of Applied Behavior Analysis, 19*, 299–305. https://doi.org/10.1901/jaba.1986.19-299

Foxx, R. M., McMorrow, M. J., & Mennemeier, M. (1984). Teaching social/vocational skills to retarded adults with a modified table game: An analysis of generalization. *Journal of Applied Behavior Analysis, 17*, 343–352. https://doi.org/10.1901/jaba.1984.17-343

Foxx, R. M., McMorrow, M. J., & Schloss, C. N. (1983). Stacking the deck: Teaching social skills to retarded adults with a modified table game. *Journal of Applied Behavior Analysis, 16*, 157–170. https://doi.org/10.1901/jaba.1983.16-157

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*, 192–199. https://doi.org/10.1111/jppi.12089

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), 1–10. https://doi.org/10.1177/108835769300800101

Greenberg, M. T., & Abenavoli, R. (2017). Universal interventions: Fully exploring their impacts and potential to produce population-level impacts. *Journal of Research on Educational Effectiveness, 10*, 40–67. https://doi.org/10.1080/19345747.2016.1246632

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

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

Harchik, A. E., Sherman, J. A., & Sheldon, J. B. (1992). The use of self-management procedures by people with developmental disabilities: A brief review. *Research in Developmental Disabilities, 13*, 211–227. https://doi.org/10.1016/0891-4222(92)90026-3

Hardman, M. L., & Drew, C. J. (1975). Incidental learning in the mentally retarded: A review. *Education and Training of the Mentally Retarded, 10*, 3–9. https://www.jstor.org/stable/23876490

Höher Camargo, S. P., Rispoli, M., Ganz, J., Hong, E. R., Davis, H., & Mason, R. (2016). Behaviorally based interventions for teaching social interaction skills to children with ASD in inclusive settings: A meta-analysis. *Journal of Behavioral Education, 25*, 223–248. https://doi.org/10.1007/s10864-015-9240-1

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*, 105–113. https://doi.org/10.1007/s00127-009-0046-0

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*, 189–207. https://doi.org/10.1111/bjc.12133

Iaquinta, A., & Hipsky, S. (2006). Practical bibliotherapy strategies for the inclusive elementary classroom. *Early Childhood Education Journal, 34*, 209–213. https://doi.org/10.1007/s10643-006-0128-5

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. https://doi.org/10.1901/jaba.1994.27-215

Jack, S. J., & Ronan, K. R. (2008). Bibliotherapy: Practice and research. *School Psychology International, 29*, 161–182. https://doi.org/10.1177/0143034308090058

Jenson, W. R., Bowen, J., Clark, E., Block, H. M., Gabrielsen, T., Hood, J., … Springer, B. J. (2011). *Superheroes social skills: A multimedia program*. Eugene, OR: Pacific Northwest Publishing.

Johnson, D. W., & Johnson, R. T. (2004). Implementing the "teaching students to be peacemakers program". *Theory Into Practice, 43*, 68–79. https://doi.org/10.1207/s15430421tip4301_0

Kearney, D. S., & Healy, O. (2011). Investigating the relationship between challenging behavior, co-morbid psychopathology and social skills in adults with moderate to severe intellectual disabilities in Ireland. *Research in Developmental Disabilities, 32*, 1556–1563. https://doi.org/10.1016/j.ridd.2011.01.053

Kinney, J. (2012). *Explore social skills curriculum*. Verona, WI: Attainment Company.

Lamb, L. M., Bigler, R. S., Liben, L. S., & Green, V. A. (2009). Teaching children to confront peers' sexist remarks: Implications for theories of gender development and educational practice. *Sex Roles, 61*, 361–382. https://doi.org/10.1007/s1199-009-9634-4

Laugeson, E. A. (2014). *The PEERS curriculum for school-based professionals: Social skills training for adolescents with autism spectrum disorder*. New York, NY: Routledge.

Laugeson, E. A., & Park, M. N. (2014). Using a CBT approach to teach social skills to adolescents with autism spectrum disorder and other social challenges: The PEERS® method. *Journal of Rational-Emotive Cognitive-Behavior Therapy, 32*, 84–97. https://doi.org/10.1007/s10942-014-0181-8

Lindsey, W., Neilson, C., & Lawrenson, H. (1997). Cognitive-behavioural therapy for anxiety in 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, UK: Routledge.

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*, 463–473. https://doi.org/10.1111/j.1365-2788.2009.01170.x

Loumidis, K. S., & Hill, A. (1997). Training social problem-solving skills to reduce maladaptive behaviours in intellectual disability groups: The influence of individual difference factors. *Journal of Applied Research in Intellectual Disabilities, 10*, 217–237. https://doi.org/10.1111/j.1468-3148.1997.tb00018.x

Margalit, M. (1990). *Effective technology integration for disabled children: The family perspective*. New York, NY: Springer-Verlag.

Margalit, M. (1995). Effects of social skills training for students with an intellectual disability. *International Journal of Disability, Development and Education, 42*, 75–85. https://doi.org/10.1080/0156655950420108

Matson, J. L. (1995). *The Matson evaluation of social skills for individuals with severe retardation (MESSIER)*. Baton Rouge, LA: Disability Consultants, LLC.

Matson, J. L., Dempsey, T., & Rivet, T. T. (2009). The interrelationships of psychopathology symptoms on social skills in adults with autism or PDD-NOS and intellectual disability. *Journal of Developmental and Physical Disability, 21*, 39–55. https://doi.org/10.1007/s10882-008-9124-6

Matson, J. L., Fodstad, J. C., & Rivet, T. T. (2009). The relationship of social skills and problem behaviors in adults with intellectual disability and autism or PDD-NOS. *Research in Autism Spectrum Disorder, 3*, 258–268. https://doi.org/10.1016/j.rasd.2008.07.001

Matson, J. L., Kazdin, A. E., & Esveldt-Dawson, K. (1980). Training interpersonal skills among mentally retarded and socially dysfunctional children. *Behaviour Research and Therapy, 18*, 419–427. https://doi.org/10.1016/0005-7967(80)90007-8

Matson, J. L., & Ollendick, T. H. (1988). *Enhancing children's social skills: Assessment and training*. London, UK: Pergamon Press.

Matson, J. L., & Shoemaker, M. (2009). Intellectual disability and its relationship to autism spectrum disorder. *Research in Developmental Disabilities, 30*, 1107–1114. https://doi.org/10.1016/j.ridd.2009.06.003

Matson, J. L., Terlonge, C., & Minshawi, N. F. (2008). Children with intellectual disabilities. In R. J. Morrie

& T. R. Kratochwill (Eds.), *The practice of child therapy* (4th ed.). New York, NY: Lawrence Erlbaum.

McQueen, M., Blinkhorn, A., Broad, A., Jones, J., Naeem, F., & Ayub, M. (2018). Development of a cognitive behavioural therapy-based guided self-help intervention for adults with intellectual disability. *Journal of Applied Research in Intellectual Disability, 31*, 885–896. https://doi.org/10.1111/jar.12447

Mehdizadeh, M., & Khosravi, Z. (2018). An inquiry into the effectiveness of bibliotherapy for children with intellectual disability. *International Journal of Developmental Disabilities*. https://doi.org/10.1080/20473869.2018.1466509

Neidert, P. L., Dozier, C. L., Iwata, B. A., & Hafen, M. (2010). Behavior analysis in intellectual and developmental disabilities. *Psychological Services, 7*, 103–113. https://doi.org/10.1037/a0018791

O'Connor, W. (1996). A problem-solving intervention for sex offenders with an intellectual disability. *Journal of Intellectual and Developmental Disability, 21*, 219–235. https://doi.org/10.1080/13668259600033151

O'Handley, R. D., Ford, W. B., Radley, K. C., Helbig, K. A., & Wimberly, J. K. (2016). Social skills training for adolescents with intellectual disabilities: A school-based evaluation. *Behavior Modification, 40*, 541–567. https://doi.org/10.1177/0145445516629938

O'Reilly, M. F., Lancioni, G. E., Sigafoos, J., O'Donoghue, D., Lacey, C., & Edrisinha, C. (2004). Teaching social skills to adults with intellectual disabilities: A comparison of external control and problem-solving interventions. *Research in Developmental Disabilities, 25*, 399–412. https://doi.org/10.1016/j.ridd.2003.07.003

Olçay Gül, S. (2016). The combined use of video modelling and social stories in teaching social skills for individuals with intellectual disability. *Educational Sciences: Theory and Practice, 16*, 83–107. https://doi.org/10.12738/estp.2016.1.0046

Panyan, M. V. (1998). *How to teach social skills*. Austin, TX: Pro-Ed.

Percy, M., Fung, W. L. A., Brown, I., & Hassiotis, A. (2017). Introduction to behavior and mental health. In M. L. Wehmeyer, I. Brown, M. Percy, K. A. Shogren, & W. L. A. Fung (Eds.), *A comprehensive guide to intellectual and developmental disabilities* (2nd ed., pp. 323–339). Baltimore, MD: Paul H. Brookes Publishing.

Provost, M. (2017). *Using bibliotherapy to support children's friendships with person with autism spectrum disorder* (Masters thesis, San Diego State University). Retrieved from https://search.proquest.com/docview/1970388414?pq-origsite=gscholar

Ratcliffe, B., Wong, M., Dossetor, D., & Hayes, S. (2015). The association between social skills and mental health in school-aged children with autism spectrum disorder, with and without intellectual disability. *Journal of Autism and Developmental Disorders, 45*, 2487–2496. https://doi.org/10.1007/s10803-015-2411-z

Reichle, J., & Wacker, D. P. (2017). *Functional communication training for problem behavior*. New York, NY: The Guilford Press.

Reichow, B., & Volkmar, F. (2009). Social skills interventions for individuals with autism: Evaluation for evidence-based practices within a best evidence synthesis framework. *Journal of Autism and Developmental Disorders, 40*, 149–166. https://doi.org/10.1007/s10803-009-0842-0

Reid, D. H. (1987). *Developing a research program in human service agencies: A practitioners guidebook*. Springfield, IL: Charles C. Thomas.

Remington, B. (1998). Applied behavior analysis and intellectual disability: A long-term relationship? *Journal of Intellectual and Developmental Disability, 23*, 121–135. https://doi.org/10.1080/13668259800033631

Reynhout, G., & Carter, M. (2007). Social Stories™ efficacy with a child with autism spectrum disorder and moderate intellectual disability. *Focus on Autism and Other Developmental Disabilities, 22*, 173–182. https://doi.org/10.1177/10883576070220030401

Reynhout, G., & Carter, M. (2008). A pilot study to determine the efficacy of a Social Story™ intervention for a child with autistic disorder, intellectual disability and limited language skills. *Australasian Journal of Special Education, 32*, 161–175. https://doi.org/10.1017/s1030011200025823

Schneider, N., Goldstein, H., & Parker, R. (2008). Social skills interventions for children with autism: A meta-analytic application of percentage of all non-overlapping data (PAND). *Evidence-based Communication Assessment and Intervention, 2*, 152–162. https://doi.org/10.1080/17489530802505396

Shure, M. B. (2001a). *I can problem solve: An interpersonal cognitive problem-solving program* (2nd ed.). Champaign, IL: Research Press.

Shure, M. B. (2001b). I can problem solve (ICPS): An interpersonal cognitive problem-solving program for children. *Residential Treatment for Children and Youth, 18*, 3–14. https://doi.org/10.1300/J007v18n03_02

Shure, M. B., & Spivack, G. (1982). Interpersonal problem solving in young children: A cognitive approach to prevention. *American Journal of Community Psychology, 10*, 341–356.

Sigafoos, J., Arthur, M., & O'Reilly, M. (2003). *Challenging behavior and developmental disability*. London, UK: Whurr.

Singh, N. N. (Ed.). (2016). *Handbook of evidence-based practices in intellectual and developmental disabilities*. Cham, Switzerland: Springer.

Skinner, B. F. (1953). *Science and human behavior*. New York, NY: Macmillan.

Solish, A., Perry, A., & Minnes, P. (2010). Participation of children with and without disabilities in social, recreational and leisure activities. *Journal of Applied Research in Intellectual Disabilities, 23*, 226–236. https://doi.org/10.1111/j.1468-3148.2009.00525-x

Stauch, T. A., Plavnick, J. B., Sankar, S., & Gallagher, A. C. (2018). Teaching social perceptions skills to adolescents with autism and intellectual disabilities using video-based group instruction. *Journal of Applied Behavior Analysis, 51*, 647–666. https://doi.org/10.1002/jaba.473

Stokes, T. F., & Baer, D. M. (1977). An implicit technology of generalization. *Journal of Applied Behavior Analysis, 10*, 349–367. https://doi.org/10.1901/jaba.1977.10.349

Sturmey, P. (2014). Adaptive behavior. In P. Sturmey & R. Didden (Eds.), *Evidence-based practice and intellectual disabilities* (pp. 29–61). Chichester, UK: Wiley Blackwell.

Sturmey, P., & Didden, R. (Eds.). (2014). *Evidence-based practice and intellectual disabilities*. Chichester, UK: Wiley Blackwell.

Sturmey, P., & Hersen, M. (Eds.). (2012). *Handbook of evidence-based practice in clinical psychology, Vol. 1: Child and adolescent disorders*. Hoboken, NJ: John Wiley & Sons.

Sukhodolsky, D. G., & Butter, E. M. (2007). Social skills training for children with intellectual disabilities. In J. W. Jacobson, J. A. Mulick, & J. Rojahn (Eds.), *Issues in clinical child psychology. Handbook of intellectual and developmental disabilities* (pp. 601–618). New York, NY: Springer. https://doi.org/10.1007/0-387-32931-5_30

Sullivan, A. K., & Strang, H. R. (2002/2003). Bibliotherapy in the classroom: Using literature to promote the development of emotional intelligence. *Childhood Education, 79*, 74–80. https://doi.org/10.1080/00094056.2003.10522773

Sundel, M., & Sundel, S. S. (2018). *Behavior change in human services: Behavioral and cognitive principles and applications* (6th ed.). Thousand Oaks, CA: Sage.

Taft, R. J., Hotchkiss, J. L., & Lee, D. (2016). Efficacy of music therapy and bibliotherapy as interventions in the treatment of children with EBD: A literature review. *International Journal of Learning, Teaching and Educational Research, 15*, 113–129. Retrieved from: http://www.ijlter.org/index.php/ijlter/article/view/777/pdf

Totsika, V., Felce, D., Kerr, M., & Hastings, R. P. (2010). Behavior problems, psychiatric symptoms and quality of life for older adults with intellectual disability with and without autism. *Journal of Autism and Developmental Disorders, 40*, 1171–1178. https://doi.org/10.1007/s10803-010-0975-1

Underwood, L., McCarthy, J., & Tsakanikos, E. (2010). Mental health of adults with autism spectrum disorders and intellectual disability. *Current Opinion in Psychiatry, 23*, 421–426. https://doi.org/10.1097/YCO.0b013e32833cfc18

Vereenooghe, L., & Langdon, P. E. (2013). Psychological therapies for people with intellectual disabilities: A systematic review and meta-analysis. *Research in Developmental Disabilities, 34*, 4085–4102. https://doi.org/10.1016/j.ridd.2013.08.030

Visser Knoth, M. (2006). What ails bibliotherapy? *The Horn Book Magazine, 82*, 273–276. Retrieved from: https://www.hbook.com/2006/05/using-books/what-ails-bibliotherapy/

Walton, K. M., & Ingersoll, B. R. (2013). Improving social skills in adolescents and adults with autism and severe to profound intellectual disability: A review of the literature. *Journal of Autism and Developmental Disorders, 43*, 594–615. https://doi.org/10.1007/s10803-012-1601-1

Wang, S., Parilla, R., & Cui, Y. (2013). Meta-analysis of social skills interventions of single-case research for individuals with autism spectrum disorders: Results from three-level HLM. *Journal of Autism and Developmental Disorders, 43*, 1701–1716. https://doi.org/10.1007/s10803-012-1726-2

Watkins, L., Kuhn, M., O'Reilly, M. F., Lang, R., Sigafoos, J., & Lancioni, G. E. (2016). Social skills. In N. N. Singh (Ed.), *Handbook of evidence-based practices in intellectual and developmental disabilities* (pp. 493–509). Cham, Switzerland: Springer.

Weiss, M. J. (2013). Behavior analytic interventions for developing social skills in individuals with autism. In P. F. Gerhardt & D. Crimmins (Eds.), *Social skills and adaptive behavior in learners with autism spectrum disorders* (pp. 33–51). Baltimore, MD: Paul H. Brookes Publishing.

Willner, P., Rose, J., Jahoda, A., Stenfert Kroese, B., Felce, D., Cohen, D., … Hood, K. (2013). Group-based cognitive-behavioural anger management for people with mild to moderate intellectual disabilities: Cluster randomized controlled trial. *British Journal of Psychiatry, 203*, 288–296. https://doi.org/10.1192/bjp.bp.112.124529

Index

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B

C

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